PRE- OR POST- SCHOOL INFLUENCES ON LEARNING ADAPTATIONS, RISKS AND DISABILITIES IN CHILDREN AND ADOLESCENTS: OVERLAPPING CHALLENGES FOR PUBLIC HEALTH, EDUCATION AND DEVELOPMENT

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Editorial: Pre- or Post- School Influences on Learning Adaptations, Risks and Disabilities in Children and Adolescents: Overlapping Challenges for Public Health, Education and Development

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Editorial on the Research Topic

Pre- or Post- School Influences on Learning Adaptations, Risks and Disabilities in Children and Adolescents: Overlapping Challenges for Public Health, Education and Development.

The study of child and adolescent learning has generally focused on aspects specifically tied to individual academic performance. However, a new emerging perspective is that any "deficit" and/or disability and conversely any achievement is not the result of a single event, such as an isolated reaction, but it is formed, through numerous biosocial contributing variables, during a child's attempt to adapt to learning conditions and settings. The fit between such adaptations and normative criteria (set by educational and social standards) is often associated with labels such as "fulfillment," "strengths" "resilience" or "weaknesses," "risk," "vulnerability" and "disability."

This Research Topic explored the overlapping challenges and themes related to developmental adaptations (as defined above) in the context of formal and informal settings for learning primarily within childhood and adolescence.

To start off in this path, Jefferies et al. investigated the relationships between the multidimensional constructs of physical literacy and resilience in 227 school Canadian children aged 9–12 years old. They found that resilience was predicted by movement capacity, confidence, and competence, environmental engagement, and overall perceptions of physical literacy. Their research highlights the importance of introducing physical literacy in schools.

In a similar vein, Ato et al. examined the impact of temperament on academic achievement and sociometric status in a sample of 295 6–7-year-old Spanish children. Parents completed the Temperament in Middle Childhood Questionnaire, while sociometric status and academic achievement were derived from teachers' reports. Latent profile analysis showed that Children in the "Negative/Undercontrolled" profile were at higher risk for academic failure and peer rejection, while "Sociable/High regulated" showed the reverse pattern. The findings have very important (so far underexplored) implications for ways in which schools could integrate "difficult children."

Two contributions focused on early developmental determinants. Firstly, Yao et al. examined the association of dopamine-related genes with mental and motor development and gene-environment interaction in 201 preterm and 111 term Taiwanese children, who were followed from 6 to 36 months and were genotyped for 15 single-nucleotide polymorphisms (SNPs) in dopamine-related genes (DRD2, DRD3, DAT1, COMT, and MAOA). MAOA SNPs were robustly associated with the mental (but not motor) development scores throughout early childhood in the premature children but not in the term counterparts. This warrants further investigations on whether the MAOA variants could help develop personalized interventions for preterm children.

Considering another early developmental period, Li et al. examined whether *executive function training* (EFT) could improve children's emotional competence (EC). Fifty-five 4-year-old Chinese children were assigned to either EFT or no-EFT group. Pre-test vs. post-test training between-and- within-subjects effects were analyzed to quantify improvement. EFT was associated with significantly higher scores on EC and changes in inhibition control and working memory abilities significantly predicted variation in EC. The findings suggest that intervening on inhibition control and working memory abilities via training may improve preschool children's emotional abilities.

At the other end of the spectrum, adult studies have shown that the concurrency of a smartphone-related task and walking can increase instability and risk of injuries. Caramia et al. recruited 29 young Italian adolescents to test whether walking with a smartphone increased fall and injuries risk, and to quantify these possible outcomes, participants were asked to walk along a walkway, with and without a concurrent writing task on a smartphone. Concurrency of walking and smartphone use resulted in reduced step length, gait speed and general aspects of gait stability, regardless of experience or frequency of use, suggesting that using the smartphone while walking may determine an increased risk of injury or falls also for young digital natives.

Adopting an approach to prevent air pollution, Zhao et al. developed the Brief Haze Weather Health Protection Behavior Assessment Scale-Adolescent Version (BHWHPBAS-AV), and tested its validity and reliability in two randomly selected districts of Baoding, China, and involving 22 middle-school classrooms and 1,025 valid questionnaires. The BHWHPBAS-AV scale showed promising reliability and validity suggesting it may be applied to assess adolescent haze weather health protection behavior, and help school and medical staff administer targeted behavioral and preventative interventions or health education programs.

In a second prevention study, Grant et al. assessed the cognitive profiles of 360 Canadian adults and children ranging from 7 to 80 years of age with disability in reading alone, mathematics alone and both (comorbidity), with tests widely used in both psychoeducational and neuropsychological batteries. Through a systematic exhaustive review of clinical neuroimaging literature, they mapped the complex set of domain-specific and domain-general impairments shown in the comorbidity of reading and mathematical disabilities to correspondingly plausible neuroanatomical substrates of dyslexia and dyscalculia. According to their hypothetical model, reading-math comorbidity seems due to atypical development of the left angular gyrus. This neuroeducational framework may assist to improve both early prediction and intervention across developmental periods.

Finally, Malboeuf-Hurtubise et al. reported a pilot study based on a new intervention, which combines mindfulness meditation and Philosophy for Children (P4C) activities, with the goal of improving mental health in pre-kindergarten children. Thirty-eight pre-kindergarten Canadian children took part in this study and were randomly allocated to the experimental or wait-list control conditions. Teachers completed pre- and post-intervention questionnaires. Although there were no significant effects, some improvement trends were found for internalized symptoms and hyperactivity. The results partly contradict previous research and suggest that mindfulness and P4C may not be effective intervention for mental health in children. However, the study also suggests a host of other confounding variables that might be responsible for the null findings and should be addressed in future research.

In summary, this Research Topic collection explored how children's preferences, profiles and predispositions are shaped by the social and biological activities that form the background of their everyday living. These papers addressed integrated multidisciplinary issues of education, development and public health, contributing practical examples of viable and sustainable local targeted programs, which, hopefully, may stimulate future research.

AUTHOR CONTRIBUTIONS

AD'A wrote the first draft. All authors contributed equally to edit to final version.

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Physical Literacy and Resilience in Children and Youth

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Background: There is growing interest in the relationship between physical and psychosocial factors related to resilience to better understand the antecedents of health and successful adaptation to challenges in and out of school, and across the lifespan. To further this understanding, a trans-disciplinary approach was used to investigate the association between the multidimensional constructs of physical literacy and resilience in children at a key stage in their development.

Methods: Cross-sectional data were collected from 227 school children aged 9-12 years old from five schools in Winnipeg, Manitoba, Canada. Resilience was measured using the *Child and Youth Resilience Measure*, and physical literacy through the *Physical Literacy Assessment for Youth* tools. Data were provided by self-report, surrogate assessors of the child (physical education teachers and parents), and trained assessors for movement skills. These data were analyzed using correlation and logistic regression.

Results: Resilience was significantly correlated with numerous indicators of physical literacy, including movement capacity, confidence, and competence, environmental engagement, and overall perceptions of physical literacy. Regressions indicated that resilience could be predicted by movement confidence and competence, environmental engagement, and overall physical literacy.

Conclusions: The findings of this study, using a constellation of sources, provide foundational evidence for the link between resilience and physical literacy among children, encouraging the importance of physical literacy development in schools. Longitudinal studies are required to further examine this relationship and how these previously unrelated fields may work together for a richer understanding of the interplay between the physical and psychological determinants of well-being.

Keywords: resilience, physical literacy, physical activity, physical education, children, youth, circus

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INTRODUCTION

A multisystemic and social-ecological understanding of resilience asserts that young peoples' capacity to thrive despite exposure to adversity depends on the quality of their interactions with aspects of their environment, and the degree to which those environments provide the resources for the development or maintenance of optimal psychological, social, and physical well-being (1–3). This approach to resilience encourages the importance of the availability and accessibility of resources that can foster resilience, and that strengths in one domain may buffer against stressors related to those in another (2). For instance, studies have shown that school-based interventions designed to facilitate improvements in cognitive and interpersonal skills not only lead to better academic performance, but can also provide protective effects against social and health risks such as delinquency and psychosocial distress (4).

This has particular implications for approaches to create healthy schools, such as the Pan-Canadian Healthy School Planner (5) and the UK Resilience Programme (6), which adopts the Penn resilience programme used in the US (7). These approaches promote resilience-bolstering practices that focus on building *psychosocial* resources (e.g., self-awareness, self-management), but at present, neglect or demote the *physical* to physical education classes (or Daily Physical Activity, intramural physical activity programming, or recess), despite known links between physical and psychological domains [e.g., Deuster and Silverman (8)].

In a parallel field, physical literacy has been defined as the competence to perform movement skills and the knowledge, motivation, confidence, and understanding to value and take responsibility for engagement in physical activity across the lifespan (9, 10). It has also been described as the physical and psychological attributes that are foundational to participation in physical activity and therefore the capacity for an active lifestyle (11, 12). As the groundwork for physical activity, physical literacy is said to be the basis for sustaining the health of individuals (13, 14), and therefore maintaining a healthy workforce and reducing the burden on health systems (15, 16). This has led to suggestions that cultivating physical literacy is as essential as developing skills in literacy and numeracy (17, 18), and has prompted global interest, such as the UNESCO program focus (19) and part of the WHO's proposal to create "active societies" (20). In Canada and the United States, recognition of the importance of physical literacy and the limitations of traditional sports-based physical education have led to a shift to a holistic physical literacy enriched curricula, which nurtures skill, confidence, motivation, and participation (21–24).

As physical literacy includes affective, social, and cognitive elements (9, 25), there are therefore commonalities with resilience. Furthermore, the core elements of both physical literacy and resilience develop when an environment is established which helps foster the ability to overcome challenges, obstacles, or adversity in physical and social settings (2, 3, 26). In the realm of resilience, this process of "steeling" suggests that limited exposure to adversity in appropriate environments can help an individual gain experience and coping strategies that can provide advantages in future encounters (27, 28), and has been linked to agency and persistence (29). Similarly, in physical literacy, the process of engaging in appropriately constructed challenges leads not only to improved basic movement competence, but concurrently the confidence and general competence to execute and assess movement in varied physical and social contexts, thus leading to the motivation for further physical activity engagement [see **Figure 1**, and for emerging evidence see Kriellaars et al. (30)]. The common process of engaging and overcoming challenges could be the basis for the association between the two constructs.

An investigation employing physical literacy could reveal whether the important foundational attributes for physical activity link to the established psychological and social factors related to resilience. Such a link would have implications for expanding school resilience programs to better support the well-being of students and trajectories for healthy development. It would also support the development of cross and cocurricular linkages and elevate the importance of physical education, as well as other movement-based school interventions. The objective of this study, then, was to investigate the association between physical literacy and resilience in children at a formative stage of their development. We hypothesized that measures of physical literacy would be significantly positively correlated with, and predictive of, resilience.

FIGURE 1 | The cycle of physical literacy. Adapted from: Cairney et al. (14).

METHOD

Design

Baseline, cross-sectional data was extracted from a 3-year longitudinal project exploring the introduction of circus arts in the physical education (PE) curriculum in Canadian schools. This project was a partnership between the National Circus School, the Resilience Research Centre of Dalhousie University, Centre de liaison sur l'intervention et la prévention psychosociale (CLIPP), the Quebec federation of school boards (FCSQ), Concordia University, University of Manitoba, and the University of Alberta. Data were supplied by participants, as well as their PE teachers, their parents, and trained assessors, in order to gain insights from a range of informative and credible sources.

Participants

Two hundred and twenty-seven elementary school children participated in the study (99 males, 122 females, 6 did not disclose) from five low-to-moderate socioeconomic status schools in Winnipeg, Manitoba. Students were in grades 4–6 with ages ranging from 9 to 12 years old ($M = 10.04$, $SD = 0.47$). Child assent and parental informed consent were obtained. Ethical approval for the study was granted by the University of Manitoba.

Measures

Six separate assessments were included in the study. Five were from the Physical Literacy for Youth Assessment (PLAY) tools

(31), designed to measure aspects of physical literacy from a variety of sources. Instructions for administration and further information can be found at physicalliteracy.ca/resources/.

The first assessment was the "PLAYfun," which is completed by a trained assessor and measures an individual's competence of performing 18 curricular-linked movement skills (including running, locomotor movements, object control, balance, and stability), graded on a four-category 100 mm visual analog scale with 25 mm categories ranging from "initial," "emerging," "competent," to "proficient" ($\alpha = 0.87$) (32, 33). We used the average of the 18 skills as a measure of an individual's *movement competence*, and the sum as a measure of their *movement capacity*. The second tool was the "PLAYself" (7-day test-retest reliability = 0.83) (Jefferies et al., in preparation), which is completed by the child and includes a 12-item *physical literacy self-description*, rated on a four-point Likert scale (sample items include "I think I have enough skills to participate in all the sports and activities I want" and "I'm confident when performing activities"). It also includes a 6-item measure of *environmental engagement* (activity in and on water, on ice, on snow, and indoors and outdoors generally).

The third tool was the self-report "PLAYinventory" form (7-day test-retest reliability = 0.76), providing a count of the *number of activities* regularly participated in during the individual's leisure time over the last 12 months (out of over 80 possible activities). The fourth tool was the "PLAYparent" (7-day test-retest reliability = 0.73), completed by a parent to measure perceptions of their child's *movement competence* (6 items scored on a 3-point Likert scale) and their *overall physical literacy* (via a two-category 100 mm visual analog scale). The fifth tool was the "PLAYpe_teacher" (7-day test-retest reliability = 0.87), a modified version of "PLAYcoach," providing measures of *movement competence* (6 items) and *confidence* (single item measure), both measured on a 5-point Likert scale ranging from "poor" to "excellent," as well as the physical education teacher's rating of the student's *overall physical literacy* (via a two-category 100 mm visual analog scale). The tool also captures an individual's *overall fitness* and *activity level* (single item measures), measured on a 5-point Likert scale ranging from "poor" to "excellent," in addition to their *body composition*—a "visual BMI"—involving a 10-picture body silhouette arranged in order of ascending BMI, which an individual is matched to (34, 35). These three fitness variables (measures associated with domain of physical activity) were included to investigate their possible relationship to resilience and to compare to the potential relationship between resilience and the physical literacy variables.

A novel sixth tool, also self-report, assessed resilience and peer relations, using the 12-item Child and Youth Resilience Measure (CYRM; $\alpha = 0.75$) (36, 37) and the peer relations subscale of the Strengths and Difficulties Questionnaire ($\alpha = 0.60$) (38). The CYRM is used to provide indications of an individual's social-ecological resilience, meaning that scores reflect the ability of individuals to engage with external resources in order to manage adversity. For ease of interpretation, the peer relations subscale was reverse-coded so that higher scores indicate better relations with peers.

Data Collection

Data collection took place in October 2016 over a 2-week period, where the participants completed the PLAYself, PLAY Inventory, and the questionnaire containing the CYRM and peer relations scale. Eight trained assessors rated the participants on the PLAYfun using a similar approach to that of Kriellaars and colleagues (30), and PE teachers completed a PLAYpe_teacher assessment for each child based on their recall of an individual after 1 month of exposure in the school setting. Parents completed a PLAYparent assessment for their own children.

Analyses

One-way ANOVAs were performed to explore differences between male and female participants, as these are often reported in the literature [e.g., O'Brien et al. (39)]. Correlational analyses were then performed to examine relationships between measures of physical literacy, resilience, and peer relations. Exploration of resilience scores suggested a threshold score of 29 was appropriate to dichotomise individuals into high ($n = 139$) and low ($n = 53$) categories, which corresponds with thresholds recommended for other versions of the measure (40). Given the interest in the identification and intervention of youth most in need of support, binary logistic regressions were employed to examine the capacity of physical literacy and fitness variables to predict whether individuals had high or low levels of resilience (linear regression using resilience as a continuous variable returned similar results but without classification information). All analyses were performed using SPSS v21 (41).

RESULTS

We discovered significant sex differences in trained assessor ratings of movement competence ($p = 0.010$) and capacity ($p = 0.009$), PE teacher ratings of movement confidence ($p = 0.016$), and overall activity level ($p = 0.011$). We also found significant differences between the sexes in parent perceptions of their child's movement competence ($p = 0.022$). In each instance, male participants scored significantly higher than their female counterparts (**Table 1**). No significant differences were observed between males and females among the remaining variables.

As hypothesized, resilience scores were positively correlated with trained assessor ratings of movement competence ($r_s = 0.16$, $p = 0.031$) and capacity ($r_s = 0.15$, $p = 0.042$), as well as PE teacher ratings of competence ($r_s = 0.17$, $p = 0.016$), confidence ($r_s = 0.25$, $p = 0.001$), overall physical literacy ($r_s = 0.21$, $p = 0.004$), fitness ($r_s = 0.26$, $p < 0.001$), and activity levels ($r_s = 0.23$, $p = 0.001$). Resilience was also positively correlated with self-reported physical literacy ($r_s = 0.30$, $p < 0.001$) and environmental engagement ($r_s = 0.22$, $p = 0.001$). Parental ratings of movement competence and overall physical literacy, body composition, and number of self-reported activities were not found to be related to resilience.

Resilience scores also positively correlated with peer relations ($r_s = 0.31$, $p < 0.001$). Additionally, peer scores correlated with PE teacher assessments of confidence ($r_s = 0.15$, $p = 0.036$), movement competence ($r_s = 0.14$, $p = 0.050$), overall physical literacy ($r_s = 0.16$, $p = 0.033$), fitness

TABLE 1 | Descriptive statistics and zero-order correlation coefficients for all variables against resilience scores.

Source	Variable	Males (n = 99) Mean (*SD*)	Females (n = 122) Mean (*SD*)	Total sample Mean (*SD*)	Resilience r_s
Self-report	Resilience	30.38 (4.44)	30.21 (3.98)	30.30 (4.17)	–
Self-report	Peer relations	10.30 (1.95)	10.32 (1.92)	10.32 (1.93)	0.31**
Self-report	No. of activities	26.72 (13.15)	25.83 (11.09)	26.24 (11.99)	0.03
Self-report	PL: Environmental engagement	24.39 (3.40)	23.90 (3.30)	24.13 (3.33)	0.22**
Self-report	PL: PL self-description	33.94 (4.96)	33.44 (5.59)	33.66 (5.28)	0.30**
Trained assessor	PL: Movement competence	33.44 (7.17)	30.76 (7.39)	31.99 (7.39)	0.16*
Trained assessor	PL: Movement capacity	601.26 (129.34)	551.85 (133.86)	574.47 (133.79)	0.15*
Parent	PL: Overall rating	23.29 (4.03)	22.79 (3.06)	23.07 (3.56)	0.14
Parent	PL: Movement competence	16.37 (2.85)	15.31 (2.10)	15.85 (2.54)	0.08
PE teacher	PL: Movement confidence	18.52 (4.46)	16.96 (4.72)	4.19 (1.73)	0.25**
PE teacher	PL: Movement competence	24.74 (6.72)	23.77 (5.98)	24.25 (6.34)	0.17**
PE teacher	PL: Overall rating	4.27 (1.77)	4.10 (1.70)	17.69 (4.68)	0.21*
PE teacher	Fitness	3.47 (1.00)	3.20 (1.13)	3.33 (1.08)	0.26**
PE teacher	Activity level	3.60 (0.92)	3.22 (1.13)	3.40 (1.06)	0.23**
PE teacher	Body composition	2.83 (1.73)	2.95 (2.27)	2.90 (2.03)	−0.06

*$p \leq 0.05$, **$p \leq 0.001$. PL, physical literacy; PE, physical education.

(r_s= 0.27, $p < 0.001$) and activity levels (r_s= 0.18, $p = 0.015$), but unlike resilience scores, peer scores negatively correlated with bodily composition (r_s= −0.18, $p = 0.015$), indicating that higher BMIs were associated with lower peer scores. Additionally, unlike resilience, peer scores were not found to correlate with assessor ratings of movement competence or capacity, nor self-reported physical literacy or environmental engagement.

We then performed a binary logistic regression, and a backwards stepwise approach involving all physical literacy variables revealed a significant and parsimonious solution at the sixth step ($\chi^2_5 = 23.731$, $p < 0.001$). The model, accounting for 33% of the variance in resilience scores (Nagelkerke R^2), correctly predicted 79% of cases (1.2% more than steps 5 and 7) and involved PE teacher perceptions of movement confidence and competence, parent perceptions of movement competence, and self-reported environmental engagement and perceptions of physical literacy. Numerous models using physical literacy variables were achievable. However, when adding fitness, activity level, and body composition in a second block of predictors, the model was also significant ($\chi^2_6 = 24.101$, $p = 0.001$), but these variables did not add predictive value (79% of cases correctly predicted, 33% of variance accounted for). Finally, we performed the binary logistic regression using the fitness, activity, and body composition variables alone, and a significant model was not produced.

DISCUSSION

Both the correlative and predictive results of the study demonstrate a clear association between resilience and physical literacy in children, arising from a constellation of sources, involving self-perception, perception of a teacher, or assessment by trained observers. Specifically, we found that an individual's resilience is associated with their competence and confidence to move, which are the key components of physical literacy (9). Only parent perceptions did not correlate with self-reported levels of resilience, which may be explained by the tendency for parents to overestimate the abilities of their children (42, 43). Additionally, no sex differences were detected in self-reports of resilience, but were found in physical literacy variables such as movement confidence, which were significantly higher in male participants compared to females, fitting with the literature (39, 44, 45).

To our knowledge, this is the first study to demonstrate the connection between resilience and physical literacy. Although the mechanism which links resilience and physical literacy remains unclear, it presents the beginning of an important transdisciplinary approach (46, 47). For instance, if the affective domains of confidence and motivation developed in physical literacy (25) go beyond just motor action, then they may also provide or help young people acquire the skills and abilities to better negotiate for, and navigate to, resources that sustain their well-being in different contexts. This is fundamental to resilience (48). Furthermore, the contexts in which movement competence develops are both physical (land, air, ice, snow, water) and sociocultural, and require that these competencies are developed with exposure to problem-solving settings in both (26). This exposure to and mastery of 'positive challenge' may position physical literacy as an antecedent of resilience. Our study is cross-sectional and therefore prohibits establishing causal mechanisms; however, either direction of action is beneficial, as improvements in resilience may lead to greater confidence and motivation in physical activity, and therefore to greater participation, health, and well-being (8). A longitudinal study is required to examine the causal couplings between the constructs.

These findings respond to the emerging interest in how physical literacy relates to health (20) and the link to mental, physical and social well-being. Our study supports the notion that encouraging physical literacy is associated with fostering resilience, and so creating the underlying conditions for individuals to thrive and participate actively in society. This has important implications for curricula development in schools. For instance, by including physical literacy development, and so acknowledging the importance of physical as well as psychosocial factors associated with resilience, such a holistic reframing broadens access to the resources for young people to thrive. Furthermore, although there have been resilience-related investigations into the protective factors and stressors involved in sport (49, 50), the study of physical literacy is broader and likely more relevant to vulnerable persons that may not be engaged in traditional sports activities; physical literacy includes movement in vocation, recreation, performance arts, and school, as well as sport (51). Therefore, the inclusion of robust physical literacy curricula in schools would likely lead to a greater participatory culture and arguably to greater human and social equity [see the "power pillar" of physical literacy (26)]. Indeed, some health and physical education curricula have already adopted a physical literacy approach (22, 24), and initial implementations have been highly successful (30).

Other findings in this study point to interesting areas of further research. For example, while peer relations were not associated with most sources of physical literacy, we found that they were associated with PE teacher ratings of elevated BMI and other physical literacy-related variables. This may indicate a separation of the factors implicated in peer relationships and resilience. In a social-ecological model of resilience which emphasizes the combined impact of psychological, social, cultural, and physical resources that sustain well-being (2, 48), peer relationships may be only one dimension of resilience as it links to physical literacy. However, as both peer relationships and body composition are known to impact resilience (48, 52), it is also possible that some resilience and physical literacy variables may begin to interact at a later age. This is encouraged by our findings reflecting the literature that fitness, activity level, and body composition were associated with each other (53) and with physical activity (54), but that they did not add to the predictive capacity of our model. Perhaps at 9–12 years old (the age of our participants) these factors may not yet have an impact on resilience, but as children move into older adolescence, levels of fitness, activity, body composition, and peer relationships become more important. Process models of physical literacy (14, 55) propose a positive feedback cycle of motivation and confidence which leads to competence (and then participation); a process which may begin pre-adolescence and become more strongly associated with resilience in adolescence [see the "journey pillar" of physical literacy (26)]. As this was an exploratory cross-sectional study, we cannot confirm these relationships, however, the study stimulates thinking about the conceptualization of physical literacy and resilience as processes that are likely interactive.

As this study is part of a longitudinal project, we will be able to track variations in resilience and physical literacy to shed light on how changes in each interact from pre-adolescence into adolescence. Such a study would be particularly valuable given the precipitous drop in physical activity that occurs around age 12 (56), and potentially inform whether the causal factors are pre-pubescent or related to the pubescent transition itself. This line of further investigation may also explain why no sex difference was found for self-reported resilience in the current study, but female participants scored more poorly than males on a number of physical literacy indicators. As physical activity levels decrease more steadily for females than for males into adolescence, investigating corresponding changes in resilience could also help to address the challenge related to sex-related differences in resilience that have been identified at this age (57–59). This may then point to critical ages for schools to invest in physical literacy development to reduce the gap in movement competence and confidence (30).

CONCLUSION

This study directly links resilience and physical literacy, bringing together two previously distinct approaches to the well-being of young people and confirming their relationship. Our findings support the value of physical literacy development in schools as part of a holistic approach toward supporting the well-being of young people and their future health. Further longitudinal research into the interaction of resilience and physical literacy processes could provide insight into creating optimal physical-psycho-social environments in schools that foster the development of both constructs, and the possible interlinking to other key processes, such as creativity.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this manuscript will be made available by the authors, without undue reservation, to any qualified researcher.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by University of Manitoba Research Ethics Board. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

MU, PA, and DK conceived of the project and contributed to the revising and editing of the manuscript. DK and PJ aided in collecting and analyzing the data, and to the writing and revising of the manuscript. All authors reviewed and approved the manuscript.

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Relation Between Temperament and School Adjustment in Spanish Children: A Person-Centered Approach

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The aim of this study was to examine from a person-centered approach the impact of temperament on academic achievement and sociometric status in a sample of 6–7-year-old Spanish children. To measure children's temperament in early childhood, parents were given TMCQ (Temperament in Middle Childhood Questionnaire), while sociometric status and academic achievement were requested for children's teachers. Using latent profile analysis (LPA) four temperament profiles were found. Children belonged to the "Negative/Undercontrolled" profile showed a higher probability of academic failure and were more rejected, and children included in the profile "Sociable/High regulated" showed higher academic scores and a lower probability of being rejected by their peers. Several implications in the Spanish educational context are discussed.

Keywords: temperament, academic achievement, social adjustment, person-centered approach, latent profile analyses

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INTRODUCTION

Academic failure and social maladaptation constitute distressful and ongoing problems in Spain. Considered by recent Ministers of Education as the main problem in Spanish education, school dropout has increased alarmingly in recent years, and is the second highest in the EU (Ministry of Education and training, 2019). Indeed, the latest available data from the PISA report (Programme for International Student Assessment) highlight Spain's poor performance, with scores below average in both Language and Mathematics (INEE, 2015), compared with other countries. Furthermore, disorders related to social maladjustment in schools, like bullying, anxiety and depression, have also been increased considerably. Concretely, bullying scaled up almost 50% from 2015 to 2017, exceeding for the first time 1000 victims annually in Spain (Report Bullying sin fronteras, 2018). In the search for the most important contributors to explain academic and social outcomes, children's temperament has emerged strongly, based on the idea that children's individual differences in emotional arousal could be a protective or a risk factor in their developmental trajectories (Leve et al., 2005).

Temperament has been defined as individual differences in reactivity and self-regulation influenced by maturity and experience (Rothbart, 1981), and its correlation with academic and social adjustment are well established. However, not all the dimensions of temperament contribute equally in explaining children's adjustment. Specifically, high levels of negative emotionality and low levels of effortful control, defined as the ability to suppress a dominant response to perform a subdominant response (Rothbart and Bates, 1998), have been significantly related to poor academic achievement (Blair and Razza, 2007; Valiente et al., 2008; Zhou et al., 2010; Hintsanen et al., 2012) and worse social adjustment (Eisenberg et al., 2000), while the contribution of positive emotionality is not so clear. Nevertheless, although these relations have been systematically reported in the literature, there are few recent studies relating these variables in Spain (Checa and Rueda, 2011; Ato et al., 2014; Checa and Abundis-Gutierrez, 2017; Galián et al., 2018). Thus, and similar to the results found in other cultures, self-regulation abilities have been proved to have a linear or indirect effect on academic achievement or social maladjustment. Particularly, individual differences in effortful control predicted academic achievement (Checa and Rueda, 2011; Checa and Abundis-Gutierrez, 2017; Galián et al., 2018), and sociometric status in Spanish children (Ato et al., 2014; Sánchez-Pérez et al., 2015) so less self-regulated children were those with lower academic performance and a higher probability of being rejected.

Nevertheless, these relations have been examined mainly thorough a variable-centered approach, which treats dimensions (or overarching factors) as independent entities that uniquely predict their relations with other outcomes. This approach, although useful, is incomplete as it tends to separate psychological processes from the individual in whom they occur and ignores the organization of multiple traits within an individual (Hart et al., 2003). To the extent that this approach lacks of a multilevel consideration of the problem, it seems to be inadequate when the objective is to establish conclusions about individuals (Crockett et al., 2006). In contrast, the person-centered approach is holistic, and the individual is viewed as the unit of analysis and each trait takes on meaning based upon its role within the entire organization of the individual (Bergman and Magnusson, 1997). Thus, from this perspective it is possible to examine how unique combinations of temperament dimensions act together in predicting child outcomes.

In the temperament field, Thomas, Chess and Birch's pioneer work (1968), based on a person-centered approach, distinguished three children temperament profiles; difficult, easy and slow to warm up, related to socioemotional adjustment, with the difficult temperament outweighing compared to the other two for greater risk of maladjustment (Giancola et al., 1998; Moffitt et al., 2001; Frick and Morris, 2004). In this vein, McClowry (2002) identified four temperament profiles, of which "Social/Eager to try" and "Industrious" would be similar to the Easy profile, "High Maintenance" would be similar to the Difficult profile, and "Cautious/slow to warm up" would be similar to the Slow to warm up profile. Another classic study on profiles is Caspi and Silva (1995), who identified five temperament profiles and examined them in a longitudinal study from 3 to 18 years old. The five categories - undercontrolled, inhibited, confident, reserved and well-adjusted - showed striking differences in adjustment scores, with the undercontrolled being those who showed the worst adjustment in adulthood. More recently, Sanson et al. (2009) identified four temperament profiles which they called reactive/inhibited, poor attention regulation, non-reactive/outgoing and high attention regulation. In their work, children assigned to the reactive/inhibited and poor attention regulation groups tended to have higher levels of later behavior problems compared to children assigned to non-reactive/outgoing and high attention regulation categories, confirming that belonging to one or another profile has a strong impact on their present and future adjustment. Nevertheless, to date there are few studies exploring the effect of temperament on academic achievement and sociometric status from a person-centered perspective, and we have found no such studies for Spain that incorporate both academic and social outcomes.

Based on the aforementioned, the aims in this study were to determine, using Latent Profile Analysis (LPA), temperament profiles in Spanish children aged 6 and 7 years, and to analyze their impact on Math and Language achievement, on the one hand, and to Acceptance and Rejection scores, on the other. Taking into account the luck of studies in the field, this study hopefully will contribute to a better understanding of the variables involved in academic failure and social rejection, which is an important issue that needs to be addressed, particularly in Spain. Besides, we consider the start of childhood a crucial period in the analysis of these relations, insofar as this is the developmental stage in which we usually observe first academic and social problems in the school.

MATERIALS AND METHODS

Participants

This research used data from a larger study of child temperament and its relations with their adjustment in several areas. The participating families were recruited from five schools in Murcia (Spain). Although the larger sample of our research included 474 children from the First and Second Cycle of Primary School, for the purposes of our study we selected only First Cycle of Primary School children. The sample comprised 295 Spanish children (51.2% boys and 48.8% girls) of 6 (44.4%), and 7 (55.6%) years old. The most important reason behind this choice was the consideration that change from first to second cycle could imply academic variations which could alter the relations between temperament profiles and academic performance. Also, as we have just mentioned, we were particularly interested in studying these relations in the start of childhood. Of the participating families, 9.3% of the parents had completed Primary School studies, 22.1% had completed Secondary School, 29.3% held a professional qualification, 28.3% were university graduates, and 1% held a Ph.D.

Measures

Temperament

Temperament was measured using the Temperament in Middle Childhood Questionnaire (TMCQ; Simonds and Rothbart, 2004). This questionnaire obtains information provided by parents on a number of daily situations and includes 160 items on a 5-point Likert scale grouped in 17 temperament scales: (T1) *Activation Control*: The capacity to perform an action when there is a strong tendency to avoid it; (T2) *Activity level*: The level of gross motor activity including the rate and extent of locomotion; (T3)*Affiliation:* The desire for warmth and closeness with others, independent of shyness or extraversion; (T4) *Anger/frustration*: The amount of negative effect related to interruption of ongoing tasks or goal blocking; (T5) *Assertiveness/dominance*: The tendency to speak without hesitation and to gain and maintain control of social situations; (T6) *Attentional Focusing*: The tendency to maintain attentional focus upon task-related channels; (T7) *Discomfort:* The amount of negative effect related to sensory qualities of stimulation, including the intensity, rate or complexity of light, movement, sound and texture; (T8) *Fantasy/Openness:* Active imagination, esthetic sensitivity and intellectual curiosity; (T9) *Fear:* The amount of negative affect including unease, worry or nervousness related to anticipated pain or distress and/or potentially threatening situations; (T10) *High Intensity Pleasure:* The amount of pleasure or enjoyment related to situations involving high stimulus intensity, rate, complexity, novelty and incongruity; (T11) *Impulsivity*: The speed of response initiation; (T12) *Inhibitory Control:* The capacity to plan and to suppress inappropriate approach responses under instructions or in novel or uncertain situations; (T13) *Low Intensity Pleasure:* The amount of pleasure or enjoyment related to situations involving low stimulus intensity, rate, complexity, novelty and incongruity; (T14) *Perceptual Sensitivity*: The amount of detection of slight, low intensity stimuli from the external environment; (T15) *Sadness*: The amount of negative affect and lowered mood and energy related to exposure to suffering, disappointment and object loss; (T16) *Shyness:* Slow of inhibited approach in situations involving novelty or uncertainty; and (T17) *Soothability/Falling Reactivity:* The rate of recovery from peak distress, excitement or general arousal. The coefficient alpha for these subscales ranged from 0.621 to 0.887.

Sociometric Status

Sociometric status data were collected using the BULL-S Questionnaire (Cerezo, 2000), a questionnaire designed to measure, among other aspects, the sociometric position of each person of the group. For that purpose, the teachers administered a sociogram in which the children answered, in order of preference, about three other children with whom they least and most liked (1) working, and (2) spending their free time (in the classroom context). After that, for each of the participating children a rejection score (RS) and an acceptance score (AS) was calculated following the procedure detailed in the reference manual (Cerezo, 2000, p. 28–31).

Academic Performance

At the end of the academic year the students' final grades in Language (LP) and Math (MP) were recorded. The scores provided by the teachers were distributed in 4 categories (Fail, Pass, Merit, and Distinction). We categorized the variable from Fail = 1 to Distinction = 4.

Procedure

A meeting was held with the head teachers of the schools in order to explain the purpose of the project to them. After consent was given by the school and parents, a second meeting was held with the tutors to instruct them in the administration of the sociometric test. At the same time, they were given the temperament questionnaire, along with a letter addressed to the parents with instructions for filling in it. A telephone number for queries was also provided. When the tests were filled in, a third meeting with tutors and parents was held to solve or correct possible mistakes detected in the questionnaires. Questionnaires that were less than 80% completed were discarded.

Data Analysis

Complete data were collected for all 295 cases of the sample. No missing data were found. We first used a descriptive analysis of all the variables measured and ANOVA tests for gender differences. Then we ran a Latent Profile Analysis (LPA) to determine children's temperament profiles with the *tidyLPA* (version 1.0.2) package of *R* platform (Rosenberg et al., 2018), interfaced with *MPlus* (version 8.1, Muthén and Muthén, 1998–2017) via *MPlusAutomationR* program (Hallquist and Wiley, 2018). LPA is a special case of the general mixture model in which latent profiles are identified based on patterns of observed indicator variables. Latent profiles differ from latent classes due to the continuous nature of measured variables (Harring and Hodis, 2016). In our study, LPA is used mainly to discern the optimal covariance matrix and the number of children's subsets (profiles) who share similar patterns of temperament attributes.

In order to help in the interpretation of profiles we also used Partial Least Squares (PLS) regression, a dimension reduction technique where predictor variables are mapped onto a smaller set of variables that maximally explain a response variable. The package *pls* of R platform (version 2.7, Mevik et al., 2019) was selected, instead of linear regression analysis, to avoid multicollinearity problems and to enable the set of the 17 temperament scales to be ranked based upon how strongly they influence on each of the 4 profiles obtained by LPA. Finally, we also used planned comparisons ANOVA to detect plausible profile differences and gender differences in social and academic adjustment measures.

RESULTS

Descriptive Statistics, Correlations and Gender Differences

Table 1 shows descriptive data and significant correlations between the variables of interest. ANOVA tests were applied to check for statistically significant differences in gender and age for each of the measures. Significant differences between boys and girls were found for T4: $F(1,264) = 3.84$, $p = 0.05$; T6: $F(1,264) = 23.17$, $p < 0.001$; T7: $F(1,264) = 6.71$, $p = 0.01$; T8: $F(1,264) = 25.60$, $p < 0.001$; T11: $F(1,264) = 6.57$, $p = 0.01$; T12: $F(1,264) = 13.96$, $p < 0.001$; T13: $F(1,264) = 6.61$, $p = 0.01$; T14: $F(1,264) = 7.81$, $p < 0.01$; Rejection: $F(1,264) = 14.20$, $p < 0.001$ and Language achievement: $F(1,264) = 8.05$, $p < 0.01$, with boys scoring higher in T4, T7, T11, and Rejection, whereas girls scored higher in T6, T8, T12, T13, T14, and Language achievement.

TABLE 1 | Descriptive data and significant correlations.

	T1	T2	T3	T4	T5	T6	T7	T8	T9	T10	T11	T12	T13	T14	T15	T16	T17	AS	RS	LP	MP
Mean	3.34	4.09	4.24	3.19	3.43	3.24	2.86	3.97	2.89	3.30	2.98	3.20	3.98	3.70	2.66	2.78	3.48	6.50	5.11	3.25	3.25
SD	0.50	0.61	0.43	0.72	0.54	0.96	0.61	0.54	0.61	0.64	0.67	0.57	0.50	0.60	0.59	0.96	0.59	5.16	7.29	0.77	0.80
AS						0.3***				−0.1*	−0.2***	0.2**	0.1**						−0.4***	0.3***	0.3***
RS	−0.1*	−0.1*		0.1*		−0.4***	−0.4***	−0.1*			0.3***	−0.2***	−0.2***				−0.1*	−0.4***		−0.4***	−0.3***
LP	0.1*					0.4***		0.1*		−0.1*	−0.2*	0.1*	0.1*					0.3***	−0.4***		0.8***
MP						0.4***					0.1*	0.1*						0.3***	−0.3***	0.8***	

T1, Activation control; T2, Activity level; T3, Affiliation; T4, Anger/Frustration; T5, Assertiveness/Dominance; T6, Attentional Focusing; T7, Discomfort; T8, Fantasy/Openness; T9, Fear; T10, High Intensity Pleasure; T11, Impulsivity; T12, Inhibitory Control; T13, Low Intensity Pleasure; T14, Perceptual Sensitivity; T15, Sadness; T16 Shyness; T17, Soothability/Falling Reactivity; AS, Acceptance Scoring; RS, Rejection Scoring; LP, Language Performance; MP, Math Performance. *$p < 0.05$; **$p < 0.01$; ***$p < 0.001$.

No significant differences between children of 6 and 7 years old were found in any of the social and academic performance temperament measures.

Latent Profile Analysis and Partial Least Squares Regression

The 17 temperament scales of TMCQ were then subjected to a latent profile analysis (LPA) in order to select the optimum number of profiles. LPA is a model-based clustering technique that provides a precise framework for choosing a relevant number of clusters/profiles with the most appropriate covariance matrix. A first look at the covariance matrix of all temperament measures showed variances in the range 0.25/0.92 and covariances in the range -0.32/ + 0.29. Then we focused our attention on models whose covariance structures show equal or varying variances and zero covariances. These structures correspond to models 1 and 2 of the *tidyLPA* R-package (Rosenberg et al., 2019) and were estimated using the most popular range between 2 and 5 profiles. **Table 2** shows the most relevant information criteria used to choose the best combination of covariance structure and the number of profiles (lower values are better). Classical Akaike Information Criterion (AIC) and Bayesian Information Criterion (BIC) give contradictory information (AIC for model 2 with 5 profiles; BIC for model 1 with 4 profiles). The adjusted sample size BIC (SABIC; Sclove, 1987) pointed again toward model 1 with 5 profiles, but with the most robust Integrated Completed Likelihood (ICL, Biernacki et al., 1998; Baudry, 2015), the best fit model would be a 4-profile model with a covariance structure assuming equal variances and zero covariances.

For the interpretation of profiles, we regressed all temperament scales on each one of the 4 profiles selected with LPA. We used cross-validation to find the optimal number of components. Finally, we extracted all the useful information and the output was formatted to obtain normalized regression coefficients (positive and negative) to be interpreted as percentages (so their absolute sum is 100) and the result was sorted. **Figure 1** summarizes this process and details the relative importance of the main temperament scales, identifying each profile.

Profile 1 represented 25.8% of the sample (48 boys and 28 girls) and was characterized by higher than average scores in T1, T2, T10, and T11, and well below the mean in T4, T7, T15, and T16. Thus, we labeled this profile as "Outgoing/Average

TABLE 2 | Selection of the covariance matrix and number of profiles.

Covariance Structure	Profiles	AIC	BIC	SABIC	ICL
Model 1: Equal	2	9090	8282	9117	9319
variances,	3	8886	9144	8922	9206
Zero covariances	4	8711	9035	8756	9108
	5	8595	8986	8649	9131
Model 2: Varying	2	9076	9330	9112	9368
variances,	3	8857	9240	8910	9280
Zero covariances	4	8661	9174	8732	9224
	5	8576	9218	8666	9269

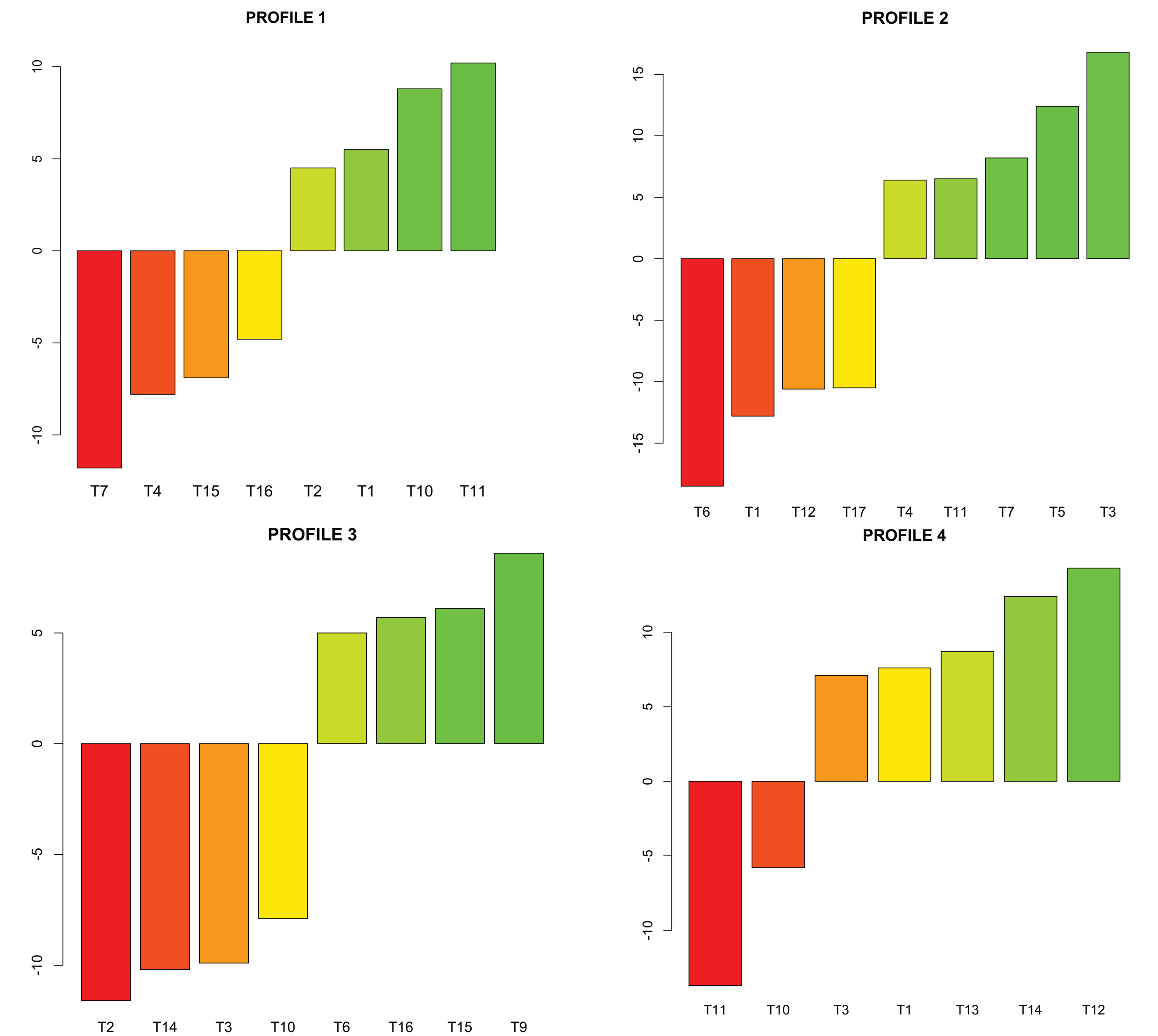

FIGURE 1 | Temperament profiles.

regulation." Profile 2 comprised 19.3% of the sample (33 boys and 24 girls) and was well above the mean in T4 and T11, higher than the mean in T5, T7, T9, T10, and T15, well below the mean in T6 and lower than the mean in T2, T12 and T17. This profile was then referred to as "Emotionally negative/Low regulation." Profile 3 represented 23.7% of the sample (36 boys and 34 girls) and was characterized by scores well above the mean in T9, higher than the mean in T6, T16, and T15 and well below the mean in T2, T3, T10, and T14. We called this profile "Inhibited/Average regulation." Finally, Profile 4 contained 31.2% of the sample (33 boys and 59 girls), and it was characterized by scores well above the mean in T12 and T14, higher than the mean in T3, T1, and T13, lower than the mean in T10 and well below the mean in T11. Thus, this profile was labeled as "Sociable/High regulation."

Profile differences in social and academic adjustment were carried out using planned comparisons ANOVA. **Table 3** shows the means of four measures by profiles and gender. Significant profiles effects were found in the explanation of Acceptance, Rejection, Language and Math achievement, in particular comparing profiles 2 and 4 with other profiles. Specifically, those belonging to Profile 2 have a higher probability of being rejected [$F(1,293) = 6.65$, $p = 0.01$] a lower probability of being popular [$F(1,293) = 6.51$, $p = 0.01$] and score lower in Language [$F(1,293) = 9.06$, $p = 0.003$], and Math achievement [$F(1,293) = 7.38$, $p = 0.007$] than children with other profiles. The inverse pattern was observed in Profile 4. Thus, children belonging to this profile scored significantly lower in Rejection scores: [$F(1,293) = 21.90$, $p < 0.001$], and

TABLE 3 | Means of main measures by profiles and gender.

Measures	Gender	Profile 1	Profile 2	Profile 3	Profile 4
Acceptance	Boys	5.250	4.152	6.611	8.212
	Girls	6.920	5.271	6.441	8.220
Rejection	Boys	7.875	9.394	5.417	3.424
	Girls	5.179	5.283	4.735	1.966
Language	Boys	3.042	2.818	3.222	3.485
	Girls	3.286	3.202	3.412	3.458
Mathematics	Boys	3.145	3.000	3.278	3.576
	Girls	3.143	3.125	3.412	3.322

significantly higher in Acceptance [$F(1,293) = 15.63$, $p < 0.001$], Language [$F(1,293) = 12.87$, $p < 0.001$] and Math performance [$F(1,293) = 7.28$, $p = 0.007$] than children with other profiles.

Finally, after comparing profiles 2 and 4 with gender variable added, we also found significant gender differences on Rejection: $F(1,292) = 8.52$, $p < 0.01$, with boys being more probably rejected, and Language: $F(1,292) = 4.65$, $p < 0.05$, with girls scoring higher in language performance.

DISCUSSION

Our study had a number of aims. First, we sought to determine temperament profiles in a Spanish sample of 6- and 7-year-old children, and second, we sought to explore how belonging to the different profiles affects children's academic and social adjustment. Regarding our first objective, we used LPA to identify 4 profiles of children differing in their temperament characteristics. Profile 1 (Outgoing/Average regulation) describes approaching and not very shy children. Children included in this profile, though considered by their parents as active and impulsive, scored as average in effortful control dimensions, such as activation control, inhibitory control and attentional focusing; profile 2 (Emotionally negative/Very low regulation) includes children high in negative emotionality dimensions, such as anger, distress, fear and sadness. On the other hand, these children scored very low in self-regulation dimensions, such as activation control, attentional focusing, inhibitory control and soothability. Profile 3 (Inhibited/Average regulation)takes ininhibited, fearful and shy children, with average scores in effortful control scales, and Profile 4 (Sociable/High regulation) describes quiet and sociable children, with high scores in self-regulation dimensions, such as activation control, inhibitory control, low intensity pleasure and perceptual sensitivity. These profiles were conceptually similar in many aspects to others found in literature. For example, Profile 1 is similar to the profiles "Confident" (Caspi and Silva, 1995) "Social/eager to try" (McClowry, 2002), and "Non-reactive/outgoing" (Sanson et al., 2009) describing approaching and confident children. Profile 2 share characteristics with the profiles "Difficult temperament" (Thomas et al., 1968), "Undercontrolled" (Caspi and Silva, 1995), "High Maintenance" (McClowry, 2002) and "Poor Attention regulation" (Sanson et al., 2009), describing emotionality negative and undercontrolled children. Profile 3 is similar to the profiles "Slow to warm up" (Thomas et al., 1968), "Inhibited" (Caspi and Silva, 1995), "Cautious/Slow to warm up" (McClowry, 2002) and "Reactive/Inhibited" (Sanson et al., 2009), describing fearful and inhibited children. Finally, Profile 4 can be assimilated with profiles "Easy temperament" (Thomas et al., 1968), "Well-adjusted" (Caspi and Silva, 1995), "Industrious" (McClowry, 2002) and "High Attention Regulation" (Sanson et al., 2009), describing adjusted and well-regulated children.

As for our second objective, we found significant differences in academic and social outcomes when we compared the different profiles. With respect to Language and Math achievement, we found that Profile 2 (Emotionally negative/Low regulation) and Profile 4 (Sociable/High regulation) explained children's performance, suggesting that only children who are outstanding for their high or low attentional capabilities are see their academic performance affected positively or negatively. As expected, children belonging to Profile 2 had a higher probability of academic failure, in both Language and Math, while children belonging to Profile 4 showed the opposite. The literature confirms the importance of self-regulation abilities in the classroom, since the focusing and maintaining attention problem puts the children at risk of feeling overwhelmed and, consequently, missing learning opportunities (Zhou et al., 2010; Hernández et al., 2017). In addition, the connection between self-regulation and learning could be particularly meaningful in Spain, where the more rigid and less spontaneous structure, with scarcity of play time, could "punish" less attentive and self-regulated children (Galián et al., 2018). Few studies have analyzed the relations between temperament and academic achievement from a person-centered perspective, but they replicated the line of results found in ours (Robins et al., 1996; Hart et al., 2003).

We also explored the relations between belonging to a particular profile and the probability of being accepted or rejected in the classroom context. As with academic performance, only Profiles 2 (Emotionally negative/Low regulation) and 4 (Sociable/High regulation) explained children's social outcomes. Concretely, children in Profile 2 showed a higher probability of being rejected, and a lower probability of being accepted by their classmates. In contrast, children belonging to Profile 4 were more popular, with a lower probability of being rejected and a higher probability of being accepted by their peers. Again, children's self-regulation abilities seem to be crucial in their social adjustment, and the result suggests that being more inhibited or approaching does not determine children's sociometric status as much as their ability to regulate their emotions in conflicts and social exchanges. Possible explanations as to why children categorized as shy and inhibited do not show a higher probability of being rejected, as occurs in other cultures, are that inhibited children in our study showed average scores in self-regulation and that Spain still shows traits of collectivistic cultures, where shyness is not as punished as in individualistic ones. Other studies that have examined in depth the relations between temperament and social adjustment from a person-centered approach have highlighted the importance of self-regulation in children's social development (Sanson et al., 2009; Laible et al., 2010).

The educational implications of these results need to be discussed. On the one hand, it is necessary to work on the awareness among educational agents of the importance of knowing the different temperament profiles that children can show and the effects that belonging to different categories could have on their academic and social trajectories. In this line, it is important to detect temperament profiles that are in higher risk of maladaptation, such as Emotionally negative/Low regulation, found in our sample, so the training of self-regulation strategies should be considered at prevention and intervention level. In this respect, some child-temperament program has proved its efficiency in schools (INSIGHTS; McClowry, 2014), but temperamental applications in educational settings in Spain remain unexplored. A deep reflection from the institution is also needed to facilitate the adaptation of “difficult children” to the system by making the structure progressively more flexible. Actions such as introducing breaks between demanding attentional tasks and letting the children move and play in the classroom at some moments could be very beneficial for children with more challenging temperamental profiles.

Some limitations of the present study should be mentioned. First, it would be interesting to include another sources of information, such as laboratory measures, in addition to parents, in order to increase future study’s inter-reliability. Indeed, a longitudinal analysis of relations between temperament and adjustment from a person-centered approach would be useful, if aimed at exploring whether temperament profiles are stable over different developmental stages, and whether their impact on academic and social adjustment changes over time. Finally, a comparison between temperamental profiles from different cultures could contribute to better understanding of how culture idiosyncrasy and temperament work interrelatedly in children’s adjustment.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation, to any qualified researcher.

ETHICS STATEMENT

This study was carried out in accordance with the recommendations of University of Murcia, with written informed consent from all participants in accordance with the Declaration of Helsinki.

AUTHOR CONTRIBUTIONS

All authors designed the study, collected the data, contributed to the interpretation of the results, and revised the final manuscript. EA ran the statistical analyses and wrote the manuscript.

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Interaction Between Prematurity and the MAOA Gene on Mental Development in Children: A Longitudinal View

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This study aimed to examine the association of dopamine-related genes with mental and motor development and the gene-environment interaction in preterm and term children. A total of 201 preterm and 111 term children were examined for their development at 6, 12, 18, 24, and 36 months and were genotyped for 15 single-nucleotide polymorphisms (SNPs) in dopamine-related genes (*DRD2, DRD3, DAT1, COMT*, and *MAOA*). An independent sample of 256 preterm children was used for replication. Since the developmental age trends of preterm children differed from those of term children, the analyses were stratified by prematurity. Among the 8 SNPs on the *MAOA* gene examined in the whole learning sample, the results of linkage disequilibrium analysis indicated that they were located in one block (all D′ > 0.9), and rs2239448 was chosen as the tag ($r^2 > 0.85$). In the analysis of individual SNPs in each dopamine-related gene, the tag SNP (rs2239448) in *MAOA* remained significantly associated with the mental scores of preterm children for the interaction with age trend ($p < 0.0001$; largest effect size of 0.65 at 24 months) after Bonferroni correction for multiple testing. Similar findings for rs2239448 were replicated in the independent sample ($p = 0.026$). However, none of the SNPs were associated with the motor scores of preterm children, and none were related to the mental or motor scores of term children. The genetic variants of the *MAOA* gene exert influence on mental development throughout early childhood for preterm, but not term, children.

Keywords: dopamine, genetics, child development, cognitive development, prematurity, longitudinal analysis

INTRODUCTION

Preterm children with very low birth weight (VLBW, birth body weight <1,500 g) face an elevated risk of neurodevelopmental impairments throughout childhood (1–3). Both genetic and environmental factors may contribute to these developmental consequences (4, 5). Recent evidence has shown that the association of preterm birth with severe brain structural abnormalities, congenital disease (6, 7), and developmental problems (8) might be accounted for by a common genetic background. The dopamine system (DA) is a major neurotransmitter in the extrapyramidal system of the brain that involves motor control, endocrine function, cognition, reward system and behavior (9). Dopamine-related genes have long been implicated in child development (9, 10). Certain single nucleotide polymorphisms (SNPs) of dopamine-related genes (e.g., dopamine receptor 2 [*DRD2*] and *DRD3* for DA release, dopamine transporter [*DAT*] for DA clearance, monoamine oxidase A [*MAOA*] and catechol-O-methyltransferase [*COMT*] for DA degradation) have been associated with neurodevelopmental disorders in children. Specifically, several SNPs in *DAT1* (rs27072 and rs2550948) and *MAOA* (i.e., rs12843268, rs2072744, rs5905859, rs3027400, rs2235186, rs2235185, rs2239448, and rs3027407) have been associated with attention deficit hyperactivity disorder (ADHD) (11–16); some SNPs in *DRD2* (rs1800497) and *DRD3* (rs167771) have been related to child affective problems, obesity, working memory impairment, or autism spectrum disorders (ASD) (17–22); and some SNPs in *COMT* (rs4818, rs4680, and rs2075507) have been associated with ASD, aggressive behavior, verbal inhibition and poor working memory in school-aged children (23–25).

A challenge in research on child development is how to incorporate longitudinal data from repeated measurements (26). To date, few studies on the role of dopamine-related genes have adopted a longitudinal view that incorporates repeated measurements of development. Furthermore, existing studies have mainly been conducted among Caucasian populations on more severe developmental morbidities. Whether dopamine-related genes are involved in more common forms of developmental variation in preterm or term children, particularly among non-Caucasian populations, remains poorly understood.

To address the gap in the literature, we turned to a prospective follow-up study of preterm children and their term counterparts. The specific aims of this study were to (1) examine the association of dopamine-related genes with mental and motor development in preterm children with VLBW and term children with normal birth weight at 6, 12, 18, 24, and 36 months of age, (2) examine whether environmental factors (preterm/term birth) interact with dopamine-related genes in children's mental and motor development, and (3) replicate the findings in an independent sample of children.

MATERIALS AND METHODS

Participants

The participants of the learning sample consisted of 201 preterm children with VLBW and 111 term children who were born in or admitted to three hospitals in northern Taiwan during the time periods of 1995–1997 (Cohort I), 2002–2004 (Cohort II), and 2006–2008 (Cohort III). Meanwhile, the replication sample had 256 preterm children with VLBW recruited during the time period of 2012–2014 (Cohort IV) from northern and southern Taiwan. More detailed descriptions of the recruitment are provided in the **Supplementary Methods** and **Figure S1**. Briefly, the inclusion criteria for VLBW preterm children from all cohorts were birth weight $< 1,500$ g, gestational age <37 weeks, and the absence of congenital abnormalities and severe neonatal diseases. The selection criteria for term children included gestational age within 38–42 weeks, birth weight $\geq 2,500$ g, and the absence of congenital abnormality and perinatal disease. All mothers were Taiwanese citizens, were over 18 years old, and had no history of psychiatric disorders or drug or alcohol abuse. All of the participants were from different families.

This study was approved by the Research Ethics Committee of National Taiwan University Hospital and the Institutional Review Board of National Cheng Kung University Hospital, and written informed consent was obtained from parents. Both the Cohort I and II studies were observational studies, while the Cohort III and IV studies were randomized controlled trials (URL: https://clinicaltrials.gov/, identifiers: NCT00173108 and NCT00946244 for Cohort III, and NCT01807533 for Cohort IV, National Taiwan University Hospital). Preterm children in Cohorts III and IV were randomly assigned to the intervention group [clinical-based intervention group and home-based intervention group in Cohort III (27); the family-centered intervention group in Cohort IV (28)] and the usual-care group (Cohorts III and IV). Children in the intervention group received 5 in-hospital and 7 after-discharge intervention sessions from hospitalization to 12 months of corrected age that emphasized environmental modulation, feeding support, massage, child developmental skills, parental support and education, and dyadic interaction activities. Children in the usual-care group received standard care. Randomizations were computer-generated and stratified by gestational age and hospital, with the sequence kept in a locked file and concealed from the parents, medical staff, and outcome examiners. However, the parents and the intervention providers were aware of group allocation. All methods were performed in accordance with the relevant guidelines and regulations.

Measurements

Children in Cohorts I to III had their perinatal and demographic data collected via chart review and parental interviews, and their developmental outcomes were evaluated at 6, 12, 18, 24, and 36 months of age using the Bayley Scales of Infant Development—2nd Edition (BSID-II) (29). Because the BSID-II was revised into the Bayley Scales of Infant and Toddler Development—3rd Edition (Bayley-III) (30) in 2006, both versions were administered to children in Cohort III at five time points (6, 12, 18, 24, and 36 months). For children in Cohort IV, the Bayley-III was administered at four time points (6, 12, 24, and 36 months), and the BSID-II was additionally administered at two time points (24 and 36 months). Because the replication sample had no BSID-II scores at 6 and 12 months, their Bayley-III scores at these ages were transformed into BSID-II scores using a linear regression procedure (31) (details in **Supplementary Methods**).

Genetic Analyses

A buccal cell sample was collected from the cheeks of each child using the Catch-All swabs of the BuccalAmp^{TM} DNA Extraction Kit (Epicenter Biotechnologies, WI, USA). Genomic DNA was extracted from the buccal cell samples following the standard protocol of a commercial kit, the QIAamp Mini Kit (Qiagen, Chatsworth, CA, USA). A total of 15 SNP markers, including 1 in *DRD2* (rs1800497) (19), 1 in *DRD3* (rs167771) (17), 2 in *DAT1* (rs27072 and rs2550948) (12), 3 in *COMT* (rs4818, rs4680; and rs2075507) (23, 24), and 8 in *MAOA* (i.e., rs12843268, rs2072744, rs5905859, rs3027400, rs2235186, rs2235185, rs2239448, and rs3027407) (13), were selected for genotyping using TaqMan analysis and a 7900HT Fast Real-Time PCR System (Applied Biosystems, Foster City, CA, USA). Quality control of the genotyping included duplications, a negative control, and a call rate > 95%. Although there is a well-known variant number of tandem repeats on *MAOA* (32), we did not include it in this study, since its genotyping requires different designs and sophisticated quality control.

None of the genotypes for any of the 15 markers showed significant deviation from Hardy-Weinberg equilibrium, as calculated using PLINK (33) (version 1.07, http://pngu.mgh.harvard.edu/purcell/plink/), with $p > 0.001$ defined as significant departure in either preterm or term children. The *MAOA* gene was examined in females only because males are hemizygous at X-linked loci.

Statistical Analysis

Both the Cohort III and IV were superiority randomized controlled trials. Based on the cognitive and motor development for preterm children for a statistical power of 80%, with an attrition rate of 20% and an alpha level of 0.05, the estimated sample size in the Cohort III study was 43 in each group of preterm children (34). For the Cohort IV study, for a power of 80% with a 20% attrition and an alpha level of 0.05, the estimated sample size in each group was 78 for the 24-month cognitive outcome and 115 for the 24-month motor outcome (27).

We used the QUANTO software (version 1.2.4, http://hydra.usc.edu/gxe) to estimate the overall sample size for the investigation of gene-environment interaction effects in both learning sample and replication sample. To achieve a power of 80%, with continuous outcome and independent individuals design, and an effect size as reported by Babineau et al. (35) for the interaction effect of *5-HTTLPR* gene and environmental factors on child behavior scores ($R^2 = 0.04$ to 0.06, from 3 to 36 months of age) as well as an attrition rate of 20%, an estimated sample size of 153 to 231 children were required.

The power analysis of our existing sample size of 111 term children and 201 preterm children with an alpha level at 0.05 using G*Power software (version 3.1.9.4, Germany) indicated a power of 60–100% for an effect size ranging from 0.11 to 0.65, derived from the standardized mean differences in mental score between the two genotype groups of rs2239448 at 6, 12, 18, 24, and 36 months of age.

Group comparisons were conducted using analysis of variance for continuous variables and chi-square tests for categorical variables. The alpha level was set at 0.05. Because some homozygous genotypes had a small number of individuals for certain SNPs, we merged two adjacent genotypes to adopt a dominant or recessive model as indicated. The pattern of linkage disequilibrium (LD) was examined using Haploview (36).

The relations of the genotypes of individual SNPs to mental and motor raw scores were first examined in the learning sample. We started with a PROC GLIMMIX model with main effects of preterm birth, genotype, and age trend (measured at 5 time points) and their pairwise two-way interactions and three-way interaction with a random intercept and adjustment for covariates, including cohort sources, intervention (yes or no), and sex. Analysis of preterm children's data was additionally adjusted for the effect of gestational age. The cohort differences in developmental scores are described in more detail in **Table S1**. An unstructured covariance structure was selected for the repeated observations based on the Akaike information criterion and the Bayesian information criterion (37). The significance level of the *p*-value obtained from the learning sample was adjusted according to the Bonferroni correction (38, 39). In correcting for multiple testing, we considered the *p*-values of the main effect variables and the interactions together. If the three-way interaction was not significant, the model was refitted by deleting the interaction term from the model. Then, the correction for multiple testing was conducted again for this refitted model without the three-way interaction. Under this circumstance, we conducted stratified analyses to examine the effect of age trend, genotype, and their interaction separately for preterm and term children, with correction for multiple testing within each stratum.

To examine the cumulative effect of significant genetic markers on developmental scores, we used the regression coefficients from mixed-effects models as weights to generate a summarized genetic risk score. The scores were compared using mixed-effects models of longitudinal data at five time points to examine whether the cumulative effects existed or not. Similar correction for multiple testing was conducted for this part of the analysis using the Bonferroni correction (38, 39) by considering the *p*-values of the main effect variables and the interactions together. All statistical analyses were conducted using Statistical Analysis Software (SAS, version 9.3, SAS Institute, Cary, NC, USA).

RESULTS

The birth and demographic characteristics of the participating children are presented in **Table 1**. In the learning sample, preterm children had lower gestational age, birth body weight, parental education, and developmental scores and were more likely to be in Cohort III than were term children (all $p < 0.05$). The results of LD analysis for the 8 SNPs of the *MAOA* in the whole learning sample indicated that they were located in one block (all $D' > 0.9$) (**Figure S2**), and rs2239448 was therefore chosen as the tag ($r^2 > 0.85$).

The distributions of the 8 SNPs (rs2239448 as the tag of *MAOA*) genotyped in this study are displayed in **Table S2**. We adopted a dominant model for the designated allele 1, which was determined by collapsing two adjacent genotypes if their distributions were closer than the remaining genotypes or one

TABLE 1 | Birth and demographic characteristics of preterm and term children.

Characteristics	Learning sample (Cohort I-III)		Replication sample (Cohort IV)
	Preterm ($N = 201$)	**Term ($N = 111$)**	**Preterm ($N = 256$)**
	Number (%)	**Number (%)**	**Number (%)**
Male sex	99 (49%)	59 (53%)	129 (50%)
Maternal education[a]			
College or above	126 (63%)	83 (82%)	197 (77%)
High school below	75 (37%)	18 (18%)	59 (23%)
Paternal education[a]			
College or above	120 (60%)	83 (81%)	192 (75%)
High school or below	81 (40%)	19 (19%)	64 (25%)
Cohort*			
I	27 (14%)	38 (34%)	–
II	39 (19%)	24 (22%)	–
III	135 (67%)	49 (44%)	–
Intervention type			
Intervention	95 (47%)	–	125 (49%)
Standard care	106 (53%)	–	131 (51%)
	Mean (SD)	**Mean (SD)**	**Mean (SD)**
Gestational age (weeks)[a]	29.5 (2.9)	39.1 (1.0)	29.6 (2.6)
Birth body weight (g)[a]	1,115.5 (265.0)	3,287.7 (380.7)	1,112.1 (260.1)
Bayley mental raw scores[a]			
6 months	57.6 (4.4)	59.1 (2.7)	57.5 (2.9)
12 months	82.4 (4.1)	85.1 (3.5)	83.1 (2.5)
18 months	104.8 (6.2)	108.5 (5.5)	–
24 months	127.3 (8.0)	131.2 (7.0)	129.3 (7.3)
36 months	150.7 (6.4)	157.3 (5.2)	149.8 (7.7)
Bayley motor raw scores[a]			
6 months	35.0 (4.1)	36.4 (3.5)	–
12 months	59.1 (2.9)	61.5 (3.1)	–
18 months	71.4 (3.1)	72.8 (2.4)	–
24 months	81.0 (3.6)	83.0 (3.2)	–
36 months	97.0 (3.9)	100.8 (3.5)	–

[a] $p < 0.05$ in comparing preterm children with term children in the learning sample using t-tests or analysis of variance for continuous variables and chi-square tests for categorical variables.

homozygous group having a very small number (one in *DRD3* and two in *DAT1*).

Effects of Genetic Factors and Prematurity on Longitudinal Child Development

The results of the mixed-effects model analysis using a full model for the mental score with Bonferroni correction for 56 *p*-values are displayed in **Table S3**. Because the three-way interaction was not significant in each of the 8 SNPs, we then removed the three-way interaction and refitted the model (**Table S4**). Regarding the main effect, age trend and preterm birth were highly significant even after Bonferroni correction for 48 *p*-values, and genotype was not. In terms of interaction, the preterm × age trend was significant for all SNPs, but both the genotype × age trend and genotype × preterm interactions were significant only for the *MAOA* rs2239448, though the latter was not significant after Bonferroni correction. One possible reason for not reaching significance for the genotype × preterm interaction is the relatively small sample size for the term group ($N = 111$) vs. the preterm group ($N = 201$).

Because there was no three-way interaction and the sample was recruited separately for preterm and term children, we then conducted stratified analyses by preterm birth to examine the effect of age trend, genotype, and their interaction. For the stratum of preterm children's mental score (**Table 2**), the effect of age trend was significant for all SNPs, but the effect of genotype and genotype × age trend were significant only for the *MAOA* rs2239448 after Bonferroni correction for 24 *p*-values and adjustment for the effect of gestational age, cohort sources, intervention and sex. For illustration, the mean differences between two genotype groups at the five time points are

TABLE 2 | Relations of genotypes with the mental raw scores at 6, 12, 18, 24, and 36 months of age in preterm children (cohort, intervention, sex, and gestational age are treated as covariates).

Gene	SNP (allele 1/2)	Difference in mental score (presence of allele 1 - absence of allele 1)					Age trend		Genotype		Genotype × Age trend	
		6 months	12 months	18 months	24 months	36 months	*F*-value	*p*	*F*-value	*p*	*F*-value	*p*
Learning sample												
DRD2	rs1800497 (G/A)	1.78	0.56	−0.92	0.18	1.56	6,798	<0.0001[b]	1.39	0.24	1.34	0.25
DRD3	rs167771 (G/A)[a]	−0.20	0.91	1.78	1.95	1.46	11,707	<0.0001[b]	1.74	0.19	1.47	0.21
DAT1	rs27072 (T/C)[a]	−1.60	0.16	−0.58	−0.05	−0.35	12,396	<0.0001[b]	2.84	0.09	0.18	0.94
DAT1	rs2550948 (T/C)[a]	−0.01	0.10	0.33	0.92	0.88	10,966	<0.0001[b]	1.09	0.29	0.56	0.69
COMT	rs4818 (C/G)	−0.29	0.03	0.09	1.33	0.82	9,005	<0.0001[b]	0.00	0.99	0.92	0.45
COMT	rs4680 (G/A)	−1.02	0.09	−1.67	−1.75	−0.91	8,548	<0.0001[b]	2.85	0.09	0.88	0.47
COMT	rs2075507 (T/C)	−0.50	0.50	−0.95	−1.50	1.45	5,389	<0.0001[b]	0.22	0.64	1.07	0.37
MAOA	rs2239448 (T/C)	0.50	1.09	3.16	5.23	2.49	11,026	<0.0001[b]	8.76	0.0032	8.27	<0.0001[b]
Replication sample												
MAOA	rs2239448 (T/C)	−0.04	0.84	–	3.11	1.38	8,140	<0.0001	4.19	0.041	3.12	0.026

The BSID-II scores at 6 and 12 months of age were transformed from Bayley-III, and the significance level was set as 0.05 in the replication sample. DRD2/3, dopamine D2/D3 receptors; DAT1, dopamine transporter 1; COMT, catechol-O-methyltransferase; MAOA, monoamine oxidase A. [a]Determined by collapsing two adjacent genotypes due to one homozygous group having a very small number. [b]The significance levels reached the threshold of Bonferroni correction for 24 p-values in the learning sample.

also displayed for each SNP in **Table 2** (more detailed mental score distributions are presented in **Table S5**). Of note, *MAOA* rs2239448 showed the smallest genotype effect ($p = 0.0032$) and genotype × age trend interaction effect ($p < 0.0001$), yet only the latter remained statistically significant after Bonferroni correction. The effect size for *MAOA* rs2239448 (i.e., mean differences between the two genotype groups divided by the standard deviation of preterm children) in relation to the mental score was 0.11, 0.27, 0.51, 0.65, and 0.39 at 6, 12, 18, 24, and 36 months of age, respectively.

For the stratum of term children's mental scores (**Table S6**), the effect of age trend was significant for all SNPs, but the SNPs showed neither genotype nor genotype × age trend effects after Bonferroni correction for 24 *p*-values.

For the motor scores, we first used a full model containing the three main variables, all of their possible interactions, and covariates (**Table S7**). Regarding the main effect, age trend and preterm birth were significant, while genotype was not. In terms of interactions, the preterm × age trend was significant, which was also expected according to a previous study (26), but the genotype × age trend and genotype × preterm interactions and the three-way interaction were not significant. Even if we refitted the model without the three-way or two-way interactions, the results still indicated that only preterm, age trend, and preterm × age trend were significant.

Replicating the Finding in *MAOA*

To examine the robustness of the influence of *MAOA* rs2239448 and its interaction with age trend on the mental scores of preterm children, we further genotyped this SNP in an independent sample of 256 preterm children. *MAOA* rs2239448 had a significant main effect ($p = 0.041$) as well as a significant interaction effect with age trend ($p = 0.026$) (**Table 2**, bottom row). For *MAOA* rs2239448 in this replication sample, its effect size with the mental score was −0.01, 0.34, 0.43, and 0.18 at 6, 12, 24, and 36 months of age, respectively. Of note, the SD at 12 months of age (2.5) was derived from the Bayley-III, whereas the developmental data at 36 months of age (7.7) was derived from the BSID-II, resulting in a larger effect size for the former even though it had a smaller group difference than the latter.

To summarize the findings for *MAOA* rs2239448, the mean mental scores at five time points for the two genotype groups (presence of allele 1 vs. absence of allele 1) are depicted separately for the preterm children in the learning sample (**Figure 1A**), the term children in the learning sample (**Figure 1B**), and the preterm children in the replication sample (**Figure 1C**).

Then, the most significant *MAOA* rs2239448 was combined sequentially with seven other markers according to the ascending order of *p*-value, and the cumulative genetic dose was subjected to mixed-effects model analysis after Bonferroni correction for 56 *p*-values (**Table S8**). Nevertheless, the combination of rs2239448 with other SNPs did not lead to any increase in the effect estimates regarding genotype, i.e., the main effect of genotype, genotype × age trend, genotype × preterm, and genotype × preterm × age trend.

DISCUSSION

In this prospective study, we evaluated the potential influence of 15 dopamine-related genetic variants on developmental scores using mixed-effects models of longitudinal data to examine three main-effects variables (preterm, genotype, and age trend) and their possible interactions. After removing the three-way interaction due to its lack of statistical significance, the three two-way interactions were significant for the *MAOA* variant. After stratification by preterm birth, the effect of the

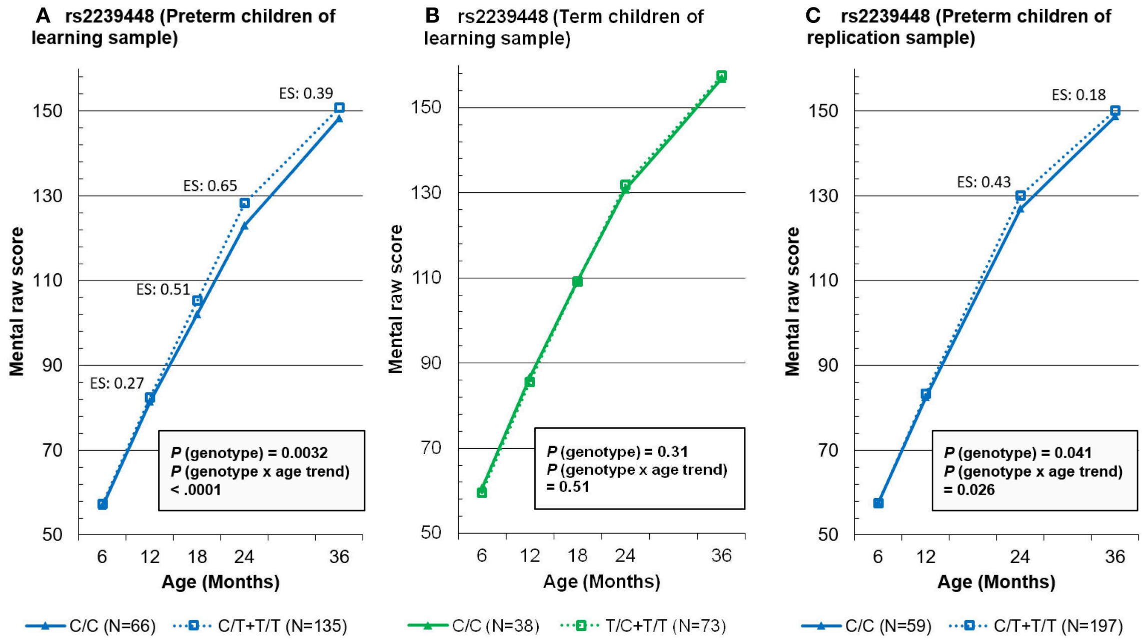

FIGURE 1 | Mixed-effects model-based mental raw score at 5 time points by the genotypes of *MAOA* tag rs2239448 in preterm and term children in the learning sample (**A,B**, respectively) and in preterm children in the replication sample **(C)**. ES, effect size.

genotype × age trend remained significant only for preterm children's mental scores for the variant of *MAOA* rs2239448 after Bonferroni correction. Furthermore, the influence of *MAOA* rs2239448 (genotype main effect and genotype × age trend interaction) in preterm children was successfully replicated in an independent sample. Nevertheless, a combination of *MAOA* rs2239448 with the SNPs of other dopamine-related genes failed to improve the association. These findings provide insightful information on the gene (*MAOA* rs2239448) × environment (preterm birth) interaction regarding the age trend of mental development.

As expected, preterm children demonstrated persistently lower mental and motor scores than their term peers did from 6 to 36 months of age. Such results were in line with previous findings indicating that preterm children showed poorer motor and mental development than their term counterparts did in their follow-up to 24 months of age (27).

Among the variants of dopamine-related genes examined, only those of *MAOA* exhibited an association with the mental scores of preterm children at different ages, with a small to medium effect size in the learning sample and a small effect size in the replication sample. For example, the preterm children with two genotypes on the *MAOA* rs2239448 had the largest difference in mental scores of 5.23 (i.e., a medium effect size of 0.65) in the learning sample and of 3.11 (i.e., a small effect size of 0.43) in the replication sample at 24 months of age. Although these *MAOA* variants have been found to be associated with some childhood-onset mental disorders, including ADHD (13) and ASD (40), this study is the first to demonstrate that the *MAOA* variants exert influence on the mental development of preterm children, providing further support for the role of *MAOA* implicated in prior animal studies (41).

On the other hand, our findings showed no association of dopamine-related genes with mental development in term children. One possibility is that the dopamine-related brain functions of term children have less variation than those of preterm children (42, 43). Hence, many previous studies have consistently reported no association between variants of dopamine-related genes and cognition in healthy children and adolescents (44, 45). In contrast, some prior studies found that the influence of dopamine-related genes could be detected only under certain environmental contexts, for example, the influence of *MAOA* on antisocial problems moderated by childhood maltreatment (46, 47), the influence of *5-HTT* on dysregulation in children moderated by prenatal depression in mothers (35), and the influence of *5-HTT* behavioral development in children moderated by child care quality (48). Therefore, the effect of dopamine-related genes on mental development may be very small unless the children underwent an adverse developmental environment, e.g., preterm birth in this study. Thus, our findings implied that preterm children are more vulnerable to the influence of *MAOA* on mental development.

A combination of the *MAOA* rs2239448 with the SNPs of other dopamine-related genes in this study did not lead to a stronger association with mental development in preterm children, as indicated in a previous study (49). One possibility is that our chosen genetic variants of other dopamine-related genes were mainly based on Caucasian populations and not on the most informative alleles in our study population.

Our failure to find any association of the variants of dopamine-related genes with motor development in preterm and term children is, to some extent, not surprising, given that our target markers were chosen based on studies focusing on childhood-onset diseases such as ADHD and ASDs. Future studies on child motor development need to consider genetic markers that may have associations with other motor-related diseases or functions.

Our findings indicate that prematurity in children with the absence of the T allele of *MAOA* rs2239448 is associated with slower mental development. In practice, the *MAOA* rs2239448 may be used to identify preterm children who are vulnerable and can benefit from early intervention to help them overcome the slowing pace of mental development. It is also warranted to investigate whether the *MAOA* variants moderate the effect of intervention in preterm children and help develop personalized interventions for preterm children.

This study has the following strengths: (1) the incorporation of a longitudinal design with follow-ups at 6, 12, 18, 24, and 36 months on both the mental and motor development of preterm and term children and (2) the successful replication of the robust results in an independent sample of preterm children. However, several limitations are important to note. First, because preterm children with severe brain damage or neonatal diseases were excluded from this study, our findings may not be generalizable to those with more severe developmental disorders. Second, this study consisted of participants from several cohorts established in different years. While clinical practice and environmental exposure might vary across time, we treated cohort membership as a covariate to control for potential confounding effects of birth year. Some information was not collected for some cohorts, such as socioeconomic status for Cohort I and Cohort II. The failure to include socioeconomic status as a covariate in our analysis might render our adjustment for potential confounders inadequate. Third, our study focused only on certain candidate SNPs selected from previous genome-wide association studies. Future research may explore the *MAOA* variable number of tandem repeats for more genetic information. Finally, the BSID-II scores at both 6 and 12 months of age in the replication sample of Cohort IV were transformed from the Bayley-III scores via regression modeling. This might not overcome the difference between the two, e.g., a smaller standard deviation of the Bayley-III scores at the age of 12 months than its counterparts of BSID-II scores.

In conclusion, this prospective study of preterm and term children demonstrated that the *MAOA* rs2239448 variants were significantly associated with the mental scores of preterm children for the main effect and its interaction with the age trend. Similar findings for *MAOA* rs2239448 were replicated in an independent sample of preterm children. However, none of the SNPs were associated with the motor scores of preterm children, and none were related to the mental or motor scores of term children. Our results have shed new light on the genetic influences of *MAOA* on mental development in preterm children and have implications for intervention.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation, to any qualified researcher.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Ethics Committee of National Taiwan University Hospital (Identifier on ClinicalTrials.gov: NCT00173108, NCT00946244, and NCT01807533). Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

N-JY conceptualized and designed the medical data collection and analysis procedures, contributed to child developmental data collection, carried out the genetic data collection and genotyping experiment, and drafted the initial manuscript. W-SH and C-HL supervised case enrollment and medical data collection in two different hospitals. C-IT carried out the DNA extraction and assisted genotyping experiment. W-YL and P-HK contributed to the statistical analysis and interpretation of the data. Y-TY helped with child developmental data collection. WC supervised the genotyping experiment and statistical analysis, contributed to the interpretation of data, and revised the manuscript critically. S-FJ designed the study, procured the grant support, coordinated all data collection of all the hospitals, supervised the child developmental data collection, and critically revised the manuscript. All authors reviewed and approved the manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fped.2020.00092/full#supplementary-material

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BRIEF RESEARCH REPORT
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Executive Function Training Improves Emotional Competence for Preschool Children: The Roles of Inhibition Control and Working Memory

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The study examined how executive function (EF) training could improve children's emotional competence (EC). Children (N = 55; M_{age} = 50.64 months) were assigned into two groups, namely the EF training group and the no-training group. The present study attempted to use a 2 (group: EF training VS no-training) × 2 (test time: pretest VS post-test) between-and- within-subjects experimental design to investigate the effect of EF training on the improvement of EC for 4-year-old children. Results showed that, (1) children in EF training group had significantly higher scores on EC than that of no-training group; (2) The change of inhibition control and working memory could significantly predict their variation of EC. These results suggested that the improvement of EC caused by EF training could be linked to the ability of inhibition control and working memory.

Keywords: executive function, executive function training, 2 months, emotional competence, preschool children

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INTRODUCTION

Emotional competence (EC) refers to the abilities of recognize, comprehend, express, and regulate emotions (Mirabile, 2009; Lahaye et al., 2011; Riquelme and Montero, 2013). Emotional competence is developed throughout childhood (Liu and Chen, 2003; Pons et al., 2003). It is a fundamental ability that can help children to handle a challenge, establish a social relationship, keep mental health, and adapt to various social environment (Wang et al., 2014; Wardiesielski et al., 2019). It has been shown that children's emotional competence is associated with their executive function (EF) (Rhoades et al., 2009; Smith et al., 2014; Kwok et al., 2015; Lantrip et al., 2015; Healy et al., 2018). However, the causal relation between them is unclear due to the limited evidence from previous experimental studies. The study aimed to determine the extent to which EF training could improve children's emotional competence.

Executive function involves four basic abilities including inhibition control, cognitive flexibility, working memory, and problem-solving (Miyake et al., 2000; Hong et al., 2004; Oh and Lewis, 2008; Weyandt et al., 2014). Children with better emotional competence tend to exhibit better ability of inhibition control, shifting and problem-solving, but less impulsivity (Zelazo et al., 2003, 2010; Silkenbeumer et al., 2016).

Previous studies found that children's emotional competence involved affective system, attentional system and self-control system (Zhou and Zhou, 2003; Zelazo and Carlson, 2012).

Silkenbeumer et al. (2016) found that children's development of emotional competence required the ability of internalization inhibition and modified the elicited emotional action. Moreover, inhibition control can influence children's emotional competence. Gerstadt et al. (1994) used Day/Night task to assess children's EF (e.g., Inhibition control ability), and Kusche (1984) used Emotional Inventory task to measure children's emotional competence ability (Kusche, 1984; Gerstadt et al., 1994). The study found that better ability of inhibition control had better performance of emotional competence (Fox et al., 2001; Fox and Calkins, 2003; Liew, 2012). Rhoades et al. (2009) demonstrated that children who had better EF were more likely to be higher on social-emotional skills and lower on problem behaviors in a sample of 146 preschools. Likewise, Garcia-Andres et al. (2010) investigated the relationship between EF and emotional regulation in 7 and 8-year-old children. They gave children Wisconsin Card Sorting Test (WSCN), Backward Digit Span (DIG), Five Digit Test (Stroop effect), and Tower of Hanoi (ToH) to measure their EFs. They found that children with the better EF performed better in the use of emotional regulation strategies (Garcia-Andres et al., 2010). Notably, children's EF (e.g., inhibition control) can promote the development of emotional strategies.

Executive function is related to individual emotional competence, but to date, it is unclear to the causal relationship between them. Previous literature has shown that children's EFs could influence children's ECs. However, to our knowledge, there is no strong causal evidence to demonstrate whether EF training can influence emotional competence. We state that children's emotional competence needs EF to better adjust to the present situation. For instance, the test of emotional comprehension includes the understanding of belief-based emotions, and it is well-documented that belief-based reasoning involves EF. Moreover, the test of emotional regulation and emotional expression might require inhibition control. For example, the children should control their own negative emotion (e.g., anger) during interaction to better manage emotions. To address this issue, we used a 2 (groups: No-training VS EF training) × 2 (test time: pretest VS post-test) between-and-within-subjects experimental design (that is experimental and control group pretest–posttest design). In EF training group, the well-trained research assistant guided children to carry out EF training activities (20∼30 min every time, twice a week, and 12 times in total). In No-training group, the children participated in daily class activities without EF training at the same time. Two hypotheses were examined: (1) EF training group would have a greater increase in emotional competence than no-training group after the intervention, (2) Children's EF change would explain the improvement of children's emotional competence.

METHOD

Participants

Fifty-five healthy Chinese preschoolers (EF training group: $N = 29$, $M_{age} = 50.65$ months, $SD = 3.11$ months; No-training group, $N = 26$, $M_{age} = 50.63$ months, $SD = 3.32$ months) participated in the study. These children were recruited from two classes (middle classes, age = 4∼5 years old) of local kindergartens. Four children of No-training group Were excluded from the analyses because they didn't participate in the post-test. The intervention took place over a 2-month period and involved fifty-one children. We Have modified them as "six children of EF training group participated EF training at One time. All children were given informed consent for this study (parental consent and participant assent for children). The study was approved by the Southwest University's Academic Ethics Committee (IRB: H19063).

Design

The study used a 2 (group: No-training VS EF training) × 2 (test time: pre VS post) between- and-within-subjects experimental design. The group (group: No-training VS EF training) was the between-subject variable but the test time (test time: pre VS post) was the within-subject variable. The No-training group was control group, and the EF training group was experimental group. Otherwise, the study was approved by the Southwest University's Academic Ethics Committee.

Measures and Materials

According to previous literatures, Facial Expression Match and Recognition, Test of Emotion Comprehension (TEC), Situational Storytelling of Emotion Expression, and Situational Storytelling of Emotion Regulation were widely used to measure the ability of children's emotional competence capacity (Dodge, 1989; Carlson et al., 2002; Pons et al., 2002; Ling and Ya, 2003; He et al., 2005; Bierman et al., 2008; Mirabile, 2009; Lahaye et al., 2011; Dong, 2012; Riquelme and Montero, 2013). The Emotional Stroop Test, Dimensional Change Card Sorting (DCCS), Memory for Picture of Wechsler Intelligence Scale and Situational Storytelling of Problem Solving was used to measure children's EF (Wechsler, 1974, 2003; Ruff et al., 1998; Miyake et al., 2000; Ruff and Capozzoli, 2003; Chrysikou and Weisberg, 2005; Zelazo, 2006; Besnier et al., 2008).

Emotional Recognition: Facial Expression Match and Recognition

The Facial Expression Match and Recognition was used to measure children's emotional recognition in this study (Bierman et al., 2008; Dong, 2012). All pictures in the task were selected from database of Nimstim Facial Expressions of Emotion (Tottenham et al., 2009). All pictures in the task were selected Chinese faces, and the picture numbers of male faces and female faces were equal. Children were asked to recognize pictures with four types of facial emotions (happy, sad, angry, and fear) and then match them with emotional words (e.g., which picture is happy?). Two points were assigned for each correct-recognization picture and one point for each correct-match picture, and thus the final score of emotional recognition should range from 0 to 12 points (Markham and Wang, 1996; Mo and Su, 2004; Quan et al., 2019).

Emotional Comprehension: Test of Emotion Comprehension (TEC)

Test of Emotion Comprehension (TEC), including four sections, was used to measure children's emotional Comprehension in this study (i.e., Belief, Desire, Reminder, and Cause) (Pons et al., 2002). The TEC test depended on a comic book with a simple cartoon scenario on each page (the size of comic book = 21 cm by 29.7 cm). There were four emotional outcomes (e.g., happy, sad, angry, and fear) on the bottom of the page. All the facial expressions typically represented each scenario. The test procedure included two steps: (1) The experimenter read the cartoon (two stories in each section). (2) The child was asked to point out the most appropriate picture of match the four possible emotional outcomes. Four sections in total with a fixed order, (I) Understanding desire-based emotions (e.g., the same situation but opposite desires individually); (II) Understanding belief-based emotions (e.g., attribution of an emotion to a chick who enjoying worms without knowing that an eagle was hiding in the tree); (III) Understanding the cue of a reminder about a present emotional state (e.g., perception of the emotion to a character that was reminded of the loss of a pet); (IV) Understanding external causes of emotions (e.g., attribution of an emotion to a character being displeased by a puck). Children would get one point when he or she answered a theme correctly. The final score of emotional comprehension was calculated by the summation of points in each.

Emotional Expression: Situational Storytelling of Emotion Expression

The Situational Storytelling of Emotion Expression was administered to assess emotional expression (Carlson et al., 2002; He et al., 2005). Children were presented with a comic book with a simple cartoon scenario in the center (the size of comic book = 21 cm by 29.7 cm). Four situational storytelling (two positive situations and two negative situations) was included. Every situation had three questions needed to be answered by pointing to facial pictures (angry, sad, calm, and happy). For example, the experimenter read a positive situation (e.g., "Today is Mike's birthday, Mike's friend bought a birthday gift to him, Mike wanted to get a toy car, but when he opened the gift and found an ugly doll there. If you were Mike, what would you choose?"). Question one is about true emotion (sad or angry): "what is Mike feeling?" Question two is about ego-oriented motivation: "If Mike was shown the true emotion (sad or angry), and his friend would never send him the birthday gift. What do you think Mike should show on his face?" Question three is about social goal orientation: "If Mike showed the true emotion (sad or angry), and his friend would be very sad. What do you think Mike should show on his face?" There were four emotional expression strategies: (1) calming: when true emotion was angry, sad, or happy, but preschoolers chose calm facial picture; (2) hiding: when true emotion was sad or angry, but children chose happy facial picture; (3) exaggerating: when true emotion was sad, but preschoolers chose angry facial picture; (4) weakening: when true emotion was angry, but kids chose sad facial picture. When the strategy was appropriate for the situation. The participants would get two points for appropriate match between strategy and situation, one point when the strategy did not coincide the situation, and zero point when the children didn't use strategy. The final score of Situational Storytelling of Emotion Expression was calculated by adding up the scores from four blocks.

Emotion Regulation: Situational Storytelling of Emotion Regulation

The Situational Storytelling of Emotion Regulation was chosen in order to assess emotional regulation (Dodge, 1989; Ling and Ya, 2003; Dong, 2012). The Situational Storytelling of Emotion Regulation was showed by a comic book with a simple cartoon scenario (the size of comic book = 21 cm by 29.7 cm). The test of emotional regulation includes four situational storytelling (two positive situations and two negative situations). Every emotional situation had four strategies to choose after the experimenter read the situational story to the child. For instance, the experimenter read a negative situation (e.g., Mike was stacking toy blocks carefully, but his good friend walked to him and said that your toy blocks stacked bad, and his friend pushed the blocks down with saying that "let me help you to stack") and asked the child, "what would you do if you were the main character?" Four options were provided: (1) Self-repression: No speaking, playing the other toys; (2) relying on the adult: Cry to the teacher; (3) Impulsive behavior: Pushing companion down; and (4) Self-assertion: Asking his friend to stack together. Two points would be got for choosing "Self-assertion," one point for "Self-repression" and relying on the adult, one point for "Self-assertion" and relying on adult, but zero point for "Impulsive behavior." The final score of Situational Storytelling of Emotion Regulation was calculated by adding up two positive and negative mean scores.

Inhibition Control: Emotional Stroop Test

The Emotional Stroop Test was administered to children's inhibitory control, which was an element of children's EF (Besnier et al., 2008). An emotional picture was used as an automatic process in the Emotional Stroop Test. Participants was asked to ignore the word meaning of words when they were instructed to read the facial expression (Bentall and Kaney, 1989; Cothran and Larsen, 2008). The Emotional Stroop Test required participants to inhibit the word on the facial expression pictures, but answered to facial expression ignoring the meaning of word. The timed reaction and accuracy were analyzed during the Stroop task. In our study, eight trials were presented to children as the exercise for understanding experimental rules. The experimenter told children "When the happy facial expression is shown on the screen, you tell me "unhappy." The final experiment included three sessions. Every session presented 16 trails with 8 happy faces and 8 unhappy faces with the random order. All pictures in the task were selected from database of Nimstim Facial Expressions of Emotion (Tottenham et al., 2009). Considering that children in the study were too young to make choice by pushing the button (aged from 46 to 56 months), the experimenter helped them push it. Given that, we only recorded accurate rate but ignored reaction time.

Cognitive Flexibility: Dimensional Change Card Sorting

We selected Dimensional Change Card Sorting (DCCS) with the version of 3∼5 years-old children (Gerstadt et al., 1994; Zelazo et al., 1996; Zelazo, 2006) to assess the cognitive flexibility in our study. Firstly, the dimensions had two parts (color dimension and shape dimension), which was relevant during the pre-switch phase as the standard version. For instance, if the color dimension had been chosen as a pre-switch dimension, and the shape was a post-switch dimension. Practicing block included six switch trails (three color switch trails and three shape trails). Secondly, the experimenter told the children, "Now we're going to play a color-shape game. In the color game, all green ones gone here [pointing to the green tray], and all white ones gone here [pointing to the white tray]. In the shape game, now we're going to play a new game, all rabbits gone here [pointing to the rabbit tray], and all boats gone here [pointing to the boat tray]." Children aged from 3 to 5 years old usually sorted correctly on all six pre-switch color trials. The child who could sort five out of six post-switch trials correctly was regarded as the pass for the post-switch. A score of 0 was assigned if the child could not pass the pre-switch phase of standard version; a score of 1 was assigned if the child passed the pre-switch phase of standard version but failed the post-switch phase; 2 points if the child passed both pre-switch and post-switch of standard version but failed the next switch. The final of score of Dimensional Change Card Sorting (DCCS) ranged from 0 point to 6 points.

Working Memory: Memory for Picture of Wechsler Intelligence Scale for Children (IV)

Memory for Picture of Wechsler Intelligence Scale for Children (IV) was administered in order to measure preschool children's working memory, which was an essential component of EF (Wechsler, 1974, 2003). In the present study, we chose the normative testing for 4 year-old children. The test of working memory included 35 sets of pictures, which ranged from one picture to six pictures in each set. Children was asked to point out the target pictures that the experimenter showed. One point was given when each question was correctly answered, and the test was ended when the child failed to answer four sets of questions continuously. The final score of children's working memory was calculated by the sum up all sets together.

Problem Solving: Situational Storytelling of Problem Solving

The Situational Storytelling of Problem Solving was administered in order to measure Problem Solving as a part of EF (Ruff et al., 1998; Ruff and Capozzoli, 2003; Chrysikou and Weisberg, 2005). The Situational Storytelling of Problem Solving relied on a comic book with a simple cartoon scenario on the page (the size of comic book = 21 cm by 29.7 cm), which included two situational storytelling (one positive situation and one negative situation). Every situation had four strategies to choose after the experimenter read the situational story. For instance, the experimenter read a positive situation (e.g., Mike and Lily play slide together after reciting children's song, Mike had recited, but Lily couldn't recite), and the experimenter asked the child, "what would you do if you were Mike to play slide with Lily as soon as possible?" The children would get two points if he or she choose "A: I would help her to recite"; he would get one point if he choose "B: Waiting for her to recite"; he would get half of a point when he choose "C: I didn't wait for him to play slide by myself," and he would get zero points when he choose "D: I don't know." The final score of Situational Storytelling of problem solving was the mean of the points of two situations (Dennis et al., 2009; Suor et al., 2017).

Procedure

After getting the consent from the parents and kindergarten's permission, children were tested individually in a quiet room. All the tests were divided into two sections. Each session lasted approximately 35–40 min. As a warm-up, the experimenters asked the child for whether he or she liked to play games and which games he or she played usually. The experimenter measured the children's EFs and ECs before and after. Of note, the Facial Expression Match and Recognition was assessed at the start of pretest and post-test, and other measures were counterbalanced across participants.

Manipulation

Children from two different kindergarten classes (Class 1: 29 children; Class 2: 26 children) were randomly assigned to EF training group (N = 29) and No-training group (N = 26).

Training Group

Twenty-nine children were randomly assigned to the EF training group. The self-developed EF training curriculum was based on four EF sub- components, which was used for the age of 4–5 year-old children. The training sessions was twice a week over 2 months (12 session in total). In order to have a better training effect, only 5 or 6 children took the 20–30-min EF training class for each time by well-trained research assistants: (1) 1^{st}–4^{th} session, the aim is to promote the inhibition control ability; (2) 5^{th}–6^{th} session, the aim is changed to promote the cognitive flexibility; (3) 7^{th}–8^{th} session, the aim is to develop the working memory; (4) 9^{th}–10^{th} session, the aim is to improve the problem solving ability; (5) 11^{th}–12^{th} session, the aim is to review all the training classes above. (Detailed curriculums see **Table 1**).

No-Training Group

Children in the No-training group took the normal course activities. All is same to training group but there is no EF training classes.

RESULTS

Firstly, in order to make the EF and EC comparable, the value of them were converted into Z scores. In the study, we got the score of EC by calculating the summation of score of emotional recognition, emotional comprehension, emotional expression, and emotional regulation. Likewise, the score of EF was calculated by summing up the scores of the inhibition control, cognitive flexibility, working memory, and problem solving.

The G*Power 3 was used to compute the statistical power in this study ($N = 55$). The test effects setting was effects in within-between-subjects designs for two groups. The results showed that the power (1-β err prob) was 0.997, suggesting a strong statistical power in our study (Faul et al., 2007).

Pretest Data Analysis

To corroborate earlier findings on the relationship between EF and emotional competence (EC), we performed correlation analysis between EF and EC during pretest. The result showed that it was a significantly positive association between EF and EC, $r = 0.679$, $p < 0.001$. The finding suggested that children's ECs could be improved by training EFs.

To examine the difference of ECs between the EF training group and No-training group during pretest, we performed independent sample t-tests. The result showed that the EC and sub-capacities had no significant difference, $ps > 0.05$. Overall, the result revealed that EF and EC didn't show significant difference during pretest, but No-training group performed better than EF training group on inhibition control, $t = 2.996$,

TABLE 1 | The syllabus of EF training class.

Training abilities	Sessions	Curricular title	Example items
Inhibition control	1–4	Big and small watermelon	**Aim:** Improve the conflict control ability. **Operation:** When research assistant gesticulate "big watermelon," children should answer "small watermelon." When research assistant gesticulate "small watermelon," children should answer "big watermelon." **Conflict condition:** Gig VS Small.
		Sun and star	**Aim:** Improve the conflict control ability. **Operation:** When research assistant show "sun picture," children should answer "night." When research assistant show "star picture," children should answer "day." **Conflict condition:** Day VS Night.
		Black and white magic wand	**Aim:** Improve the conflict control ability. **Operation:** When research assistant gesticulate "white," children should answer "black." When research assistant gesticulate "black," children should answer "white." **Conflict condition:** Black VS White.
		The pony across the river	**Aim:** Improve the conflict control ability. **Operation:** When research assistant show "white river," children should walk "black river." When research assistant show "black river," children should walk "white river." **Conflict condition:** Black VS White.
Cognitive flexibility	5–6	Mike's birthday	**Aim:** Improve the cognitive flexibility. **Operation:** Shown six positive emotional scenes and six negative emotional scenes (e.g., getting birthday gift). **Shifted condition:** From positive emotion to negative emotion.
		Book was trampled	**Aim:** Improve the cognitive flexibility. **Operation:** Shown six negative emotional scenes and six positive emotional scenes (e.g., the book was trampled). **Shifted condition:** From negative emotion to positive emotion.
Working memory	7–8	Fruit platter	**Aim:** Improve the working memory. **Operation:** Reciting fruits on forward and backward [range: 2∼5] (e.g., recited forward "apple\pear\banana" and recited backward "banana\pear\apple"). **Processing sequence:** First reciting forward and backward.
		Digital overturning	**Aim:** Improve the working memory. **Operation:** Reciting digits on forward and backward [range: 2∼5] (e.g., recited forward "1\5\2" and recited backward "2\5\1") **Processing sequence:** First reciting forward and backward
Problem solving	9–10	Lamb was eaten	**Aim:** Improve the problem solving. **Operation:** (1) Acting the story; (2) Asked the children"if you were the master, what would you do?" (e.g., Milk's lamb was eaten by a wolf. If you were Milk, what would you do?). **Training core abilities:** problem solving\inhibition control
		Flowers was crushed	**Aim:** Improve the problem solving. **Operation:** (1) Acting the story; (2) Asked the children"if you were the master, what would you do?" (e.g., Milk's flowers were crushed by a dog. If you were Milk, what would you do?) **Training core abilities:** problem solving\inhibition control
Combined training	11–12	Kitchen parade	**Aim:** Reconsolidating. **Operation:** (1) Divided into two groups, each group had 3 children; (2) One group cooked, the other group waited; (3) Limited ingredients, asked other children for help to cook each other; (4) Swapped position. **Training core abilities:** Combined multiple capacities.
		Painting emotional pictures	**Aim:** End the class **Operation:** (1) Shown four emotional pictures; (2) Asked children paint pictures, but different colors; (3) Limited colors, asked other children for help to paint each other. **Training core abilities:** Combined multiple capacities.

$p < 0.01$. To control the impact of inhibition control during pretest, we used children's inhibition control change to make further analysis during post-test.

Pre–Post-test Data Analysis

The analysis revealed that the increase in the EF training group was higher than that of no training group 2 months later (see **Table 2**).

Repeated Measures

To investigate whether EF training could enhance children's EC, we performed 2 (group: No-training VS EF training) × 2 (test time: pre VS post) repeated measures ANOVA. The emotional competence was the dependent variable. Results revealed that test time main effect was not significant, $F(1,49) = 0.547$, $p > 0.05$; the group main effect was not significant, $F(1,49) = 2.066$, $p > 0.05$; but the interaction between group and test time was significant, $F(2,49) = 21.10$, $p < 0.001$, $\eta 2 = 0.373$.

Further simple effect analyses showed that EF training group and No-training group were not significant during the pretest, M_{diff} (I–J) = 0.20, $p > 0.05$. But EF training group children performed better than No-training group during post-test, M_{diff}(I–J) = 0.51, $p < 0.001$, $\eta 2 = 0.272$ (see **Figure 1**).

The findings suggested that EF training can improve children's emotional competence.

To illustrate the effect of children's EF on the sub-capacities of children's emotional competence.

First, in order to investigate whether training EF improves children's emotional recognition, we performed 2 (group: No-training VS EF training) × 2 (test time: pre VS post) repeated measures ANOVA. The emotional recognition was the dependent variable. The results revealed test time main effect was not significant, $F(1,49) = 0.291$, $p > 0.05$; the emotional recognition ability of training group was significantly greater than that of No-training group, $F(1,49) = 5.896$, $p < 0.01$, $\eta 2 = 0.144$, and the interaction between group and test time was significant, $F(2,49) = 10.159$, $p < 0.001$, $\eta 2 = 0.222$. Further simple effect analyses showed that EF training group and No-training group were not significant during the pretest, M_{diff}(I–J) = 0.086, $p > 0.05$, but EF training group children performed better than No-training group after the intervention, M_{diff}(I–J) = 1.111, $p < 0.001$, $\eta 2 = 0.343$ (see **Figure 2-I**). The findings suggested that EF training can improve children's emotional recognition. Secondly, to determine whether EF training could promote children's emotional comprehension, we performed 2(group: No-training VS EF training) × 2(test time: pre VS post) repeated measures ANOVA and emotional comprehension was regarded as dependent variable. Results revealed test time the main effect was not significant, $F(1,49) = 0.104$, $p > 0.05$, and group main effect was not significant, $F(1,49) = 0.630$, $p > 0.05$, but the interaction effect between condition and test time was significant, $F(2,49) = 4.389$, $p < 0.05$, $\eta 2 = 0.110$. Further simple effect analyses showed that EF training group and No-training group were not significant during the pretest, M_{diff}(I–J) = 0.210, $p > 0.05$, but EF training children during post-test performed was better than No-training group, M_{diff}(I–J) = 0.721, $P < 0.001$, $\eta 2 = 0.093$ (see **Figure 2-II**). The findings suggested that EF training can improve children's emotional comprehension.

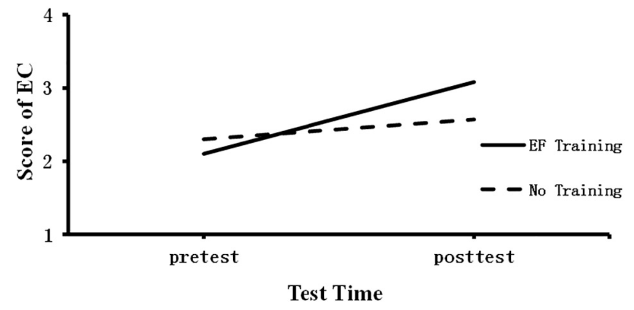

FIGURE 1 | Children's Score of EC during pre–posttest.

TABLE 2 | Description statistics between pretest and post-test.

Variable	No training		EF training	
	Pretest	Post-test	Pretest	Post-test
EC	2.30	2.57	2.10	3.08
E-Reco	6.32	7.84	6.04	10.63
E-Com	2.58	2.63	2.39	2.97
E-Exp	3.97	3.45	3.63	4.14
ER	1.86	2.73	1.52	3.04

*EF, executive function; EC, emotional competence; IC, inhibition control; Flex, cognitive flexibility; WM, working memory; PS, problem solving; E-Reco, emotional recognition; E-Com, emotional comprehension; E-Exp, emotional expression; ER, emotional regulation. *p < 0.05; **p < 0.01; ***p < 0.001.*

Thirdly, to elucidate whether EF training could predict children's emotional expression, we performed 2(group: No-training VS EF training) × 2(test time: pre VS post) repeated measures ANOVA and emotional expression was regarded as the dependent variable. The results revealed that test time main effect was not significant, $F(1,49) = 0.096$,$p > 0.05$. The main effect of group was not significant, $F(1,49) = 0.185$,$p > 0.05$, and the interaction between group and test time was significant, $F(2,49) = 5.170$,$p < 0.01$,$\eta 2 = 0.127$. Further simple effect analyses showed that EF training group and No-training group were not significant during the pretest, M_{diff}(I–J) = 0.131, $p > 0.05$, and EF training group and No-training group were significant during the post-test, M_{diff}(I–J) = 0.410, $p < 0.01$, $\eta 2 = 0.132$ (see **Figure 2-III**). The findings suggested that EF training can improve children's emotional expression.Fourthly, in order to examine whether EF training could promote children's emotional regulation, we performed 2(group: No-training VS EF training) × 2(test time: pre VS post) repeated measures ANOVA. The emotional regulation was the dependent variable. The results revealed that test time main effect was not significant, $F(1,49) = 0.990$, $p > 0.05$. The main effect of group was significant, $F(1,49) = F(1,49) = 3.977$, $p < 0.05$, $\eta 2 = 0.090$, and the interaction between group and test time was significant, $F(2,49) = 3.495$, $p < 0.05$, $\eta 2 = 0.127$. Further simple effect analyses showed that EF training

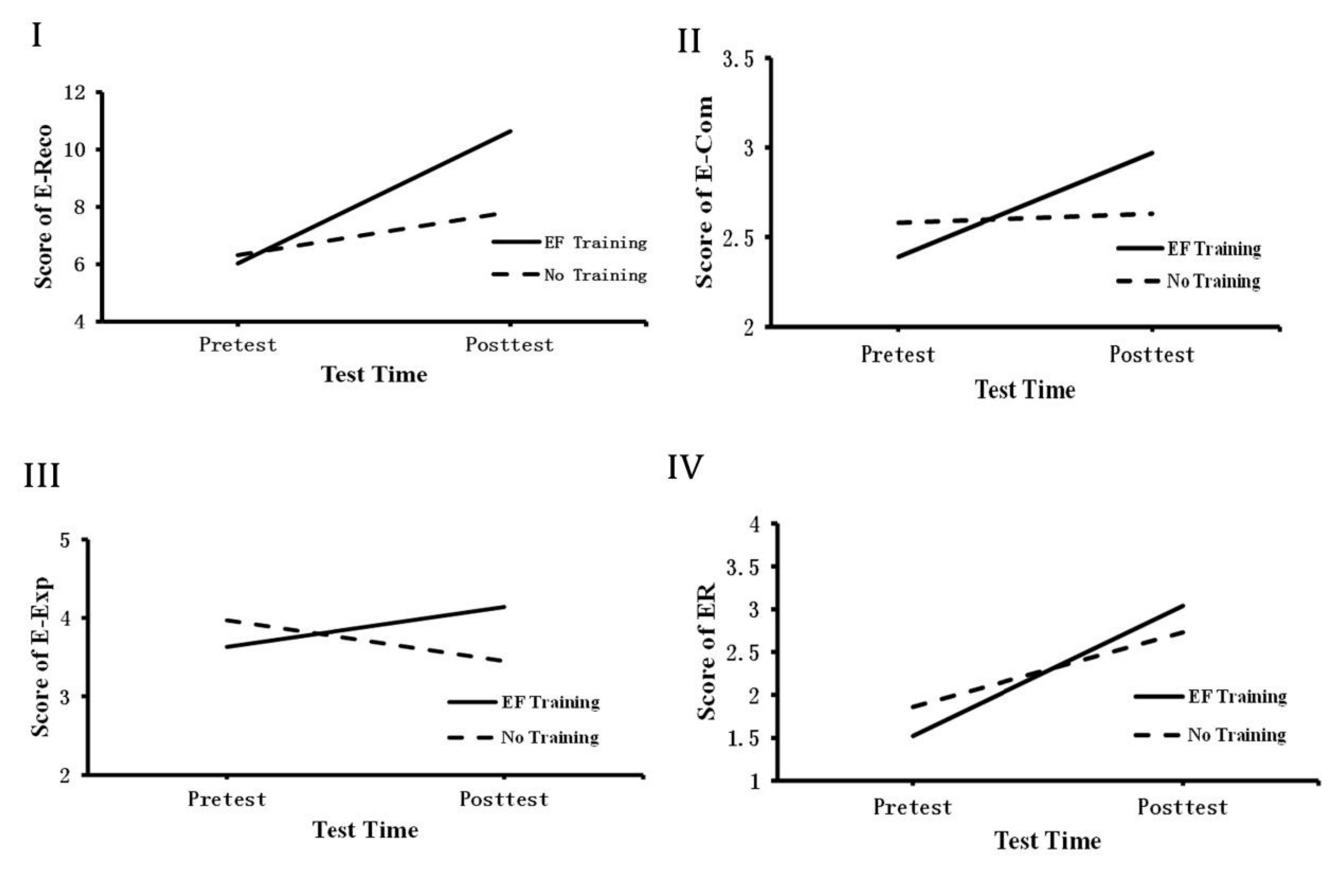

FIGURE 2 | Group by test time interaction on the EC. **(I)** The scores of emotional recognition during pre–posttest; **(II)** the scores of emotional comprehension during pre–posttest; **(III)** the scores of emotional expression during pre–posttest; **(IV)** the scores of emotional regulation during pre–posttest.

group and No-training group were not significant during the pretest, M_{diff}(I–J) = 0.299, $p > 0.05$, and EF training group and No-training group were significant during the post-test, M_{diff}(I–J) = 0.400, $\eta 2 = 0.152$ (see **Figure 2-IV**). The findings suggested that EF training can improve children's emotional regulation.

EF Change Predicted EC Change

To further discern the contributions of children's EC to variance in later EF training effect, correlation and regression analyses were used to achieve this goal. Io order to control the difference of inhibition control during the pretest, we calculated the difference by using post-test minus pretest to represent the development of children's emotional competence. Firstly, we performed correlation between ΔEF and Δ EC.

As shown in **Table 3**, there was a significantly positive correlation between ΔEF and ΔEC, $r = 0.493$, $p < 0.01$; the relationship between ΔIC and ΔEC was a significant, $r = 0.465$, $p < 0.01$; the association between ΔWM and ΔEC was significant, $r = 0.359$, $p < 0.01$, and there was a significant relationship between ΔPS and ΔEC, $r = 0.320$, $p < 0.05$, but $\Delta Flex$ was not significantly associated with ΔEC, $r = -0.007$, $p > 0.05$. Interestingly, there was a significantly positive relationship between ΔIC and ΔER, $r = 0.498$, $p < 0.01$, and the relationship between ΔWM and $\Delta ECom$ was significant, $r = 0.426$ $p < 0.05$. The results suggested that the change of internal sub-capacities (e.g., ΔIC and ΔWM) could impact the change of emotional competence (e.g., ΔER and Δ $ECom$).

To examine EF's contributions to variance in total emotional competence, we performed the hierarchical linear regression analysis. Regression models include the following two models: (1) first model only included inhibition control and (2) the second model included inhibition control and working memory.

The results showed only inhibition control and working memory were significant predictors compared with all four EF measures. The cognitive flexibility and problem solving cannot be successfully entered into the regression models. These results indicated that only inhibition and working memory can positively predict the development of emotional competence. In summary, these results suggested that the training for children's EF can improve the development of emotional competence, and both of them could explain 25.3% variation in children's emotional competence.

To further examine the contributions of the change of inhibition control and working memory to variance in total emotional competence for the No-training group, we also performed the hierarchical linear regression analysis for No-training group. The results showed that children's change in

TABLE 3 | The correlations between the change of EF and EC during pre–posttest.

	ΔEF	ΔEC	ΔIC	ΔFlex	ΔWM	ΔPS	ΔERoc	ΔECom	ΔEExp	ΔER
ΔEF	1									
ΔEC	0.493**	1								
ΔIC	0.716**	0.465**	1							
ΔFlex	0.409**	−0.007	0.096	1						
ΔWM	0.568**	0.395**	0.324*	−0.150	1					
ΔPS	0.73**	0.320*	0.309*	0.149	0.221	1				
ΔERoc	0.266	0.518**	0.348	−0.009	0.091	0.159	1			
ΔECom	0.253	0.606**	−0.047	0.049	0.426*	0.069	0.023	1		
ΔEExp	0.033	0.393*	0.168	0.07	0.044	0.193	0.205	0.062	1	
ΔER	0.418*	0.622**	0.488**	0.230	0.148	0.148	0.418*	0.025	0.102	1

*N = 32. The coefficients of 0.10, 0.30, and 0.50 as small, medium, and large. *p < 0.05; ** < 0.01; *** < 0.001. ΔEF: the change of executive function; ΔIC: the change of inhibition control; ΔFlex: the change of flexibility cognitive; ΔWM: the change of working memory; ΔPS: the change of problem solving; ΔEC: the change of emotional competence; ΔERoc: the change of emotional recognition; ΔECom: the change of emotional comprehension; ΔEExp: the change of emotional expression; ΔER: the change of regulation.*

EF could not predict the change in EC, adjusted $R^2 = 0.061$, $Beta = 0.218$, $t = 1.190$, $p > 0.05$.

DISCUSSION

Consistent with our hypotheses, the results showed that EF training significantly promoted children's EC. In terms of pre-and-post-test differences, the results found that (a) EF training group significantly improved the emotional competence (ECs) compared to those of in No-training group. (b) Children's inhibition control and working memory significantly predicted the development of emotional competence. In our study, we trained children's EF to improve emotional competence. Training effects were found for emotional comprehension and emotional regulation. Our results suggested that the EF training was able to promote children's emotional competence.

EF Training Improved Children's Emotional Competence

Our results demonstrated that our EF training improved emotional competence in preschool-aged children. Components of EF (e.g., inhibition control) plays an important role in children's emotional competence. This provides a new evident effect on children's emotional competence intervention during preschool children. Riggs et al. (2006) found that the better EF performance was always along with the better emotional competence performed. One possible explanation for the finding was that the ability of EF processing needed has some overlapping with emotional competence (Phillips et al., 2003; Chen et al., 2009; Carlson et al., 2013). Traue and Pennebaker (1993) stated that the ability of emotion and inhibition control were significantly correlated. Additionally, children's effortful control can promote the development of emotional competence (Liew, 2012). Moreover, in clinical studies, inhibition control and working memory played crucial roles in emotional tasks, such as fear extinction (Dillon and Pizzagalli, 2008). Hence, the EF training effect may result from that children's EF change was used to regulate their emotional competence change, and children's inhibition control and working memory were the vital factors to the development of emotional competence.

Inhibition Control

These findings revealed that children's inhibition control may explain a great amount of variance of children's emotional competence, especially emotional regulation. Children's self-control ability could influence the ability of emotional regulation (Carlson and Wang, 2007). In our study, children's emotional regulation was improved by EF training (e.g., inhibition control). Previous studies have revealed that regulating emotions required ability of inhibition control at the age of 4 to 6. If participants established the link between inhibition and emotional regulation, it would guide them to give better behavioral responses to emotional situations (Denham et al., 2015; Silkenbeumer et al., 2016). It has also been found that participants with better inhibition control ability exhibited better emotional regulation ability compared to lower inhibition control ones (Nakamichi, 2017). And some researchers suggested that participants' inhibition control could predict their emotional regulation and social competence during preschool (Penela et al., 2015). Our findings were consistent with previous studies. Furthermore, our study provided a new direction of intervening emotional regulation in preschool children by training inhibition control and it would make a contribution to the field from theoretical, methodological, and practically perspective.

Working Memory

We found that working memory could explain the training effects of EF on emotional competence. There is a considerable evidence for the influence of children's emotional competence by their working memory (Baddeley, 2013). Neural evidence had proved that the activity in ventral emotional comprehension processing regions was consistent with activity in brain regions related to working memory among PTSD participants (Morey et al., 2009). Another study stated that working memory may explain the frontal lobe involvement in the task processing of emotional comprehension (Mitchell, 2007). Previous research had stated that children's working memory could influence their emotional comprehension. In this study, the results suggested that training children's working memory may improve their emotional comprehension, which provides a further evidence to the mental mechanism. The development of working memory had a decisive impact on the development of children's comprehension between 5 and 11 year-old children (Morra et al., 2011). And a study stated that individual working memory could predict preschoolers' emotional comprehension performance (Pons et al., 2002; Mutter et al., 2006). In a word, our study can provide new evidence for effectiveness of clinical interventions given that working memory training could improve children's emotional comprehension.

Limitations and Future Directions

Our study has some limitations. First, our sample size limited the use of more complex analytical method to detect effect sizes. Thus, recruiting a larger sample may enable researchers to examine these results in the future study. Further longitudinal studies are needed to assess how long the EF training effect can last. Furthermore, the exact neural mechanism will still be needed to investigate in the future by using electroencephalograms (EEG), Functional near-infrared spectroscopy (fNIRS), and other neuroimaging methodologies. Finally, the self-report on daily performance of research assistants will be also needed to collect to examine how the EF training curriculum influences children's EC as well.

CONCLUSION

Our study has revealed that training EF may improve children's EC. Importantly, children's inhibition control and working memory could make the change of emotional regulation and emotional comprehension more effectively. These findings in our study can make a theoretical and practical contribution to the field of developmental psychology.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author FT.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Ethics Committee of Southwest University. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

QL performed the experiments and wrote the manuscript. PL revised the manuscript. TF and NY provided professional guidance and revised the manuscript.

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Smartphone-Based Answering to School Subject Questions Alters Gait in Young Digital Natives

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Smartphone texting while walking is a very common activity among people of different ages, with the so-called "digital natives" being the category most used to interacting with an electronic device during daily activities, mostly for texting purposes. Previous studies have shown how the concurrency of a smartphone-related task and walking can result in a worsening of stability and an increased risk of injuries for adults; an investigation of whether this effect can be identified also in people of a younger age can improve our understanding of the risks associated with this common activity. In this study, we recruited 29 young adolescents (12 ± 1 years) to test whether walking with a smartphone increases fall and injuries risk, and to quantify this effect. To do so, participants were asked to walk along a walkway, with and without the concurrent writing task on a smartphone; several different parameters linked to stability and risk of fall measures were then calculated from an inertial measurement unit and compared between conditions. Smartphone use determined a reduction of spatio-temporal parameters, including step length (from 0.64 ± 0.08 to 0.55 ± 0.06 m) and gait speed (1.23 ± 0.16 to 0.90 ± 0.16 m/s), and a general worsening of selected indicators of gait stability. This was found to be mostly independent from experience or frequency of use, suggesting that the presence of smartphone activities while walking may determine an increased risk of injury or falls also for a population that grew up being used to this concurrency.

Keywords: smartphone use, texting, adolescents, gait parameters, risk of injury

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INTRODUCTION

In the modern world, the use of a smartphone has become a main characteristic of people's lives (1). In particular, children and teens have grown up with an easy and early access to mobile phones; for this reason, they are part of the group of the so-called "digital natives" (2). For young adolescents, the smartphone is an important tool for communication, education, and entertainment purposes (3); moreover, at that age, the web-based social networks built around a mobile app represent the main connection between peers (4). As a consequence, young adolescents typically spend more than 3 h a day interacting with their smartphone (5), and this practice leads them to get familiar with using it while doing a variety of physical tasks (6). Considering this, the use of the smartphone as a concurrent task during activities of daily living is quite common among young adolescents, regardless of the risk that it can represent; for these subjects, texting is the most frequent activity during walking, given its central importance in social network applications (4).

For young and older adults, it has been demonstrated that the use of a smartphone during everyday walking is increasingly resulting in injuries for pedestrians at all ages (5), in a way similar to the effect of texting and internet navigation while driving (5). While being generally considered as automatic, walking requires attention resources (7) and it is governed by a number of higher cognitive processes (8). The main agreed source of risk associated with smartphone use while walking is identified in its distracting power (9); it has however been demonstrated that most gait parameters linked to stability and to fall risk are also altered in controlled laboratory settings (10), where distractions do not represent the main source of injury risk. Thus, risk may increase also as a consequence of biomechanical alterations.

From a biomechanical point of view, the presence of a secondary task determines posture alterations (11) and a higher risk of fall when walking (12); this is commonly associated with variations in gait patterns, such as specific spatio-temporal and stability parameters of gait (13, 14). Moreover, gait alterations were found to be greater in children and adolescents (15, 16) than in adults (17), suggesting the idea that this effect is the result of different concurring phenomena that cannot be generalized across different age groups.

Among secondary tasks, smartphone use while walking has been increasingly studied (18), given its importance in the everyday life of people of all ages. However, no studies have tried to quantify this effect in a younger population of digital natives yet. In this paper, we recruited a population of digital native young adolescents (11–13 years old) in order to check whether smartphone use during gait has a significant effect on the aforementioned parameters and to quantify these variations. The question that we want to answer with this study is whether, given the rather high experience in texting while walking for this age group, the effects that the concurrent task plays on gait performance are negligible in terms of injury risks.

MATERIALS AND METHODS

Participants

Twenty-nine young adolescents (15 girls and 14 boys, age 12 ± 0.5 years, height 1.56 ± 0.08 m) were recruited from a local secondary school: none of them had special educational needs or certified disabilities. Participants and parents were informed about the procedure, and informed permission of parents was obtained before performing the experiments. The protocol was designed in accordance with the Declaration of Helsinki and approved by the local ethics committee (Applied Electronics section of the Engineering Department).

Procedure

Participants were asked to walk along a 12-m long straight path under two different conditions:

- Baseline: walking at a self-selected speed with no additional concurrent task. No specific instructions added.
- Smartphone: walking while texting messages to the experimenter using an instant messaging app on the smartphone. The concurrent activity involved answering questions sent by the experimenter and taken randomly from a specified list.

The list of questions was defined by the teachers from the Mathematics and English language syllabi of the class the participants were attending. The teachers provided math questions (e.g., "What is the area of a trapeze?," "What is the area of a rectangle?") and translation exercises (e.g., "Translate the following verbs in the English language"). The participants were informed about the fact that they would be asked questions regarding the subjects while walking, but no specific information on the questions was given in advance to them. Each participant received the same number of questions.

Prior to the experiment, participants were asked two questions regarding smartphone expertise and frequency of use: (A) How long have you been using a smartphone? (B) How many hours a day do you use it? They were then shown the path to follow and instructed on the activity to perform. Specifically, for the Smartphone condition, they started to walk just after receiving and reading the first question. All the participants used the same smartphone, and a brief familiarization period was allowed. The order of the two conditions was randomized. While no explicit indication on how to handle the smartphone was given, during the experiments all the participants used a two-handed grip to text while walking.

Instrumentation

A single triaxial accelerometer (Shimmer3, Shimmer Sensing, Dublin, Ireland) was placed on the back of the lumbar zone around L3 (19), through an elastic belt (see **Figure 1**), to acquire linear accelerations along the three main directions (anteroposterior, AP; mediolateral, ML; vertical, VT), in the range ±2 g. Sampling frequency was set at 102.4 samples/s, and data were stored on an on-board SD card. During the experiment, notes were taken to record possible deviations from the defined path or from the activity required to be performed, so as to exclude them from the analysis.

Data Processing and Parameters Extraction

After realignment with global coordinates, accelerometer data were low-pass filtered with a cut-off frequency of 20 Hz (Butterworth, 4th order), and segmented into gait cycles based on the method proposed by McCamley et al. (20); initiation and termination steps were removed to exclude gait cycles affected by the presence of acceleration and deceleration phases. For each gait cycle, the following gait parameters, arranged into two groups, were extracted.

Spatio-Temporal Parameters

- *step length* (m), estimated following the inverted pendulum model (21);
- *step time* (s), the time interval between two successive initial contacts of different feet (22);

FIGURE 1 | Inertial sensor placement for the experimental procedure.

- *stride frequency* (Hz), obtained from the power spectra of the acceleration components (23);
- *gait speed* (m/s), the ratio between step length and step time (23).

Overall, decreases in values of these spatio-temporal parameters have been linked to a diminished progression performance, and have been associated with an increased risk of falls in elderly adults (24). Moreover, normalized versions of step length and gait speed with respect to height were also calculated, to exclude any dependence on the results from height. For all spatio-temporal parameters, values were extracted from each gait cycle, and averaged along the whole trial for each condition.

Gait Stability Indicators

From the normalized autocorrelation function of the accelerometer data, along the three directions, the following gait stability indicators were calculated:

- *step symmetry*, outlining similarity in walking patterns between left and right steps (25);
- *step regularity*, referring to the similarity between successive left (or right) steps (25);
- *stride regularity* indicating the similarity between successive strides (25).

For the vertical and anteroposterior directions, step symmetry is given by the ratio of the first and second amplitude positive peak at time lags different from zero, while for the mediolateral direction, it is given by the ratio of the first negative amplitude peak at time lag different from zero. Step regularity is represented by the value of the first amplitude positive peak (the second one for stride regularity) for the vertical and anteroposterior components, and by the value of the first negative peak for the mediolateral one (the first positive amplitude peak for the stride regularity).

In general terms, it is expected to have values of symmetry and regularity closer to 1 in normal conditions; values away from 1 may indicate a decrease in gait stability (26, 27).

Then, to use a gait speed-independent measure of walking smoothness, from each component of the acceleration vector the following indicator was calculated:

- *spectral arc length* (SPARC), as a measure of walking smoothness (28).

SPARC quantifies smoothness by computing the negative value of the arc length of the normalized Fourier spectrum of the modulus of the acceleration signal, in the frequency range of the movement. The maximum frequency for SPARC calculation has been defined as the frequency above which the normalized spectrum remains lower than 0.01 (29). A smoother gait pattern results in a higher (i.e., closer to zero) value of SPARC. In normal gait patterns, SPARC results were higher than in the presence of pathologies (29).

For all the gait stability indicators, values were calculated by considering the whole trial for each condition.

Smartphone Use Habit Sub-grouping

All participants declared they used the smartphone every day. To assess whether frequency of use or smartphone expertise were factors in possible modifications on gait behavior, the analysis on both spatio-temporal gait parameters and gait stability indicators was done by splitting participants into sub-groups considering reported years of use (question A) and frequency of use (question B). In particular:

- A1 subjects have been using the smartphone for 1–2.5 years
- A2 subjects have been using the smartphone for more than 2.5 years
- B1 subjects (moderate users) regularly use the smartphone up to 2 h/day
- B2 subjects (frequent users) regularly use the smartphone for more than 2 h/day.

TABLE 1 | Descriptive statistics for the spatio-temporal parameters (group mean ± standard deviation), and results of the corresponding statistical analysis.

Spatio-temporal parameters	Baseline	Smartphone	*p*-value
Step length (m)	0.64 ± 0.08	0.55 ± 0.06	<0.001
Normalized step length	0.41 ± 0.04	0.35 ± 0.04	<0.001
Step time (s)	0.53 ± 0.03	0.61 ± 0.06	<0.001
Stride frequency (Hz)	0.96 ± 0.06	0.83 ± 0.09	<0.001
Gait speed (m/s)	1.23 ± 0.16	0.90 ± 0.16	<0.001
Normalized gait speed (s^{-1})	0.79 ± 0.10	0.58 ± 0.11	<0.001

Statistical Analysis

Descriptive statistics included measures of central tendency and dispersion, and it was calculated for each parameter under both conditions. The distribution of data for each parameter was tested for normality by group using Lilliefors test. To check for the presence of an effect during texting, a one-way ANOVA (with condition as factor) was performed on the gait parameters that showed normality. If normality was rejected at the chosen significance level, a Kruskal-Wallis test was used. To study the effect that the years and frequency of smartphone use could have on the gait parameters, two 2-way ANOVA tests were applied considering the condition (baseline/smartphone) and either smartphone expertise (A1 or A2) or frequency of use (B1 or B2) as factors. Tests significance was set at 0.05.

RESULTS

Mean, standard deviation, and *p*-values for all extracted parameters are shown in **Tables 1**, **2**.

Spatio-Temporal Parameters

A significant effect driven by the use of the smartphone appeared for all spatio-temporal parameters. In particular, the use of smartphone during walking increased the step time, and decreased step length (and its normalized version), stride frequency, and both versions of gait speed. The numerical results are reported in **Table 1**.

Gait Stability Indicators

When using the smartphone, the statistical analysis on symmetry parameters yielded a significant increase of the step symmetry components along the anteroposterior and vertical directions, while a significant decrease appeared for the mediolateral direction. Most gait regularity parameters decreased significantly in the smartphone use condition, with only the step regularity along the mediolateral direction being unaffected. Gait smoothness in the vertical direction was not

TABLE 2 | Descriptive statistics for the gait stability indicators (group mean ± standard deviation), and results of the corresponding statistical analysis (n.s. for *p*-value > 0.05).

Gait stability indicators	Baseline	Smartphone	*p*-value
Step symmetry AP	1.00 ± 0.11	1.11 ± 0.11	< 0.001
Step symmetry VT	0.99 ± 0.11	1.13 ± 0.27	0.003
Step symmetry ML	−0.91 ± 0.26	−1.07 ± 0.31	0.01
Step regularity AP	0.78 ± 0.09	0.72 ± 0.11	< 0.001
Step regularity VT	0.80 ± 0.10	0.67 ± 0.18	< 0.001
Step regularity ML	−0.49 ± 0.12	−0.48 ± 0.14	n.s.
Stride regularity AP	0.79 ± 0.09	0.66 ± 0.14	< 0.001
Stride regularity VT	0.82 ± 0.11	0.62 ± 0.20	< 0.001
Stride regularity ML	0.56 ± 0.14	0.47 ± 0.13	0.01
SPARC AP	−4.22 ± 0.08	−4.29 ± 0.09	0.002
SPARC VT	−4.26 ± 0.12	−4.26 ± 0.07	n.s.
SPARC ML	−4.27 ± 0.07	−4.34 ± 0.07	< 0.001

affected by the presence of the concurrent task, which, in turn, led to significantly lower smoothness in both components of the transverse plane. The corresponding numerical results are reported in **Table 2**.

Effect of Smartphone Use Habit on Gait Parameters and Indicators

The questionnaire answers showed that 10 participants have been using a smartphone up to 2.5 years (sub-group A1), while 19 for more than 2.5 years (sub-group A2); 14 individuals reported using the device for up to 2 h/day (sub-group B1), the remaining 15 declared regular use of more than 2 h/day (sub-group B2).

The statistical analysis showed no significant modifications of any spatio-temporal parameter based on either sub-group splitting (for both A and B). A significant modification of some gait stability indicators based on frequency of use (sub-group B) appeared. In particular, step regularity along antero-posterior and vertical direction, and stride regularity along the antero-posterior direction were all significantly higher for frequent users, as compared to moderate users; likewise, SPARC resulted lower for frequent users (see **Table 3**). No significant effect from years of use appeared. Both spatio-temporal gait parameters and gait stability indicators were dependent from condition in both sub-groups, while no interaction between condition and either frequency or years of use was found.

DISCUSSION

This study aimed at determining the influence of smartphone use while walking on a variety of gait parameters recorded on a population sample of young adolescents. Technology is a constant part of their everyday life and their approach to smartphone use started at a young age (30). Despite their familiarity and expertise with the use of such devices, we were able to confirm that smartphone use during walking determined a variation of multiple gait parameters, including measures of gait symmetry, regularity, and smoothness.

Spatio-Temporal Parameters

When using smartphone while walking, step time increased and step length decreased, which is indicative of a slower walk. This is similar to results obtained in multiple dual-task studies on gait involving children of different age ranges (31–33). In the presence of a concurrent task, young individuals tend to walk slower and with smaller steps, as do adults. In terms of effect size, we could not draw a direct comparison with published research on the elderly (34) and young adults (35), given the specific nature of the additive concurrent task employed in this study; however, the relative reduction we observed on gait speed corresponds to the upper limits of the reported range of reduction in adults (34), thus suggesting that the effect on the studied age group is relevant. Regarding spatio-temporal parameters, we could not exclude that modifications of gait speed and step length may also depend on the altered posture caused by handling the phone, as disentangling purely postural effects from cognitive ones would have needed a "mock" condition where subjects were requested to handle the phone without answering questions. However, the amount of changes caused by maintaining a fixed elbow has been quantified in around 0.03–0.05 m/s (36), thus well below the overall effect observed in this study. These findings suggest that the nature of these modifications is mostly determined by the attention share of the secondary task.

A significant decrease also appeared for the normalized version of step length, thus highlighting that step reduction is independent from height.

We could speculate that such an amount of reduction might be associated with the adolescents prioritizing texting over motor function, and that the significant alteration of all spatio-temporal parameters might be linked to a decrease in attention to the surrounding environment.

Gait Stability Indicators

All gait stability indicators showed a worsening caused by texting: participants showed a less symmetrical, less regular, and less smooth gait. In particular, gait symmetry in the sagittal plane was detrimentally affected; while we could not exclude a higher involvement of the dominant hand when texting on the smartphone, we were positive of the absence of visible postural trunk asymmetries. In this, we were supported by the observation that all the involved individuals used the smartphone in a 2-handed holding configuration.

The observed decrease of regularity parameters mainly in the sagittal plane is in line with what has been found in a variety of dual-task studies involving adults and the elderly (12, 35), and it has been directly linked to increased task-related motor and cognitive demand, as confirmed by a higher central involvement when texting while walking (37). The hypothesis of a similar involvement also in the observed adolescent sample might explain our findings on regularity parameters.

Variations of smoothness in both components of the transverse plane may be linked to a less adaptive walking pattern, since both SPARC indicators showed a decrease when texting. Even if the effect coming from the presence of the concurrent task on SPARC is rather low, we outline here that this metric has been reported to be found in robust to walking speed

TABLE 3 | Descriptive statistic (group mean ± standard deviation) and *p*-value for the gait stability indicators influenced by condition and frequency of use (n.s. denotes *p*-value > 0.05).

Gait stability indicators		Moderate users	Frequent users	Main effect (condition)	Main effect (frequency of use)	Interaction (condition × frequency of use)
Step Regularity AP	Baseline	0.74 ± 0.09	0.82 ± 0.05	$p < 0.01$	$p = 0.007$	n.s.
	Smartphone	0.67 ± 0.10	0.76 ± 0.09			
Step Regularity VT	Baseline	0.75 ± 0.08	0.85 ± 0.08	$p < 0.001$	$p = 0.04$	n.s.
	Smartphone	0.62 ± 0.19	0.71 ± 0.14			
Stride Regularity AP	Baseline	0.77 ± 0.08	0.80 ± 0.09	$p < 0.001$	$p = 0.04$	n.s.
	Smartphone	0.61 ± 0.15	0.71 ± 0.11			
SPARC AP	Baseline	−4.21 ± 0.08	−4.24 ± 0.08	$p < 0.001$	$p = 0.008$	n.s.
	Smartphone	−4.25 ± 0.09	−4.34 ± 0.06			

variations (28, 29), and it can thus accurately capture differences in smoothness that are not the result of step time variations. The decrease of these measures has been linked to less steady walking patterns and it has been hypothesized as a predictor for fall risk in people with Parkinson's disease (29); changes in smoothness were interpreted as caused by the competition for resources between cognition and gait (38) and for the reduced visual fixation time at the travel path (39).

Smartphone Use Habit and Implications for Fall and Injury Risks

We observed that being a digital native does not protect from risks identified in older populations when texting while walking, and this is aligned with observed modifications on selected gait parameters in a similar population (40). Despite the different level of familiarity with smartphone use (39), we found that the effect of this concurrent task for digital natives resembles the one reported in the literature on samples of young adults. We also observed that no difference appeared on spatio-temporal parameters between more familiar users and less familiar ones, and this result may confirm the hypothesis of an experience-independent effect of the secondary task on gait. However, we found some elements of difference between the frequent users and the others, i.e., frequent users displayed higher regularity, at the expense of a reduced smoothness. We do not have a clear explanation for these results, but it may be speculated that frequent smartphone users have a tendency to base on rhythmicity when walking, with a reduced emphasis on stability indicators, such as smoothness. We stress here that the reduced statistical power of the analysis, when performed on each sub-group, prevents us from formulating robust interpretations on this.

One of the main factors for observed changes of gait parameters in adults has been hypothesized in the tendency to prioritize texting over walking (41). While we could not directly apply this hypothesis to the observed young adolescent population sample as we did not collect error data on the concurrent task, one possible explanation would be the following: while digital natives may be more used to texting while walking, they may not be efficient enough in governing the concurrency between the activities, according to the reported observation that they do not have a higher ability to multitask than digital new-comers, who are a generation of people that acquired familiarity with a smartphone as adults (2). The ability to multi-task effectively has in fact not been directly linked to the frequency of engagement in multiple tasks simultaneously (2). As a result, young adolescents too may prioritize texting over walking, because they are very proficient in the smartphone use. Since we did not directly collect data on the number of text errors made, we could not verify this hypothesis. At the same time, we could not exclude that another effect may come into play: the agreed overconfidence displayed by young adolescents in a variety of tasks, as compared to adults (42). Being involved in multiple tasks, they may experience high interference on gait, as their ability to execute both functions may be lower than the self-perceived one. It may be interesting to verify if this overconfidence phenomena exacerbates in the presence of elements of disturbance to gait (39). We could not exclude that, in this process, a possible role may be played by the development of motor and cognitive functions being non-complete at this age (43), also in terms of the ability to govern the attentional resources required to control gait (15).

Regarding gait parameters as possible predictors for the risk of injury and fall, it has been shown that most parameters of gait actually change secondary to the main observed change, the decrease of gait speed (44, 45). While we could not exclude that this may be also the case for many observed parameters in our study, the presence of modifications on a substantially velocity-independent parameter of smoothness, i.e., SPARC, comforted us on the validity of the findings. Other measures of smoothness were linked to an increased risk of falls in elderly adults (46). Even if, to our knowledge, a thorough test of the link between SPARC values and the risk of fall is still missing from the literature, the robustness of SPARC with respect to gait speed variations, and the presence of an effect coming from a secondary task on this parameter for the studied population, could call for new studies on this topic.

Walking behavior while using a smartphone is altered in young adolescents. Despite the familiarity of this age group with the everyday use of such devices, the concurrent use of smartphones during gait determines a general worsening of those parameters that are associated with gait performance and stability; as a matter of fact, we observed a general decline of gait speed to values that are lower than 1.1 m/s, a value which is suggested as a threshold of safety in crossing roads for pedestrians (47, 48). Considering this, we can conclude that this kind of concurrent task on walking in this population might lead to biomechanical alterations and decreased stability; in addition, the non-complete motor control development may amplify the effect of a different cognitive load while walking, increasing all the risks associated with smartphone use during daily life.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Applied Electronics section, Department of Engineering, ID 02/18. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

The study was designed by CC, MS, and SC. Recruitment and data collection was run by CC and SR. Data processing and statistical analyses was conducted by CC, CD'A, and SR. The manuscript was written by all authors. CC drafted the Material and Methods and Results sections. MS, SC, CD'A, and SR drafted the Introduction and the Discussion. CD'A led the additional analysis on subgroups and the revision to the original submission. The final version was approved by all authors.

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Developing and Testing the Reliability and Validity of the Brief Haze Weather Health Protection Behavior Assessment Scale-Adolescent Version (BHWHPBAS-AV)

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Objectives: To develop a Brief Haze Weather Health Protection Behavior Assessment Scale-Adolescent Version (BHWHPBAS-AV).

Methods: Considering primary prevention, secondary prevention and tertiary prevention as a theoretical basis, researchers developed a Brief Haze Weather Health Protection Behavior Assessment Scale-Adolescent Version-**I(**BHWHPBAS-AV-**I)**. After performing 6 reviews by related experts, and after conducting six adolescent tests for BHWHPBAS-AV-**I**, researchers developed an updated BHWHPBAS-AV-II. Out of the 20 districts in Baoding, two districts were randomly selected; moreover, two middle schools from these two districts were also randomly selected. Considering one class as a unit, researchers subsequently randomly selected 22 classes by using stratified sampling. In the end, 1,025 valid questionnaires were used as part of the study. At which point, researchers investigated the validity and reliability of the scale and obtained the final scale (BHWHPBAS-AV).

Results: BHWHPBAS-AV Cronbach's $\alpha = 0.878$, content validity = 0.948, and factor cumulative contribution rate = 54.058% using exploratory factor analysis. By confirmatory factor analysis, Chi square value (χ^2) = 271.791, degrees of freedom (df) = 94, Chi square value/degrees of freedom (χ^2/df) = 2.891, root-mean-square error of approximation (RMSEA) = 0.051, normed fit index (NFI) = 0.930, incremental fit index (IFI) = 0.953, goodness of fit index (GFI) = 0.955, Tueker-Lewis index (TLI) = 0.940, comparative fit index (CFI) = 0.953. BHWHPBAS-AV was composed of 16 items as well as 3 dimensions.

Conclusions: A BHWHPBAS-AV scale that has an acceptable reliability and validity can be applied to assess adolescent haze weather health protection behavior, and can also help school teachers, as well as medical staff working in community health care institutions, to perform targeted behavioral interventions and deliver health education programs to adolescents.

Keywords: adolescent, haze, health protection behavior, reliability, validity

INTRODUCTION

Air pollution has become a serious global problem, influencing the health level of millions of people around the world (1). In China, many serious public health problems are directly a result of air pollution. In 2012, the Chinese Government introduced its National Plan directed toward Air Pollution Control. The plan outlined stringent air pollution prevention goals and control measures. The Chinese Government promulgated the first National Action Plan focusing on Air Pollution Prevention and Control (2013–2017) in September 2013, which hoped to lead to a significant improvement in national air quality by 2017 (2). As an important manifestation of air pollution, haze weather may cause serious short-term harm, as well as have a long-term negative impact on human health. Subsequently, this may lead to further issues in public health, especially regarding the growth and development of children and adolescents (3–8). Owing to the fact that the adolescent period of life is a critical stage in growth and development, adolescent physiological functions remain immature and are thus susceptible to the effects of haze (1, 9–13). The particulate matter that contributes to haze weather can directly enter the respiratory system and subsequently adhere to the respiratory mucosa or deposit in the alveoli, which results in damage to the respiratory tract and alveolar epithelial cells. This may reduce the lung capacity of adolescents and cause asthma, rhinitis, bronchitis, pneumonia, and lung cancer (9, 11, 14). In addition, the pollutant in haze weather can weakens the immunity of adolescents, triggers allergic reactions, damages the circulatory and nervous system, and has the potential to give rise to metabolic diseases (15, 16). Compared with adults, adolescents have a lesser understanding of the harm that haze weather can cause; moreover, a lower proportion of the youth tend to adopt health protection behaviors in haze weather (16). Undoubtedly, it is critical for the safety of adolescents that we carry out social awareness campaigns to educate the public, especially adolescents, about haze weather, and how to protect oneself against its hazards (17). Implementing protective behaviors against haze weather can help prevent youngsters from suffering from haze weather related health conditions and can promote adolescent normal growth and development. Besides that, an increased social awareness will surely reduce the burden haze weather has on families, schools, the government, and society in general (18).

At present, the majority of research to do with haze weather is focused on the experimental research field, for example, its harmful effects on tissues and organs (9, 11, 12). However, the findings and implications from these experimental studies have not been widely related to the health protection behavior of adolescents (16). Therefore, it is important to develop a haze weather health protection behavior assessment scale for adolescents, in order to better encourage them to adopt protective health behaviors against haze weather. Deguen carried out a research in eight French cities with the aim of developing an air quality perception scale. Deguen's study indicated that the scale has good validity and reliability and can be used to understand the community residents' perceptions of air quality (19). Ye developed a questionnaire that explored the level of environmental health literacy, attitude, and habits of Beijing-Tianjin-Hebei region residents, surrounding the issue of haze weather (17). In Iran, Mirzaei-Alavijeh developed a self-care behavior related to air pollution protection questionnaire. The questionnaire is a promising instrument that assesses self-care behavior related to air pollution protection from the perspective of college students (20). However, currently, there is no haze weather health protection behavior assessment scale for adolescents. Therefore, our research aims to firstly develop a Brief Haze Weather Health Protection Behavior Assessment Scale-Adolescent Version (BHWHPBAS-AV), and secondly to test its validity and reliability. Wu pointed out that the conceptual framework of a scale should be based on previous theoretical literature or some exploratory empirical researches (21). Zhan pointed out that prevention strategies can be divided into three stages, which include primary prevention, secondary prevention and tertiary prevention (22). In our study, using these three stages of prevention as the theoretical basis, and by investigating related literature references (22–24), research team developed the Brief Haze Weather Health Protection Behavior Assessment Scale-Adolescent Version. BHWHPBAS-AV will help teachers in schools, as well as medical staff in health care institutions, to carry out health education and targeted behavioral interventions, for adolescents who are susceptible to the effects of haze.

METHODS

Development of BHWHPBAS-AV-I

Considering primary, secondary and tertiary prevention as the theoretical foundation (22–24), in addition to investigating extensive literature references, researchers developed an initial scale (Brief Haze Weather Health Protection Behavior Assessment Scale-Adolescent Version-I, BHWHPBAS-AV-I). The initial scale included 20 items and 4 dimensions. Researchers named the four dimensions as follows: Dimension 1, the way to obtain relevant knowledge before haze weather; Dimension 2, the source of relevant knowledge before haze weather; Dimension 3, self-protection in haze weather; Dimension 4, self-adjustment after haze weather. These items of BHWHPBAS-AV-I were presented in way that was simple and easy to understand so that the participants were able to comprehend the meaning of each item with ease (25, 26).

Development of BHWHPBAS-AV-II

Six experts (two clinical doctors, two clinical nurses and two health care experts) were invited to assess the scale content validity. The evaluation standard of content validity ranged from 1 (not related) to 3 (strongly related). Based on the expert reviews, four items from the BHWHPBAS-AV-**I** were removed,

Abbreviations: BHWHPBAS-AV, Brief Haze Weather Health Protection Behavior Assessment Scale-Adolescent Version; CVI, Content validity index; MIIC, Mean inter-item correlation coefficient; EFA, Exploratory factor analysis; PCA, Principal component analysis; KMO, Kaiser-Meyer-Olkin; CFA, Confirmatory factor analysis; χ^2, Chi square value; df, Degrees of freedom; χ^2/df, Chi square value/degrees of freedom; RMSEA, Root-mean-square error of approximation; GFI, Goodness of fit index; NFI, Normed fit index; IFI, Incremental fit index; TLI, Tueker-Lewis index; CFI, Comparative fit index.

which resulted in an updated scale (BHWHPBAS-AV-II). As part of the BHWHPBAS-AV-II, 16 items and 4 dimensions were included. The names of each of the dimensions remained unchanged. BHWHPBAS-AV-II used the Likert 5-point method (1 = disagree; 2 = agree a small part; 3 = moderately agree; 4 = agree most; 5 = completely agree). The total score of the scale was determined by the sum of items' scores. A higher total score suggested better adolescent haze weather health protection behavior. Subsequently, the research team asked 6 adolescents to complete BHWHPBAS-AV-II so that researchers could test and possibly improve the wording and comprehension of statement expressions. Every item in the BHWHPBAS-AV-II was presented in plain and simple to understand language so that the adolescent participants could easily comprehend the meaning of the items (25, 26).

Development of the Final BHWHPBAS-AV and Large Sample Test

From June 2015 to April 2016, in Baoding, China, the research team randomly selected two districts from the twenty districts of Baoding. Subsequently, researchers randomly selected two middle schools from these two districts. Considering a class as a unit, and by conducting stratified sampling, the research team randomly selected five first-grade classes, five second-grade classes, and five third-grade classes from the one middle school (a total of 750 adolescents and 50 adolescents per class). In addition, three first-grade classes, two second-grade classes, and two third-grade classes were selected from the second middle school (a total of 350 adolescents and 50 adolescents per class). In total, this study involved 1100 adolescents. Inclusion criteria: (1) Adolescents have a satisfactory capacity to comprehend, no intellectual disabilities and no reading disabilities; (2) Participants volunteered for the study; (3) Participants do not suffer from any organic brain disease, mental condition or other serious health problems. Sample size: Both exploratory factor analysis and confirmatory factor analysis were appropriately used when the hypothesized measurement model was evaluated. The sample size should be at least 10-15 individuals per item for the factor analysis (27). If the sample size was more than 20 individuals per item for the factor analysis, the results of factor analysis would be more stable and more reliable (28). Since this scale contained 16 items, the sample size of the large sample test should be more than 320. However, to produce more stable reliable results, after comprehensively considering the feasibility of the study, we appropriately increased the number of samples. Ultimately, the research team distributed 1,100 questionnaires. Twenty-one of the participants did not complete demographic characteristic questionnaires or scales, and 54 participants did not complete the scale. Thus, these 75 participants were excluded. Ultimately, 1,025 valid questionnaires were returned. Valid recovery rate = 1,025 valid questionnaires ÷1,100 total questionnaires = 93.18%. Therefore, the valid recovery rate was 93.18%. After analyzing data from 1,025 valid questionnaires, we tested the validity and reliability of the scale and developed the final version of the scale (BHWHPBAS-AV). The study participant flow diagram is shown in **Figure 1**.

Ethical Consideration and Survey Method

The study was approved by the Health and Family Planning Commission of Hebei Province (NO.20150072). The study was also approved by the Medical Ethics Committee of Hebei University. This study was carried out according to the standards set by the Declaration of Helsinki. Researchers explained the purpose of research to the middle school teaching management departments, parents/guardians of the minors, as well as the adolescents themselves. After receiving consent from the school coordinators, parents/guardians of the minors and the adolescents themselves, the research team demonstrated to the participants how to answer the questionnaires. The questionnaires used standardized language and unified instruction.

Statistical Analysis

In order to complete a data consistency check, the research team used the Epidata 3.1 software to input the data into the computer twice. The data were analyzed using the AMOS 17.0 software and SPSS 18.0 software. The research team used descriptive statistics (frequency/percentages or medians/interquartile ranges) to explore adolescent demographic characteristics. The following list represents the methods we used for measuring the validity and reliability of scale: ①The exploratory factor analysis (EFA) and the confirmatory factor analysis (CFA) were used to assess construct validity; ②Content validity index (CVI) was applied to assess content validity of the scale; ③Mean inter-item correlation coefficient (MIIC) and Cronbach's α coefficient were used to evaluate the reliability of the scale. This study set the level of significance at $p < 0.05$. The following criteria were used for the retention of factors: ①Eigenvalues > 1; ②EFA scree plot; ③Items equal to, or >2 being retained; and ④The factor loadings > 0.500 (29). The research team considered the model fit of scale structure as acceptable if root-mean-square error of approximation (RMSEA) <0.08, Chi square value/degrees of freedom (χ^2/df) <3, normed fit index (NFI) >0.90, goodness of fit index (GFI) >0.90, Tueker-Lewis index (TLI) >0.90, incremental fit index (IFI) >0.90 and comparative fit index (CFI) >0.90 (26, 29, 30). The factor loading cut-off point for CFA was 0.50 (28).

RESULTS

Characteristics Pertaining to the Large Sample of Adolescents

A total of 1,100 questionnaires were distributed to the study participants, and 1,025 satisfactory questionnaires were returned. The valid recovery rate of the questionnaires was 93.18%. Subjects included 515 men (50.2%) and 510 women (49.8%) from urban (66%) and rural areas (34%). The age of adolescents was 14 (13, 16) years old, medians and interquartile ranges (IQR). Participant demographics included Han race (96.6%) and minority race (3.4%). Adolescents were categorized into three groups according to monthly expenses: <300 yuan, 300–599 yuan and ≥600 yuan, accounting for 33.4, 52.4, and 14.2%, respectively. Adolescents were also classified into

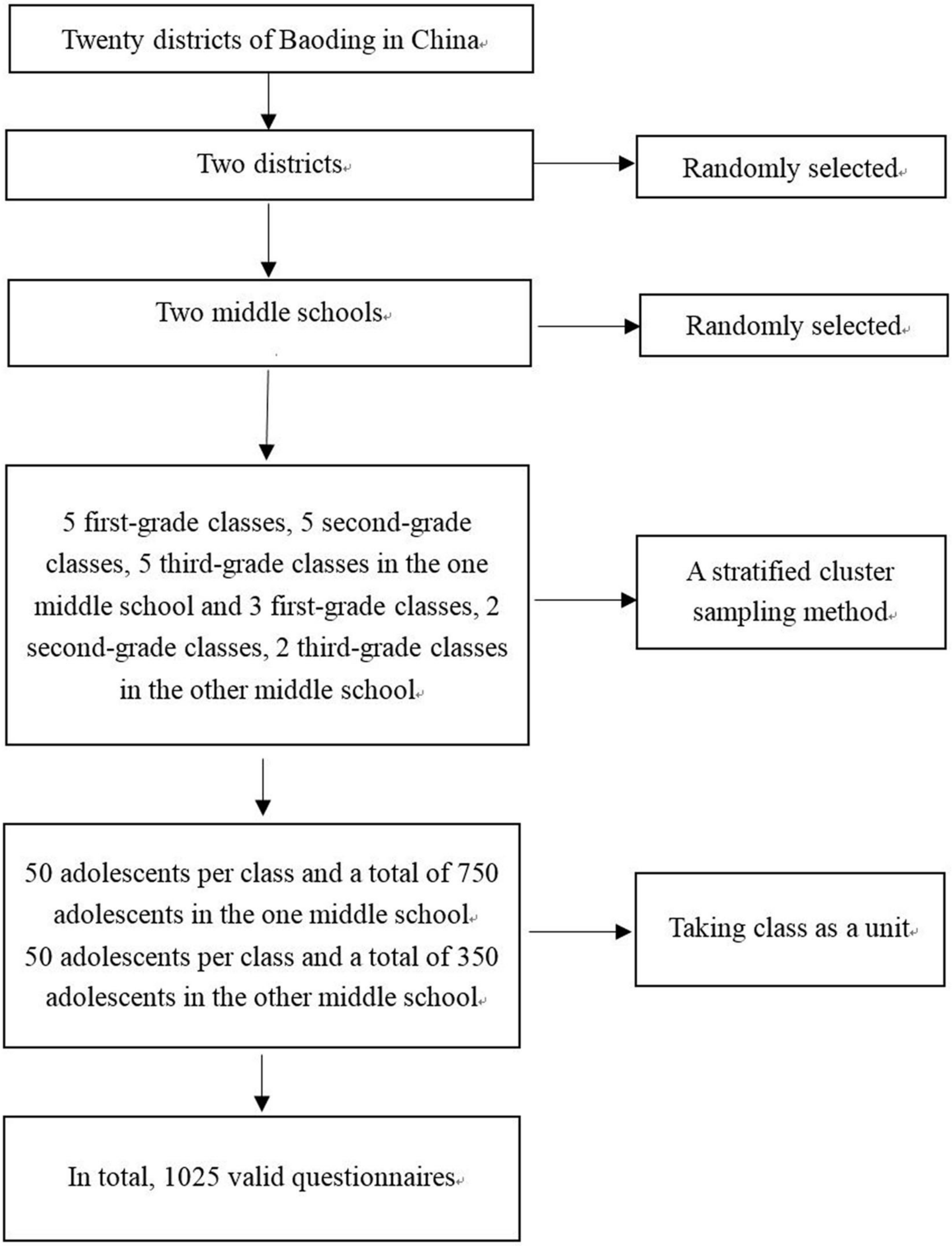

FIGURE 1 | Study participant flow diagram.

three groups according to the method of medical insurance they used, including urban medical insurance, new rural cooperative medical system and self-paying, accounting for 58.0% 29.9 and 12.2% respectively. The data characteristics of the large sample of adolescents are shown in **Table 1** in detail.

Analysis of Reliability and Validity

Construct Validity

Exploratory factor analysis (EFA)

The research team analyzed the BHWHPBAS-AV-II of 1,025 subjects. Principal component analysis (PCA) and maximum variance orthogonal rotation were applied to conduct EFA

TABLE 1 | The characteristics data of the large sample adolescents.

Characteristics	Frequency/Medians	Percentage (%)/ Interquartile ranges, (IQR)
Gender		
Male	515	50.2
Female	510	49.8
Age	14	13, 16
Race		
Han	990	96.6
Minority	35	3.4
Monthly expenses (yuan)		
<300	342	33.4
300∼	537	52.4
600∼	146	14.2
Do you have a religious faith		
No	952	92.9
Yes	73	7.1
Place of residence		
Urban area	677	66.0
Rural area	348	34.0
Way of medical insurance		
Urban medical insurance	594	58.0
New rural cooperative medical system	306	29.9
Self-paying	125	12.2
Do you live with your family		
Yes	958	93.5
No	67	6.5

The characteristics data of the large sample were presented as frequency (percentage) or medians (interquartile ranges, IQR).

of BHWHPBAS-AV-II. The results revealed that the Bartlett sphericity test value was 5668.801 (df = 120, $p < 0.001$) and the Kaiser-Meyer-Olkin value (KMO) was 0.895. The results indicated that the scale data were suitable for factor analysis. Factor extraction was conducted under a condition of undefined factor number. Three factors (Eigenvalue >1) were extracted and the cumulative variance contribution rate (%) was 54.058%. The scree plot of BHWHPBAS-AV-IIfactor analysis indicated an inflection point between the 3rd factor and the 4th factor. The scree plot of exploratory factor analysis also revealed that the 3-factor structure was suitable (**Figure 2**). After performing the aforementioned analyses, the final version of the scale (BHWHPBAS-AV) included three factors and 16 items. Researchers renamed the final three factors: Factor 1, obtain relevant knowledge before haze weather (7 items); Factor 2, self-protection in haze weather (four items); Factor 3, self-adjustment after haze weather (five items) (**Table 2**). The detailed content of Brief Haze Weather Health Protection Behavior Assessment Scale-Adolescent Version (BHWHPBAS-AV) is shown in **Table 3**.

Confirmatory factor analysis (CFA)

In order to confirm the best dimensional structures of BHWHPBAS-AV, using AMOS17.0 software, the research team randomly selected 70% of samples (718 subjects) and applied the maximum likelihood method to conduct the CFA of 16-item, and 3-factor structures of BHWHPBAS-AV. The Chi square value (χ^2) was 271.791, degrees of freedom (df) was 94, Chi square value/degree of freedom (χ^2/df) was 2.891, root-mean-square error of approximation (RMSEA) was 0.051, goodness of fit index (GFI) was 0.955, normed fit index (NFI) was 0.930, incremental fit index (IFI) was 0.953, Tueker-Lewis index (TLI) was 0.940, comparative fit index (CFI) was 0.953 (**Table 4**). The standard path and parameter estimation of CFA is shown in **Figure 3**.

Internal Correlation Test

Among the factors of BHWHPBAS-AV, the correlation coefficients ranged from 0.458 to 0.547 ($p < 0.01$). Correlation coefficients between all factors and the whole scale of BHWHPBAS-AV ranged from 0.798 to 0.812 ($p < 0.01$) (**Table 5**).

Content Validity

According to the findings obtained from six expert reviews, the content validity index (CVI) of scale was 0.948. The CVI of every item ranged from 0.667 to 1.00. After improving the item statement expression and the item wording, six participants reported that they could clearly understand the meanings of all items without any difficulty.

Reliability

The BHWHPBAS-AV mean inter-item correlation coefficient value (MIIC) was 0.312. The MIIC values for all factors ranged from 0.368 to 0.498. The BHWHPBAS-AV Cronbach's α coefficient value was 0.878, and the Cronbach's α coefficients of all factors ranged from 0.744 to 0.837 (**Table 6**).

DISCUSSION

Nowadays, medical systems globally, are gradually changed from the old health care model, which regarded medical staff as leading decision makers in patient care, to a newer and more patient centralized health care model, which involves both medical staff and individuals in the decision making process of a patient's health management (31, 32). Therefore, by significantly improving adolescent health protection ability against haze weather, and by fully mobilizing adolescents' enthusiasm of health protection, the detrimental health impact caused by haze weather may be significantly reduced. The current guidelines, which China's medical system adopts, predominantly focus on the diagnosis, treatment, and care of specific diseases. However, it does not places enough importance on investing in health education of families, schools, and communities. Adolescent knowledge regarding the harmful impact of haze weather is inadequate, and their health protection behaviors pertaining to this issue need to be further improved. The BHWHPBAS-AV developed in our research enables teachers at schools, as well as medical staff in community health care institutions, to

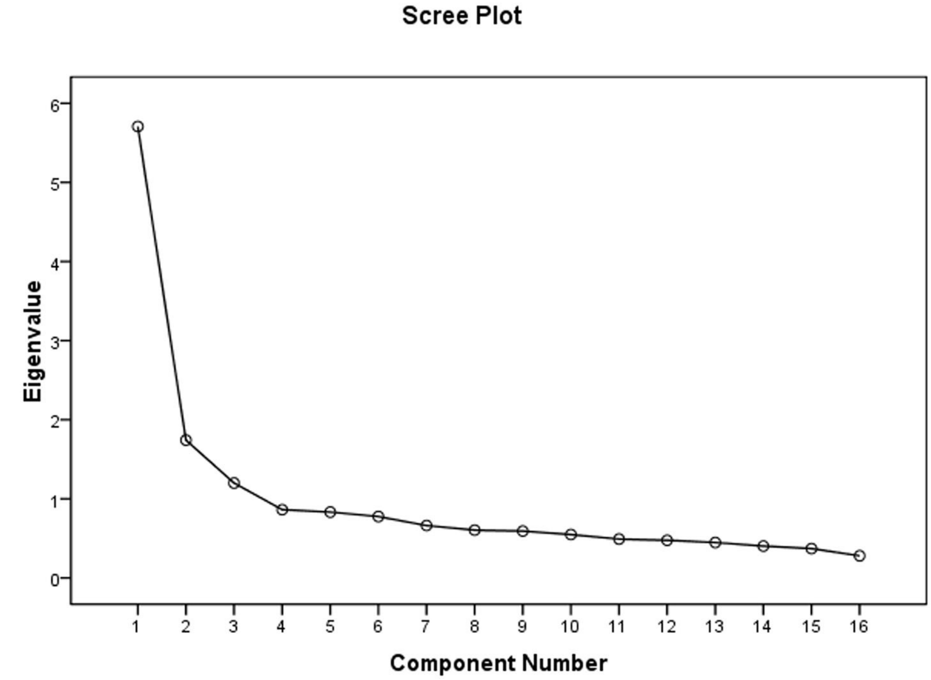

FIGURE 2 | Scree plot of exploratory factor analysis.

TABLE 2 | Rotated component matrix, eigenvalue and cumulative variance contribution rate.

	Items	Factor 1	Factor 3	Factor 2
	Q1	0.624	_	_
	Q2	0.641	_	_
	Q3	0.685	_	_
	Q4	0.714	_	_
	Q5	0.744	_	_
	Q6	0.742	_	_
	Q7	0.583	_	_
	Q8	_	_	0.795
	Q9	_	_	0.817
	Q10	_	_	0.737
	Q11	_	_	0.554
	Q12	_	0.702	_
	Q13	_	0.560	_
	Q14	_	0.589	_
	Q15	_	0.691	_
	Q16	_	0.714	_
Eigenvalue		5.707	1.742	1.200
Variance contribution rate (%)		35.670	10.890	7.498
Cumulative variance contribution rate (%)		35.670	46.560	54.058
Factor naming		Obtain relevant knowledge before haze weather	Self-adjustment after haze weather	Self-protection in haze weather

Factor 1, obtain relevant knowledge before haze weather; Factor 2, self-protection in haze weather; Factor 3, self-adjustment after haze weather. This symbol "_" indicated that values were <0.500. Suppress absolute values <0.500.

TABLE 3 | The content of Brief Haze Weather Health Protection Behavior Assessment Scale-Adolescent Version (BHWHPBAS-AV).

Dimensions	Items	Completely agree 5	Agree most 4	Moderately agree 3	Agree a small part 2	Disagree 1
Obtain relevant knowledge before haze weather	Q1. I often learn the health protection knowledge of haze weather through television media.	5	4	3	2	1
	Q2. I often learn the health protection knowledge of haze weather through Internet media.	5	4	3	2	1
	Q3. I often learn the health protection knowledge of haze weather through related books.	5	4	3	2	1
	Q4. I often learn the health protection knowledge of haze weather through school related publicity activities.	5	4	3	2	1
	Q5. I often learn the health protection knowledge of haze weather through my teacher.	5	4	3	2	1
	Q6. I often learn the health protection knowledge of haze weather through my classmates.	5	4	3	2	1
	Q7. I often learn the health protection knowledge of haze weather through my parents.	5	4	3	2	1
Self-protection in haze weather	Q8. When the haze weather happens, I clean my nasal cavity in time.	5	4	3	2	1
	Q9. When the haze weather happens, I clean my eyes in time.	5	4	3	2	1
	Q10. When the haze weather happens, I clean my face in time.	5	4	3	2	1
	Q11. When the haze weather happens, I wear a protective mask.	5	4	3	2	1
Self-adjustment after haze weather	Q12. After haze weather is over, I eat more fruits and more vegetables.	5	4	3	2	1
	Q13. After haze weather is over, I eat less spicy and irritating food.	5	4	3	2	1
	Q14. After haze weather is over, I increase my physical exercise time.	5	4	3	2	1
	Q15. After haze weather is over, I participate in some recreational activities to relax.	5	4	3	2	1
	Q16. After haze weather is over, I increase my sleep.	5	4	3	2	1

effectively and rapidly asses the degree of haze weather health protection behaviors that adolescents have. Teachers at school, as well as the medical staff in the community, will be able to carry out targeted behavioral interventions and provide adolescents with relevant and necessary health education. These measures will help adolescents develop good health protection habits against the harms that haze weather causes. This will reduce the incidence of related diseases. Finally, this will also reduce the relevant medical burden placed on families, schools, and government bodies (33).

In this study, when researchers needed to asses a hypothesized measurement model, both exploratory factor analysis and confirmatory factor analysis are appropriately applied (27). The sample size in the study should involve at least 10–15 participants per variable for the factor analysis of the scale (27). Our sample size was large enough for the analysis. In the exploratory factor analysis, in order to evaluate the suitability of factor analysis, the research team conducted the Bartlett's test of sphericity and calculated the KMO value. Bartlett's test of sphericity (5668.801, df = 120, $p < 0.001$) was significant,

TABLE 4 | The results of confirmatory factor analysis.

χ^2	df	χ^2 /df	RMSEA	GFI	NFI	IFI	TLI	CFI
271.791	94	2.891	0.051	0.955	0.930	0.953	0.940	0.953

χ^2, Chi square value; df, degrees of freedom; χ^2/df, Chi square value/degrees of freedom; RMSEA, root-mean-square error of approximation; GFI, goodness of fit index; NFI, normed fit index; IFI, incremental fit index; TLI, Tueker-Lewis index; CFI, comparative fit index.

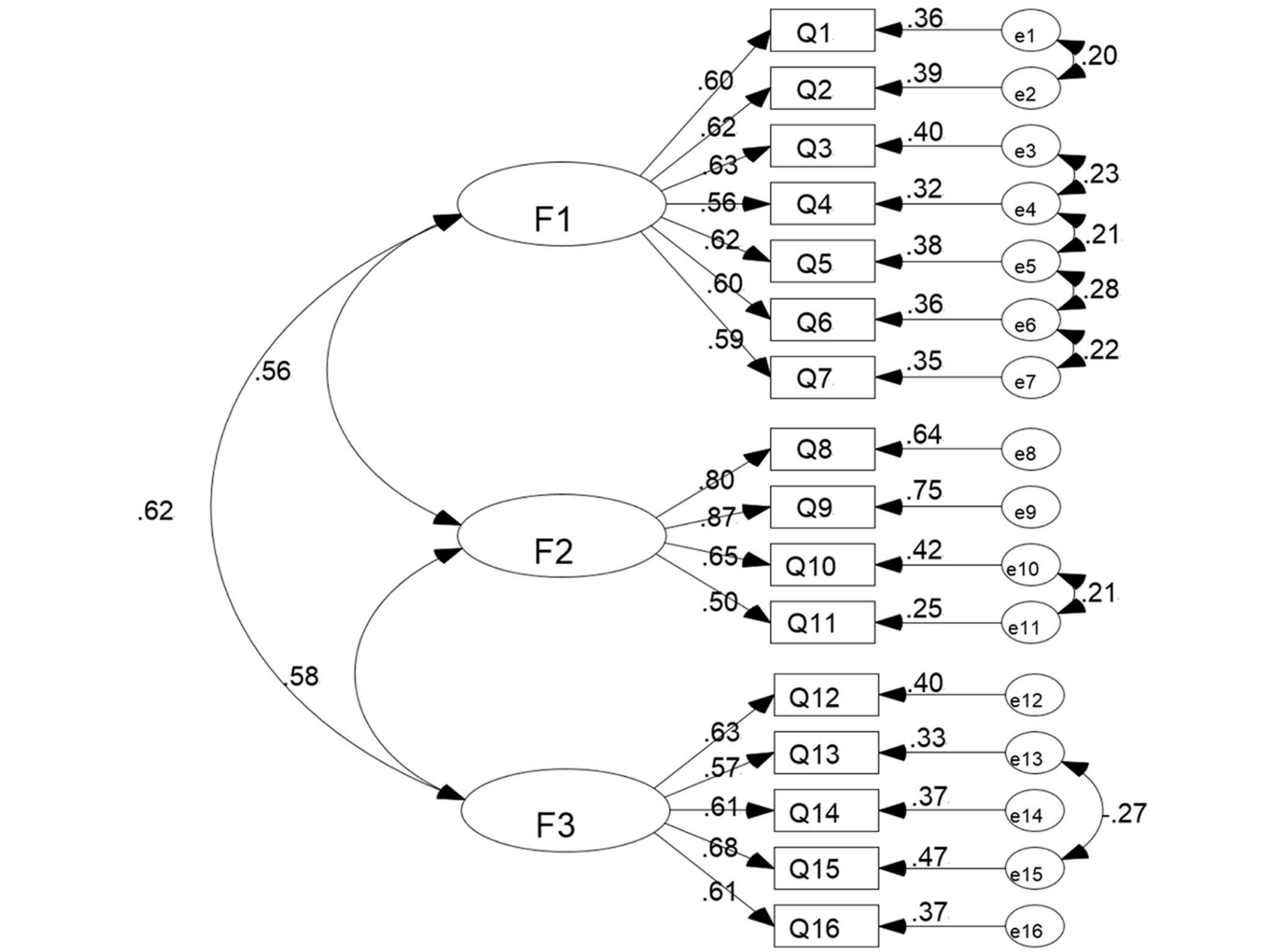

FIGURE 3 | Standard path and parameter estimation of confirmatory factor analysis. F1, Factor 1, obtain relevant knowledge before haze weather; F2, Factor 2, self-protection in haze weather; F3, Factor 3, self-adjustment after haze weather.

TABLE 5 | Correlation coefficients among the factors of BHWHPBAS-AV and between the factors and the whole BHWHPBAS-AV.

Factor	Factor 2	Factor 3	BHWHPBAS-AV
Factor 1	0.458**	0.467**	0.812**
Factor 2	_	0.547**	0.798**
Factor 3	_	_	0.809**

*Factor 1, obtain relevant knowledge before haze weather; Factor 2, self-protection in haze weather; Factor 3, self-adjustment after haze weather. This symbol "_" indicated no such correlation coefficient. ** p < 0.01.*

TABLE 6 | The MIIC and the Cronbach's α coefficient of each factor and the total BHWHPBAS-AV.

Factor	Number of Items	MIIC	Cronbach's α
Factor 1	7	0.424	0.837
Factor 2	4	0.498	0.799
Factor 3	5	0.368	0.744
BHWHPBAS-AV	16	0.312	0.878

Factor 1, obtain relevant knowledge before haze weather; Factor 2, self-protection in haze weather; Factor 3, self-adjustment after haze weather. MIIC, mean inter-item correlation coefficients.

and the KMO value (0.895) in our research was >0.6 (34). The aforementioned findings revealed that the collected data were suitable for factor analysis. Exploratory factor analysis suggested that 16 items loaded substantially onto three conceptually clear factors. Dimension 1 (the way to obtain relevant knowledge before haze weather) and dimension 2 (the source of relevant knowledge before haze weather) in the BHWHPBAS-AV-II were merged into the dimension 1 (obtain relevant knowledge before haze weather) to create the final version of BHWHPBAS-AV. The reason for changing the two dimensions was because, essentially, dimension 1 (the way to obtain relevant knowledge before haze weather) and dimension 2 (the source of relevant knowledge before haze weather) both measure the status of obtaining relevant knowledge prior to the occurrence of haze weather. Therefore, finally, researchers merged dimension 1 (the way to obtain relevant knowledge before haze weather) and dimension 2 (the source of relevant knowledge before haze weather) into dimension 1 (obtain relevant knowledge before haze weather). This helped the BHWHPBAS-AV structure become clearer, more concise, and easier to use. For the confirmatory factor analysis, the model goodness of fit is assessed by RMSEA (<0.08 acceptable), IFI (>0.90 acceptable), GFI (>0.90 acceptable), NFI (>0.90 acceptable), CFI (>0.90 acceptable), and TLI (>0.90 acceptable) (26, 29, 30). All the results of confirmatory factor analysis in this research met the above criteria. The results of confirmatory factor analysis show that both the stability and the fit of the 3-factor model structure of BHWHPBAS-AV are good.

Typically, content validity indicates whether items of a scale are able to identify the topic and content that the researcher aims to measure. The CVI of an item represents the number of expert choices of 3 and 2 divided by the total number of experts. The total CVI value of a scale is the average value of all of items' CVI values (26). The total CVI value of scale in this study was 0.948. The CVI values of all items ranged from 0.667 to 1.00. This indicates that BHWHPBAS-AV is able to reflect the variables measured. Every item was able to measure the correct content, and the BHWHPBAS-AV has shown a satisfactory content validity. The internal correlation test results of BHWHPBAS-AV showed that there was a certain degree of correlation between these factors; furthermore, there were also some differences between these factors. Thus, these factors were able to reflect different aspects of adolescent haze weather health protection behaviors. Thus, the three factors were able to effectively and comprehensively assess the status of adolescent haze weather health protection behaviors.

By using the mean inter-item correlation coefficient and Cronbach's alpha coefficient, researchers can evaluate the scale reliability (35, 36). If the scale MIIC value is > 0.3, the internal reliability of the scale is acceptable (35). In this study, MIIC values of the entire BHWHPBAS-AV and all factors of BHWHPBAS-AV were all >0.3. A general criterion for a good internal reliability of scale is usually the Cronbach's alpha coefficient of ≥ 0.7 (26). In this research, Cronbach's alpha coefficients of the whole BHWHPBAS-AV and all factors of BHWHPBAS-AV were all > 0.7. Therefore, based on the above comprehensive analysis, BHWHPBAS-AV developed in this study has a satisfactory reliability.

LIMITATIONS AND FUTURE DIRECTION

Due to several limitations, future, studies should widen the scope of sampling. Moreover, BHWHPBAS-AV will be more widely verified and used in more areas of the country, so that BHWHPBAS-AV can be better revised and improved in the future. Owing to the lack of an appropriate golden criterion for measuring the adolescent haze weather health protection behavior, testing the criterion validity of BHWHPBAS-AV is a difficult task (29). Therefore, in the future, the criterion validity of BHWHPBAS-AV needs to be further evaluated. Furthermore, in the process of applying BHWHPBAS-AV in other countries, further cross-cultural BHWHPBAS-AV revision and cross-cultural BHWHPBAS-AV validation are also needed.

CONCLUSION

In conclusion, our research team developed and validated the BHWHPBAS-AV with proven reliability and validity. BHWHPBAS-AV can be applied to evaluate adolescent health protection behaviors taken against haze weather. BHWHPBAS-AV can also help teachers at schools, as well as medical staff in community health care institutions, to carry out targeted behavioral interventions and to conduct health education for teenagers regarding protection against haze weather. Ultimately, this will help teenagers establish better haze weather health protection behaviors, which will allow for their healthy growth.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Health and Family Planning Commission of Hebei Province (NO. 20150072). The study was also approved by Medical Ethics Committee of Hebei University. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

QZ, CY, and LT conceived, designed the study, and wrote manuscript. CY, ST, YZ, HD, YC, and YL collected data, analyzed data, and revised manuscript critically. All authors contributed to the article and approved the submitted version.

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Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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From Schools to Scans: A Neuroeducational Approach to Comorbid Math and Reading Disabilities

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We bridge two analogous concepts of comorbidity, dyslexia-dyscalculia and reading-mathematical disabilities, in neuroscience and education, respectively. We assessed the cognitive profiles of 360 individuals (mean age 25.79 ± 13.65) with disability in reading alone (RD group), mathematics alone (MD group) and both (comorbidity: MDRD group), with tests widely used in both psychoeducational and neuropsychological batteries. As expected, the MDRD group exhibited reading deficits like those shown by the RD group. The former group also exhibited deficits in quantitative reasoning like those shown by the MD group. However, other deficits related to verbal working memory and semantic memory were exclusive to the MDRD group. These findings were independent of gender, age, or socioeconomic and demographic factors. Through a systematic exhaustive review of clinical neuroimaging literature, we mapped the resulting cognitive profiles to correspondingly plausible neuroanatomical substrates of dyslexia and dyscalculia. In our resulting "probing" model, the complex set of domain-specific and domain-general impairments shown in the comorbidity of reading and mathematical disabilities are hypothesized as being related to atypical development of the left angular gyrus. The present neuroeducational approach bridges a long-standing transdisciplinary divide and contributes a step further toward improved early prediction, teaching and interventions for children and adults with combined reading and math disabilities.

Keywords: reading and mathematical disability, comorbidity, dyslexia, dyscalculia, psychoeducational testing, neuroimaging, developmental learning disabilities

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INTRODUCTION

The classification, diagnosis, and treatment of learning disabilities are important topics of research in both psychoeducational and neuroscience literature. Researchers in these two fields often measure similar constructs but use differing approaches to work with individuals with learning disabilities. Consequently, each field has produced different concepts and theories over time, leading to a sort of disconnect between the identification of learning disabilities in educational settings and the identification of learning disabilities based on neuroscientific evidence, respectively. Furthermore, the identification and development of comorbid learning disabilities, while a prevalent topic in psychoeducational literature, remains relatively understudied in neuroscience; a testable model of the neuroanatomical substrates of comorbidity is greatly needed. We developed a novel neuroeducational approach to bridge the corresponding concepts on learning disabilities in the two disciplinary fields.

Learning Disabilities

Learning disabilities are a type of neurodevelopmental disorders that impede the acquisition, retention, or application of verbal or non-verbal information, affecting a person's ability to use specific cognitive skills (1, 2). The most prevalent learning disability is *reading disability*, a specific difficulty in learning to read, interpret, and manipulate written words, also known as *dyslexia*. The second most prevalent is *mathematical disability*, a specific difficulty in learning arithmetic and performing mental calculations, also known as *dyscalculia* (3, 4).

Current research on comorbid math and reading disabilities and their developmental origins is far from exhaustive. As recently as 2007, a systematic review of the U.S. Department of Education's Educational Research Information Center (ERIC) database revealed that the number of published studies on reading disability outnumbered the number of studies on math disability by a ratio of 14 to 1 (5). This disparity in knowledge translates into disproportionate diagnoses and asymmetric interventions for individuals with comorbid math and reading disabilities. Therefore, defining a robust neuroeducational model of dyslexia-dyscalculia comorbidity is a priority for the early identification and treatment of learning disabilities (6–8).

The Psychoeducational Approach to Identifying Learning Disabilities

The psychoeducational evaluation is the traditional method of classifying and identifying learning disabilities. The goal of the evaluation is to examine the student's performance on standardized tests of general academic achievement (9), and will determine if the student qualifies for special education or remedial training (10). A psychoeducational assessment typically consists in obtaining an IQ score and selected domain-specific standardized tests—psychometric measures that directly assess abilities in reading, writing, or arithmetic (8, 11).

In this approach, the IQ-achievement discrepancy criterion provides the framework for identifying an unexpected difficulty with learning. To be classified as having a learning disability, the discrepancy model requires that there is a significant discrepancy (usually 1.5 standard deviations) between the person's academic ability or potential (defined by the IQ score) and academic achievement (as defined by their scores on a general reading or math test). This model rests on the questionable assumption that intelligence tests are not confounded by more basic processes for which domain-specific psychoeducational tests provide independent measures (12), and regrettably exclude the possibility of identifying learning disabilities in people with intellectual disabilities (13).

Notably, a study by Tanaka et al. (14) reported evidence based on brain activity demonstrating the diagnostic inappropriateness of the IQ discrepancy criterion. Replicating previous findings based on psychoeducational tests (15) they showed that brain activity and structures associated with reading difficulties in individuals with intact general intellectual ability and in individuals with lower intellectual ability show very similar profiles. The latter supports the recent removal of the IQ discrepancy from the definition of specific learning difficulties in the DSM-V.

The Neuropsychological Approach to Identifying Learning Disabilities

Compared to psychoeducational evaluations, neuropsychological assessments are greater in the depth of their assessment. They are more fine-tuned to examine specific cognitive deficits (such a phonological processing deficits) that underlie learning disabilities. A neuropsychological assessment is performed by licensed clinical neuropsychologists who combine elements of brain anatomy, cognitive neuroscience, and neurodevelopment to infer the neurological correlates of differences in specific cognitive abilities (7).

Secondly, neuropsychological assessments are greater in the breadth of the assessment. In contrast to a psychoeducational evaluation (which typically consists of an IQ score and a few standardized tests), a full neuropsychological assessment includes a structured clinical interview with the client (and interviews with the client's family and/or significant others, if possible), a review of the client's relevant medical records, and the administration of tests that measure domain-general functions such as selective attention, sensory perception, fine motor skills, visuospatial reasoning, and working memory (7). All the available information will be used to make a specific diagnosis of the client's learning disability, instead of relying on psychometric measures alone.

Third, the interpretation of test scores from a neuropsychological assessment is guided by different principles than in a psychoeducational evaluation. Instead of applying the IQ-discrepancy model as in the psychoeducational approach, clinical neuropsychologists define the severity of a learning disability by introducing a cut-off threshold on the tail end of a distribution of academic achievement (9). While cut-off points are useful for providing a post-assessment diagnosis, given the limits of current causal models, they do not precisely reflect the neurobiological basis of a learning disability (13, 16); they rather emphasize normativity and address pragmatic issues related to early intervention.

The identification of math and reading disabilities is a predominant topic in *neuroeducation*—an emerging field at the intersection of neuropsychology, neuroscience, and psychoeducational research (17–19). The goals of neuroeducation are to develop curricula and teaching methods that are based on a scientific understanding of neural mechanisms of learning (20, 21). In line with this paradigm, the central theme of the present paper is linking in the most direct way possible corresponding constructs in education and neuroscience. The aim is to improve prediction of the biological and psychological factors that yield poor academic outcomes seen in individuals with learning disabilities. By examining the cognitive profiles of individuals with dyslexia and dyscalculia and mapping the observed deficits to their neuroanatomical correlates from existing research, the neuroeducational model proposed in the current research provides educators and neuroscience researchers with a working framework for designing effective teaching and interventions specific to individuals with comorbid learning disabilities.

The Neuropsychology of Dyslexia

Approximately 10% of North American children experience developmental dyslexia (22, 23), a disorder characterized by difficulties with reading fluency that are not better explained by visual or cognitive impairments, psychosocial challenges, or poor language instruction. Observable symptoms include inaccurate or effortful reading, poor spelling ability, and the avoidance of leisure or work-related activities that involve reading (22, 24).

The Cognitive Profile of Reading Disability and Dyslexia

There are several overlapping theories about dysfunctional cognitive processes that impair reading fluency in developmental dyslexia (25), however three theories have garnered widespread support in current research literature: the *phonological deficit theory*, *double-deficit theory* and the *visual deficit theory*.

The *phonological deficit* theory is the most widely-promoted and well-established theory in dyslexia research (26, 27). This theory proposes that the core impairment in dyslexia is a deficit in phonological processing—a pervasive difficulty with forming associations between phoneme combinations and the correct corresponding sounds, known as grapheme–phoneme correspondence (28, 29). Deficits in phonological processing can be identified early in development (30, 31) and persist into adulthood (32, 33). Dyslexic children exhibit marked difficulties in manipulating *pseudowords* (non-sensical words made up of valid phonemes in a particular language), and display poor reading fluency when asked to read written words, but not when the words are read to them by another individual (6, 23). In addition, specific training to improve phonological processing leads to significant improvements in reading ability (34, 35).

The *double-deficit theory of dyslexia* builds on the notions presented in the phonological deficit theory. In addition to impaired phonological processing, this theory suggests dyslexia is characterized by a deficit in rapid automatized naming (RAN). RAN is the measure of how quickly an individual can recognize and name aloud a series of familiar objects, pictures, colors, or symbols, or letters (36). While recent studies have suggested that poor RAN performance may reflect impaired functional connectivity between brain structures that control visual processing and speech, poor RAN performance in sight-word reading can indicate phonological deficits in individuals with dyslexia and are more likely to underlie the difficulties in recognizing words (37, 38).

The *visual deficit theory* states that reading disabilities arise due to atypical development of the visual system, whereby there is disruption in the processing of visual information from letters and words in written text. Some neuropsychological studies have shown that individuals with reading disabilities exhibit impaired temporal processing, atypical eye movement regulation, and more frequent visual scanning errors in comparison to normal readers (39). While below-average performance on visual attention tasks in preschool has been shown to predict reading disability (40), it is unclear whether a visual system deficit is a root cause or a result of long-term reading disabilities (41), and seems to contradict recent findings of heightened visuospatial reasoning in dyslexic adults (42–44).

The Neuroanatomical Correlates of Dyslexia

Converging evidence from functional neuroimaging studies has pinpointed three neuroanatomical regions in the left hemisphere which primarily facilitate the multimodal processing of written words: the *left inferior frontal gyrus*, the *fusiform gyrus*, and *temporoparietal parietal junction* (displayed in **Figures 1A,B**). The inferior frontal gyrus contains *Broca's area*, a region that is well-known in neuropsychological literature for its mediating role in speech production, but less recognized for its role in processing phoneme sequences and phonological segmentation (46–48). The *fusiform gyrus* (also known as the *occipitotemporal gyrus*) contains the *Visual Word Form area*, which enables humans to distinguish between the symbols that form letters and numbers, and symbols that are otherwise arbitrary shapes (49, 50). The *temporoparietal junction* (a group of structures including the *angular gyrus*, *supramarginal gyrus*, and the *superior temporal gyrus*) facilitates semantic processing and is also involved in the analysis of phoneme sequences (16, 51, 52).

Dyslexia is associated with atypical development of the left hemisphere language network. Compared to age-matched controls, individuals with dyslexia show atypical physiological activity and white-matter connectivity in several frontal, parietal, and temporal structures in their dominant hemisphere (23, 26, 53). Studies that used functional neuroimaging to examine the neural correlates of phonological decoding consistently found that individuals with dyslexia typically exhibit lower cerebral blood oxygenation at the posterior regions of their language network—usually the left *fusiform gyrus* and the structures of the left *temporoparietal junction* (6, 16). Meta-analyses of neuroimaging studies that compared functional brain abnormalities between individuals with dyslexia have identified a variety of other brain regions that exhibit atypical activity during reading tasks. A meta-analysis of 28 studies identified hypoactivation of the left inferior frontal gyrus, left fusiform gyrus, left temporoparietal cortex, left occipitotemporal cortex, left precuneus, left frontal operculum, left precentral gyrus, and right superior temporal gyrus, as well as hyperactivation in the left anterior insula (54). Two other meta-analyses identified atypical hypoactivity in bilateral *superior temporal gyri*, left *middle* and left *inferior temporal gyri*, left *precuneus*, left *thalamus*, *right postcentral gyrus*, and the *right fusiform gyrus* during reading tasks (55, 56).

The Neuropsychology of Dyscalculia

Developmental dyscalculia is characterized by difficulties in processing numerical information and performing basic mathematical operations, impeding the acquisition of age-appropriate mathematical skills (24, 57). It is estimated that dyscalculia affects 3–6% of the world population (58, 59), but dyscalculia is considerably unrepresented in research literature

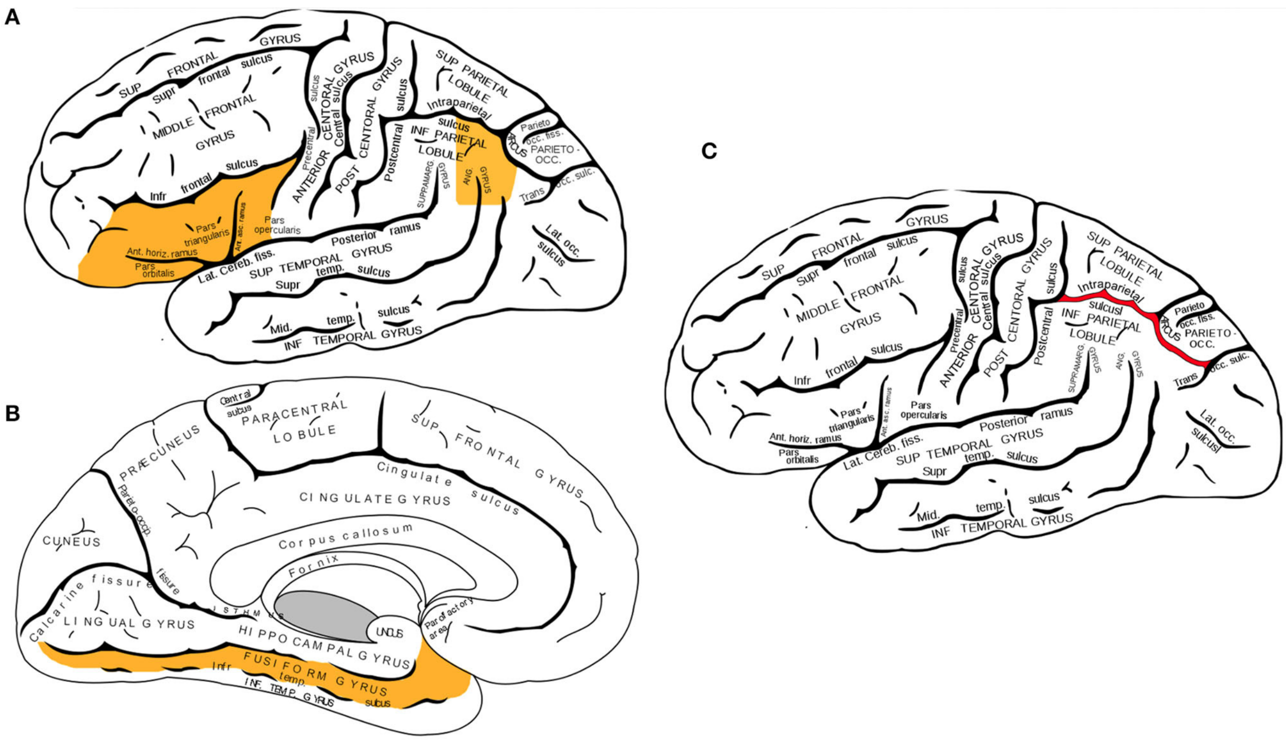

FIGURE 1 | (A,B) Three neuroanatomical structures of the left hemisphere language processing network. **(A)** The left inferior frontal gyrus and the left angular gyrus are highlighted in this lateral view of the cerebral cortex. **(B)** Sagittal view of the cerebral cortex shows the left fusiform gyrus. **(C)** Lateral view of the left intraparietal sulcus. While activation of several brain regions correlates with various aspects of mathematical cognition, intraparietal sulcus (IPS) is the primary activation site during tasks that test numerical magnitude processing (see text). Brain diagrams were adapted from (45). Anatomy of the Human Body. Retrieved from https://www.bartleby.com/107/189.html.

on learning disabilities [5, (57)]. Anywhere from 17 to 66% of individuals with dyscalculia also fit the diagnostic criteria for dyslexia (60, 61). Indeed, students with a math disability are just over two times more likely to also have a reading disability than those without a math disability (62).

The Cognitive Profile of Mathematical Disability and Dyscalculia

Dyscalculia is characterized by impaired *non-symbolic and symbolic numerical processing*, the ability to quickly estimate and manipulate numerical magnitudes and quickly perform mental operations without writing out procedures (63) or relying on verbally-based strategies such as counting (64, 65). The most popular view of mathematical cognition, and consequently of math disabilities [i.e., Triple Code Model; (66, 67)], entail that all development of symbolic number skills derive (through alternative format re-coding), and are ultimately grounded on the innate endowed ability of "number sense." Accordingly, humans would form mental representations of numerical quantities using a *mental number line*, an imaginary line of numbers ordered in an ascending series. Thus, an individual would estimate the place any number or quantity on the number line and perform operations using their approximation of the number—a cognitive function known as *numerical magnitude processing* or the *approximate number system (ANS)*. The acuity of a person's ANS is often measured using *numerical magnitude comparison* tasks with *non-symbolic* quantities (e.g., a group of dots) as opposed to symbolic Arabic digits (e.g., the number 9). In a non-symbolic numerical comparison task, the individual is asked to approximate the correct place for non-symbolic quantity (without counting each item one-by-one) on a visually presented number line. A greater degree of error in ANS has been identified as the core deficit underlying developmental dyscalculia (68, 69). In comparison to typically-developing controls, children with dyscalculia demonstrate lower accuracy in approximating the number of non-symbolic items in a group, and lower accuracy in determining which group of items is greater in magnitude (70, 71). ANS is assumed to rely heavily on spatial representations of numbers (72, 73); individuals with dyscalculia often perform poorly on neuropsychological tests of visuospatial ability (74).

However, a survey of the spectrum of quantity cognition for animals and humans (75) shows that number sense can directly account only for a fraction of the acquired skills, mainly involving approximate small (subitizing for numerosities 1–4) and large quantity assessment and comparison. Oral and written language account for most of the learning spectrum [see Figure 2 in (75)]. On this background, the "primacy" of the number sense has been most recently challenged, since the bulk of multidisciplinary existing evidence demonstrates that the alleged mapping between number sense and symbolic, more complex notions are not as direct as postulated (for example in the most influential Triple Code Model). A review of current neuroscience and behavioral evidence (76) suggests that several alternative possible and plausible routes of normal and atypical non-symbolic to symbolic correlations could occur which provide a better empirical account of math achievement than direct effects of number sense.

One perspective alternative to ANS contends that numerical ability is grounded on representing, understanding, and manipulating *symbol-symbol associations* (SSA). That is, small numerical symbols are initially mapped on a precise representation (e.g., the subitizing range) which, supported by increasing counting and linguistic competency, eventually leads to an independent and exact symbolic system based on order relations between symbols (77). Most magnitude estimation and comparison effects found in studies confirming ANS can be equally explained and modeled in terms of the SSA, and there is also sufficient evidence of distinct brain mechanisms associated to symbolic and non-symbolic numerical processing (78). Further, recent meta-analyses show that symbolic numerical processing tasks are a strong predictor of arithmetic and have consistently been found to be deficient in dyscalculia [see, for instance (79)]. Critically, representatives of this alternative view of numerical cognition differentiate between a non-symbolic deficit and an *access deficit* in dyscalculia, which reflects intact ANS, but deficient access to number semantics from numerical symbols (65).

At the same time, a wealth of evidence in research and practice shows that during formal schooling children with dyscalculia experience learning challenges in *symbolic and linguistic-based quantitative reasoning* related to academic mathematics such as arithmetic, not only numerical skills. These difficulties include learning and remembering exact number words and concepts, and applying skills in: addition/subtraction, multiplication and fraction strategies, commutation and percentages, using place value, as well as geometry, time, measurement, and word problems (80). Without discounting the important role that basic numerical processing might play, the scope of the present work is more narrowly focused on the latter higher-level symbolic and linguistically-based quantitative abilities as they are more directly linked with achievement in educational settings (64).

The Neuroanatomical Correlates of Dyscalculia

The current literature of brain imaging studies reveals that the brain recruits a wide variety of interconnected regions during mathematical tasks, including prefrontal, posterior parietal, occipito-temporal, and hippocampal areas (81). Neuroimaging studies on individuals with dyscalculia have identified two parietal regions associated with the manipulation of numerical quantities: the bilateral *intraparietal sulci* and the left *angular gyrus*. Multiple functional neuroimaging studies have shown that the right and left *intraparietal sulci* (shown in **Figure 1C**) become activated during calculation tasks that involve numerical magnitude processing (69, 82, 83). In contrast, the *angular gyrus* becomes activated during the retrieval of arithmetic facts from long-term memory, such as when finding the solutions to simple multiplications (84, 85).

While the neuroanatomical evidence of atypical brain function in developmental dyscalculia is not quite exhaustive, several functional neuroimaging studies have reported atypical activation patterns at the intraparietal sulci. Compared to age-matched controls, children with dyscalculia exhibit reduced activation at the right intraparietal sulcus when performing non-symbolic numerical comparison tasks [for example (86)]. In addition, applying TMS to the right intraparietal sulcus can severely impede performance on numerical magnitude tasks, artificially producing deficits that are equivalent to those observed in adults with dyscalculia [for example (87, 88)].

An Overview of Cognitive Profiles of Comorbid Math and Reading Disabilities

A body of work has focused on the cognitive profiles of individuals with comorbid math and reading disabilities, establishing a design (here dubbed as the "four-groups design") which has become pivotal in this research area. Specifically, Willburger et al. (89) examined cognitive performance in children with reading disability alone (RD), mathematical disability alone (MD), or with comorbidity of both disabilities (MDRD) as compared to typically developing and/or achieving children (TD and/or TA). MDRD children exhibited additive deficits in rapid automatized naming; this suggested that the deficits associated with comorbidity are additive and not qualitatively different from the deficits in the single disabilities. Later, this team (90) examined how domain-specific processes (e.g., symbolic and non-symbolic numerical processing, phonological processing) and domain-general processes (e.g., working memory, computations) contribute to comorbidity. MDRD children exhibited domain-specific deficits in phonological processing and numerical magnitude processing, performing at the same level as individuals with RD or MD. Unexpectedly, both the latter groups demonstrated better short-term working memory than the MDRD and the TD group, hinting that some domain-general processes may contribute to comorbidity.

Furthermore, Moll et al. (91) showed how three domain-general processes—namely processing speed, temporal processing, and verbal memory—can correlate differentially with reading and mathematical performance and are also associated with inattentive behavior. Both RD and MD children exhibited deficits in verbal memory. However, after controlling for parent-reported difficulties with inattention, deficits in verbal processing became associated with reading ability alone, whereas slowed temporal processing and visuospatial memory deficits were associated with mathematical ability alone. The authors concluded that deficits in processing speed, temporal processing, and verbal memory reflect variations in subclinical attention difficulties, and that reading and mathematical disabilities may thus be the outcome of multiple impaired cognitive systems rather than individual domain-specific processes. Relatedly, Wilson et al. (74) demonstrated that MDRD adults exhibited additive deficits in rapid naming and working memory, equivalent to the sum of the deficits exhibited by adults with the single disabilities. These authors concluded that additive domain-general deficits were likely correlates (not the underlying cause or the eventual consequence) of comorbidity.

More recent studies have shown that some processes traditionally considered as domain-specific may play an important role in comorbidity. Slot et al. (92) showed that children's rapid automatized naming and phonological awareness were associated with reading performance, whereas number sense and visuospatial working memory were associated with mathematical performance. However, phonological awareness was also predictive of mathematical performance, suggesting that a shared deficit in phonological processing may underlie both RD and MD. Similarly, Raddatz et al. (93) found that MD children showed deficits in various non-verbal and verbal tasks related to number processing, whereas RD children showed deficits in verbal tasks.

In contrast to this literature, neuroscience studies have rarely adopted the four-groups design. Improving on this limitation, the following study was designed to start filling some gaps in clarifying the nature of domain-specific and domain-general deficits of reading-math comorbidity with reference to the currently known neural underpinnings of dyslexia and dyscalculia.

The Present Study: Research Questions, Design and Hypotheses

The primary objective of this study was to outline a neuroeducational model of dyslexia-dyscalculia comorbidity—a framework for understanding the psychoeducational and neuropsychological characteristics of individuals with comorbid math and reading disabilities that can be tested for validity in future research. The neuroeducational model proposed here is [as defined by (94)] a preliminary theory or set of hypotheses to synthesize current knowledge and then guide and refine evidence-based practice in education, public health and the allied fields. It should not by any means be interpreted as proof of established knowledge or theory. We fully expect this "probing" model to be tested and re-tested and in this process modified, refined, or even falsified based on future research. This model was established in three phases. In the first phase—using a psychoeducational approach—we examined the performance of individuals with math and reading disabilities on a series of psychoeducational tests and drew conclusions about the specific cognitive deficits they exhibited. In the second phase—using a neuropsychological approach—we performed a systematic review of existing clinical studies on the neuroanatomical correlates of dyslexia and dyscalculia; then, we identified the involvement of key neuroanatomical structures displaying abnormal function. In the third phase, we mapped the deficits as measured by psychoeducational tests to their most plausible neuroanatomical correlates obtained in the systematic review, creating a neuroeducational model of comorbidity that unites the broad psychoeducational definitions of math and reading disabilities with neuropsychological evidence of the biological characteristics of dyslexia and dyscalculia. This series of operations allowed us to build correspondence between the diagnostic tools used to identify learning disabilities in psychoeducational context and the neurodevelopmental theories of dyslexia and dyscalculia in the neuropsychological literature.

To determine the cognitive profile of comorbid dyslexia-dyscalculia, performances were measured from a sample population with math disability, reading disability, and dual math, and reading disability via a comprehensive battery of psychoeducational tests. The statistical analyses of their psychoeducational outcomes were used to (i) determine if there were any measurable cognitive deficits that were unique (in nature or in magnitude) to the participants with comorbid reading-math comorbidity; (ii) determine if the cognitive deficits were domain-specific (within the realm of reading or numerical cognition) or domain-general (working memory and/or executive functions outside the realm of reading or numerical cognition) in nature; (iii) determine the nature of the relationship between math and reading deficits in the comorbid group. Two sets of hypotheses were assessed:

First set of hypotheses: It was hypothesized that the deficits in the comorbid participants would either be *additive* (where the approximate sum of the deficits in the reading-disabled participants and the math-disabled participants is measured), *synergistic* (an over-additivity caused by an interaction between math and reading deficits), or *antagonistic* (an under-additivity caused by an interaction between math and reading deficits). The nature of the relationship between math and reading disabilities was determined using the same 2×2 factorial design previously used in the four-groups design literature (74, 90). A significant interaction between the math disability and reading disability indicates a synergistic over-additivity or an antagonistic under-additivity in the mean scores of a particular test, and lack thereof indicates an additive effect.

Second set of hypotheses: it was hypothesized that the comorbid participants in this study would exhibit: (1) impaired reading fluency and phonological processing equivalent to those shown by individuals with reading disability alone, (2) impaired quantitative reasoning skills equivalent to those shown by participants with mathematical disability alone, and (3) deficits in working memory equivalent to those shown by individuals with reading disability alone. Consequently, it was also hypothesized that, consistent with the proposed neuroeducational approach, it should be possible to derive a mapping of correspondence between the pattern found in the psychoeducational findings and known neuroanatomical correlates in the clinical neuroimaging literature, which can be empirically tested with further neuroimaging studies.

MATERIALS AND METHODS

Psychoeducational Tests

Tests of Achievement for Identifying Math and Reading Disabilities

Wide Range Achievement Test 3rd edition (WRAT3) arithmetic subscale

The WRAT3 Arithmetic subscale is a test of written arithmetic problems, which included number addition, subtraction, multiplication, and problems involving fractions and decimals.

Wide Range Achievement Test 3rd edition (WRAT3) reading subscale

The *Reading subscale* is a single word reading test, where participants were asked to read aloud a series of increasingly difficult words.

Testing Phonological Processing

Rosner Auditory Analysis Test

The Rosner Auditory Analysis test is the first of two Phoneme Deletion tasks included in this study. Participants were instructed to repeat a list of 40 common English words. Next, the test administrator asked the participant to repeat each word without pronouncing a specific phoneme, thereby "deleting" the first, last or embedded phoneme from the word and pronouncing the word fragment(s) that remained.

Pseudowords Phoneme Deletion task

In this second Phoneme Deletion task, participants were instructed to listen to 30 pseudowords and then repeat it by "deleting" a specific phoneme.

Woodcock Reading Mastery Tests-Revised (WRMT-R) Word Attack subtest

The Word Attack subtest examines a participant's phoneme-grapheme awareness without relying on a verbal demonstration by the test administrator (95). Participants were instructed to read a list of 45 pseudowords. The level of difficulty gradually increased throughout the test; the number of syllables in each pseudoword increased intermittently from 1 syllable to 4 or 5 syllables by the end of the list.

Testing Quantitative Reasoning

KeyMath revised, interpreting data subtest

Participants completed the Interpreting Data subtest of the revised KeyMath Assessment (KeyMath-R) (96). Participants were tasked with solving a written mathematical problem (i.e., "Kareem can read sixty pages in two and one–half hours. How many pages can he read in 1 hour?").

Testing Intellectual Functioning

All participants, aged 17 and over, completed three subtests of the Wechsler Adult Intelligence Scale (WAIS-R) (97). Participants aged 6 to 16 completed three analogous subtests from the Wechsler Intelligence Scale for Children (WISC-III) (98).

WAIS-R/WISC-III vocabulary subtest

The Vocabulary subtest measures a person's semantic memory retrieval. Participants performing the WAIS-R were asked to orally define a series of 30 vocabulary words, gradually increasing in difficulty. Participants performing the WISC-III were asked to name pictures representing each word.

WAIS-R/WISC-III Block Design subtest

In the Block Design subtest, participants were asked to re-create a model or a picture of a design using up to nine red and white blocks within a time limit. This test was included as a measure of visuospatial reasoning.

WAIS-R/WISC-III Digit Span subtest

The Digit Span subtest examines verbal working memory. Participants were presented orally with a series of single-digit numbers. In the first half of the trials, they were required to orally repeat the presented numbers in the same order they heard (forward digit span); in the second half of the trials, they were to repeat the presented numbers in the reverse order (backward digit span).

Procedure

Participants were tested individually for a 3 h session (including two 10 min breaks). Each testing session began with the administration of the Vocabulary, Block Design, and Digit Span subtests from the WAIS-R or the WISC for participants aged 6–16. Successively, after the first break, each participant completed the Reading, Spelling, and Arithmetic subscales of the WRAT3. After the second break, each participant completed a series of psychoeducational tests. All participants completed four tests of phonological processing: The Rosner Auditory Analysis task, the Pseudowords Phoneme Deletion task, followed by the Word Attack and Word Identification subtests of the Woodcock Reading Mastery Tests (WRMT-R), the latter being excluded from this analysis. Lastly, participants completed the KeyMath Interpreting Data subtest.

Sampling

The participants in this study were selected from a database resulted from a 10 year prospective cohort research study at the University of British Columbia (UBC). A total of 585 participants ranging from 7 to 77 years of age were recruited from around the Greater Vancouver Area as well as the graduate and undergraduate student population at UBC. They were recruited through a publicly advertised free comprehensive psychoeducational assessment offered as compensation for their participation and in exchange for use and publication of the resulting anonymous group data and findings. This study was approved by the UBC institutional research ethics boards in accordance with the 1964 Declaration of Helsinki ethical standards and in strict adherence of the Tri-Council Policy Statement (https://ethics.gc.ca/eng/policy-politique_tcps2-eptc2_initiatives.html). Participants or their parents/guardians (for children $<$ 12 years of age), signed a consent form; parental/guardian's consent was conditional on children's active assent. Participants or parents/guardians completed a brief questionnaire on demographic and socioeconomic information about themselves or their family.

The testing format varied over the decade of data collection, and over 50 different types of test scores were entered into database. The initial database was reduced so as to only include the participants who completed specific psychometric tests in the same specific format and which therefore provided information about the cognitive profiles of individuals with dyslexia and dyscalculia permitting to test the objectives of this study.

Inclusion criteria for the present study entailed: (1) completion of The WRAT3 Arithmetic subscale and WRAT3 Reading subscale, which served as the main diagnostic indicators; (2) completion of domain-specific tests that examine phonological processing (the reading domain) or quantitative reasoning (the mathematical domain), and domain-general tests that examine executive functions (such as working memory and spatial reasoning). In the mathematical domain, only one test was selected: the Interpreting Data subtest of the revised KeyMath Assessment; (3) completion of three neuropsychological tasks that test domain-general cognitive functions were selected: The Vocabulary, Block Design, and Digit Span subtests of the WAIS-R (for participants ages 17 and older) or the WISC-III (participants ages 16 and younger); and finally (4) Estimated IQ scores [calculated using the sum of the WAIS-R Vocabulary subtest and the WAIS-R Block Design subtest, as in (12)] had to be > 70, which we adopted as the clinical threshold for low IQ (99).

The final analysis included data from *360* participants. The average age of the participants (on the day of testing) in each group are presented in **Table 1**. A one-way ANOVA followed by Tukey *post-hoc* multiple comparisons revealed that the average age of the RD group was significantly lower than the average age of the TA group ($p = 0.008$), the MD group ($p = 0.017$) and the MDRD group ($p = 0.007$). A two-way MANCOVA was conducted to examine the effect of age on the scores from all seven psychoeducational tests, with math disability and reading disability as the independent variables and age as a covariate. Using the Bonferroni procedure to correct for multiple ANOVAs (with a significant threshold of $p < 0.008$), there were no significant interactions between age and math disability, nor between age and reading disability, on the mean scores for any of the psychoeducational tests (Wilks' Lambda = 0.012). Furthermore, a three-way MANOVA was conducted with math disability, reading disability, and age category as independent variables. The participants were divided into two age categories: below age 16 and above age 16. MD × Age Category interaction was not significant (Wilks' Lambda = 0.780) nor was the RD x Age Category interaction Wilks' Lambda = 0.349). As a result, the low average age of the dyslexic did not appear to have a significant effect on the psychoeducational test results as whole.

Other MANOVA and MANCOVA analysis using a similar approach as the one used to investigate age effects showed no significant sex differences.

Diagnostic Criteria and Subgroups

Using previously established cut-off criteria (15, 100), each participant was assigned to one of four groups: the *math disability (MD)* group (participants who scored 25th percentile or lower on the WRAT3 Arithmetic subscale), the *reading disability (RD) group* (25th percentile or lower on the WRAT3 Arithmetic subscale), the *comorbid math and reading disability (MDRD)* group (25th percentile or lower on both WRAT3 subscales), and the *typical achievement (TA)* control group (higher than the 25th percentile on both WRAT3 subscales). The mean percentile scores for each group and mean age of the participants (on the day of testing) are reported in **Table 1**.

Three measures of socioeconomic status (SES)—education, occupation, and median income—were evaluated in the present study (reported in **Table 1**). To measure SES, we used the Kuppuswamy's socioeconomic ranking scale validated for urban communities (101). Each participant received a numerical rating between 1 and 7 for the highest level of education they had achieved by the day of testing (1 = elementary school certificate or currently enrolled, 2 = middle school certificate, 3 = secondary school diploma, 4 = some college/university or post-secondary diploma, 5 = college/university degree, 6 = graduate degree, 7 = professional degree). Each participant received an individual rating for their occupation status (1 = unemployed, 2 = unskilled worker, 3 = semi-skilled worker, 4 = skilled worker, 5 = clerical, shop-owner, farmer, 6 = semi-profession, 7 = profession). For the participants ages 16 and younger, the highest level of occupation status achieved by either one of their parents

TABLE 1 | Characteristics of the four groups.

	TA	MD	RD	MDRD	All groups
N	158	69	46	87	360
Mean age (years)	26.47 (14.66)	26.80 (12.69)	19.22 (10.96)	27.22 (12.99)	25.79 (13.65)
n Female	77	35	16	47	175
% Female	48.73%	50.72%	34.78%	54.02%	48.61%
WRAT3 Arithmetic	57.82 (19.38)	13.62 (7.34)	50.26 (17.05)	10.34 (7.66)	36.84 (26.81)
WRAT3 Reading Mean Score	63.42 (19.24)	53.00 (17.18)	12.09 (8.20)	10.14 (8.11)	41.99 (28.77)
Estimated IQ	109.61 (14.28)	97.75 (13.45)	100.80 (16.42)	88.74 (13.57)	101.17 (16.51)
Education Rating	3.23 (1.41)	3.10 (1.29)	2.43 (1.19)	3.15 (1.22)	3.08 (1.31)
Occupation Rating	3.65 (1.30)	3.69 (1.53)	3.67 (0.87)	3.20 (1.44)	3.54 (1.37)
Median Income[a] Rating	3.52 (1.63)	3.41 (1.36)	3.50 (1.34)	3.49 (1.28)	3.49 (1.48)

Numbers in parentheses represent one standard deviation from the mean. Underlined numbers are mean percentile scores that are within the 25th percentile on the WRAT3 Arithmetic or WRAT3 Reading subtest.

[a]The distribution of income relative to the period studied was relatively stable in Vancouver and comparable to other big cities (>1M) in Canada (i.e., Toronto, Montreal, Ottawa, Calgary). Although the distribution was positively skewed around the mean (60–70 K in CND$) relative to other cities, because we do not intend to generalize our results to the entire Canada, what is most relevant is that there were no differences in income distribution between our four groups.

was used as a proxy for their own occupation rating. Median household income ratings were generated for each participant by the postal code of the home address that they provided on the day of testing (1 = $50,000 or less, 2 = $50,000–$60,000, 3 = $60,000–$70,000, 4 = $70,000–$80,000, 5 = $80,000–$90,000, 6 = $90,000 or more), based on most temporally proximal Canadian population census data (102). Preliminary one-way ANOVAs and Tukey *post-hoc* multiple comparisons were conducted to identify any between-group differences in the three SES measures. There were no significant between-group differences in occupation or median income rating. There was only a significant difference in mean education rating *between* the TA and RD groups ($p = 0.007$); as previously noted, this is explained by the lower average age of the RD group, when the contrast on mean education rating was run controlling for age this difference was no longer significant.

Statistical Analysis

The analysis involved two separate sets of tests for the first and second set of hypotheses (see section The Present Study: Research Questions, Design and Hypotheses). Relative to the first set of hypotheses, a two-way ANOVA was conducted for each of the seven psychoeducational tests, to assess just the *interaction* between math disability and reading disability across the four groups. The model followed a 2 × 2 factorial design, where the two between-subject factors were math disability (with two levels, math disability vs. no math disability) and reading disability (also two levels, reading disability vs. no reading disability). We followed the same procedures consolidated in previous studies on individuals with comorbid math and reading disabilities (74, 90) whereby, the interaction term serves as an indicator of the type of relationship between deficits in individuals with MDRD. A significant 2-way interaction between math disability and reading disability would indicate a synergistic or antagonistic relationship between math and reading disability—an over-additivity or under-additivity of deficits in domain-specific or domain-general cognitive processes. Main effects were irrelevant to the objectives of the study and are not considered, to avoid redundancy. Nonetheless, for rigor, they were counted in the correction for Type I error inflation due to multiple testing (see below). To assess the second set of hypotheses, one-way ANOVAs and *post-hoc* Tukey pairwise comparisons were used to analyze focused between-group differences in performance between TA, RD, MD, and MDRD groups, with each psychoeducational test measurement as dependent variable, and learning disability groups as levels of the independent variable/factor. This followed directly from the second set of hypotheses for this study.

To counteract Type I error inflation, we adopted the standard Bonferroni criterion; effects were deemed significant if p was below 0.00192; this corresponded to the *p*-value adjustment: 0.05/26 tests, which included all interaction and main effect tests of the two-way ANOVA as well as all one-way ANOVAs. Correspondingly, for the Tukey procedure, the same correction was applied to keep *p*-values below adjusted 0.05 level.

Systematic Review of the Neuroanatomical Correlates of Dyslexia-Dyscalculia Comorbidity

The protocol for this review followed the guidelines established by the *Preferred Reporting Items for systematic Review and Meta-Analysis Protocols* (103); a detailed checklist with inclusion/exclusion criteria, search terms, and methods is presented in **Table 2**.

The systematic review was performed in four stages. The first stage was a preliminary search to identify well-cited authors on math and reading disabilities and the avenues for future research using Google Scholar. The second stage of review provided working definitions of dyslexia and dyscalculia using PsycINFO and the Education Resources Information Center (ERIC). The third stage served to identify the neural correlates unique to dyslexia, the neural correlates unique to dyscalculia, and the neural correlates that are shared between the disorders. A detailed search of biomedical literature was performed to examine evidence from four types of studies: (i) functional neuroimaging studies, (ii) structural neuroimaging studies, (iii) functional connectivity studies, and (iv) lesion-symptom mapping studies. This stage of the review was conducted using literature available through the Web of Science and the National Center for Biotechnology Information (PubMed). The fourth and final stage of the review was initially conducted in April 2015 and identified 26 empirical studies met inclusion criteria. This stage of the review was repeated in April 2020 to add updated findings from the literature; 24 additional empirical studies were identified.

The functional neuroimaging studies included compared the brain physiology of people with and without dyslexia during reading tasks or compared the brain physiology of people with and without dyscalculia while they performed mathematical tasks. The structural neuroimaging compared the white and/or gray matter volume in specific neuroanatomic regions among people with dyslexia and/or among people with dyscalculia and normal controls. Similarly, the functional connectivity studies used the same types of comparisons applying MRI tractography in multiple neuroanatomic regions. To support the model with direct evidence, the searches also identified empirical clinical studies that examined patients with *alexia* (acquired dyslexia) or *acalculia* following traumatic brain injury.

RESULTS

Tests of Phonological Processing

Mean scores on the tests of phonological processing (Rosner Auditory Analysis: RAA, Pseudowords: PW, Word Attack: WA) are shown in the panels of **Figure 2**. The results of the two-way ANOVA did not reveal a significant interaction between math disability and reading disability for any of these tasks (all F's < 2.89; $p > 0.10$, $\eta^2_p \leq 0.01$). The one-way ANOVA identified a significant effect of learning disability group on the mean scores of the RAA [$F_{(3, 356)} = 28.90$, MSE = 2448.46, $p < 0.0001$, $\eta_p{}^2 = 0.196$], PW [$F_{(3, 356)} = 39.28$, $MSE = 2067.86$, $p < 0.0001$, $\eta_p{}^2 = 0.25$], and WA [$F_{(3, 356)} = 49.92$, $MSE =$

TABLE 2 | PRISMA-P Protocol for Systematic Review (103).

Rationale	To identify any neuroanatomical structures whose atypical function may be associated with the cognitive deficits exhibited by individuals with dyslexia and dyscalculia
Objectives	The review answered the following questions: • "What brain regions show atypical activity in dyslexia alone?" • "What brain regions are show atypical activity in dyscalculia alone?" • "What brain regions are atypical activity in comorbid dyslexia-dyscalculia?
Eligibility criteria	Studies published in academic research journals since January 1, 2004. This marks the beginning of the current definition of specific learning disability (104) • The studies involved 20+ participants, males and females ages 6 and older • The studies followed a quasi-experimental design with at least two groups: one group with a learning disability (dyslexia or dyscalculia) and a control group • The studies did not involve individuals with any medical condition (other than dyslexia or dyscalculia) or any other life circumstance that could have influenced their performance on the cognitive tasks (ADHD, neurodegenerative disease, lack of education, etc.) • The investigators applied one of the three following techniques: a) Functional magnetic resonance imaging (fMRI) to examine physiological correlates of cognitive activity during phonological or numerical magnitude comparison tasks b) Diffusion tensor imaging (DTI) to examine structural differences in white or gray matter composition between key neurological structures c) Lesion-symptom mapping (caused by either a stroke or a brain tumor)
Information sources	• Google Scholar • American Psychological Association (PsycINFO) • Education Resources Information Center (ERIC) • NIH MEDLINE Database (PubMed) • Web of Science
Search strategy	Step 1: Preliminary Search A preliminary search was performed using Google Scholar to find the leading authors in learning disabilities research, identify their seminal publications on dyslexia, dyscalculia, and provide a working definition for each disorder Step 2: Existing Meta-Analyses A secondary search was performed to using PsycINFO and ERIC to identify studies that examined comorbid dyslexia-dyscalculia, and to identify any existing meta-analyses on the cognitive or neurological correlates of each learning disability Step 3: Detailed Search A detailed search of medical literature was performed using PubMed and Web of Science to identify empirical studies that used functional or structural MRI to examine individuals with (i) dyscalculia and (ii) dyslexia, and that report the neuroanatomical structures where atypical function, white matter composition, or functional connectivity is associated with each disorder a) A combination of the following search terms were used to identify functional and structural neuroimaging studies on dyslexia: "neurobiological dyslexia" "neurobiological reading disability" "brain region dyslexia" "brain region reading disability" "neuroimaging dyslexia" "neuroimaging reading disability" "fMRI dyslexia" "fMRI reading disability" "MRI dyslexia" "MRI reading disability" "DTI dyslexia" "DTI reading disability" b) A combination of the following search terms were used to identify functional and structural neuroimaging studies on dyscalculia: "neurobiological dyscalculia" "neurobiological math disability" "brain region dyscalculia" "brain region math disability" "neuroimaging dyscalculia" "neuroimaging math disability" "fMRI dyscalculia" "fMRI math disability" "MRI dyscalculia" "MRI math disability" "DTI dyscalculia" "DTI math disability" Step 4: Lesion-symptom Mapping Studies A second detailed search of medical literature was performed using PubMed and Web of Science to identify empirical studies that examined patients with alexia (acquired dyslexia) or acalculia (acquired dyscalculia), who had suffered damage to structures that mediate in math or reading ability, caused by an ischemic stroke or brain tumor a) A combination of the following search terms were used to identify lesion-symptom case studies of patients with alexia: "neurobiology AND acquired dyslexia" "neurobiology AND alexia" "lesion AND acquired dyslexia" "lesion AND alexia" "stroke AND acquired dyslexia" "stroke AND alexia" "brain tumor AND acquired dyslexia" "brain tumor AND alexia" b) A combination of the following search terms for acquired dyscalculia: "neurobiology AND acquired dyscalculia" "neurobiology AND acalculia" "lesion AND acquired dyscalculia" "lesion AND acalculia" "stroke AND acquired dyscalculia" "stroke AND acalculia" "brain tumor AND acquired dyscalculia" "brain tumor AND acalculia"
Study records	One independent reviewer selected the published studies that fit the eligibility criteria. The selected publications were legally stored and classified using Mendeley Desktop (Version 1.15.2) for Windows 10
Outcomes and prioritization	The desired outcome was a list of brain regions that are involved in dyslexia and dyscalculia. Priority was given to studies that included participants all four groups (participants with dyslexia, dyscalculia, comorbid dyslexia-dyscalculia, and controls)
Synthesis	The results of the systematic review are synthesized using a table as displayed below. Each brain region identified in the review is classified by learning disability (whether the region is associated with dyslexia, dyscalculia), as well as by the type of atypical functionality displayed (whether the brain region is generally more active or inactive in individuals with the learning disability). Of primary interest are the neuroanatomical structures whose atypical function is common to dyslexia and dyscalculia; these structures are underlined in the table

4528.74, $p < 0.0001$, $\eta_p{}^2 = 0.30$]. For all three tasks, Tukey *post-hoc* multiple pairwise comparisons revealed a similar pattern of significant intergroup mean scores differences, showing TA > RD, and TA > MDRD (all *p*'s < 0.01). For all other comparisons, RD = MD = MDRD (For more details see Note in **Figure 2**).

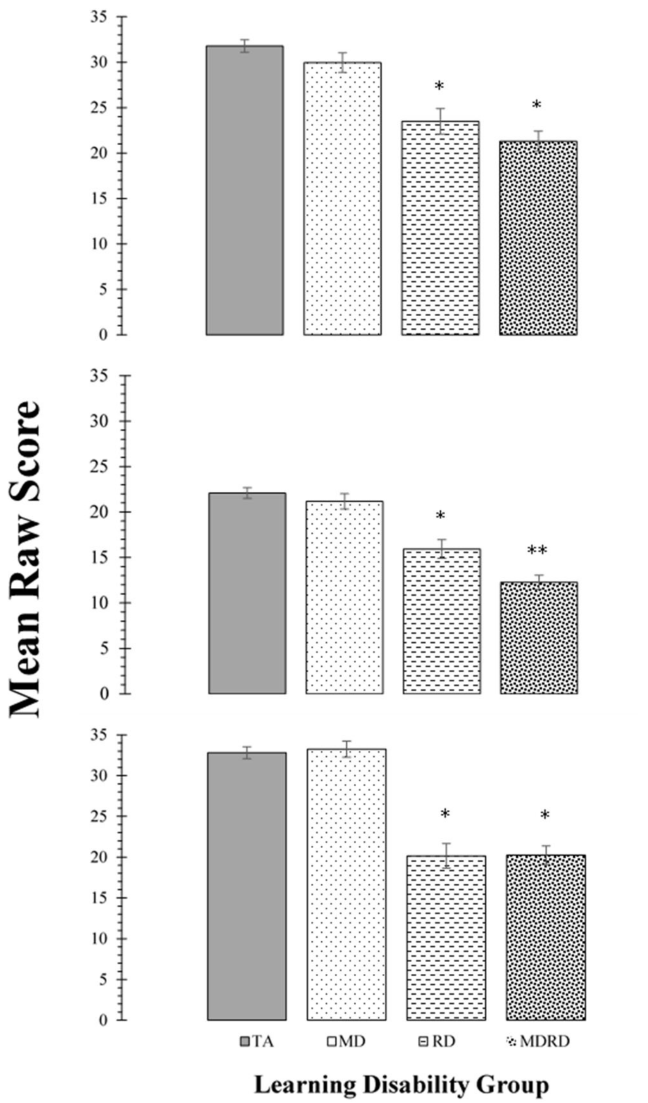

FIGURE 2 | Top Panel: Mean scores for the Rosner Auditory Analysis task. Middle Panel: Mean scores for the Pseudowords Phoneme Deletion task Bottom Panel: Mean scores for the WRMT-R Word Attack subtest. For all panels, bars represent one standard errors. Asterisks summarize the results of *post hoc* Tukey test comparisons: "*" indicates a significant difference from the typical achievement (TA) control group at $p < 0.05$; "**" indicates a significant difference from the reading disability (RD) group at $p < 0.05$. TA > RD (Rosner Auditory Analysis: 8.29 [CI: 4.31 to 12.27]; Pseudowords: 6.15 [CI: 3.02 to 9.29]; Word Attack: 12.65 [CI: 8.53 to 16.76]; all *p*'s < 0.01). TA > MDRD (Rosner Auditory Analysis: 7.30 [CI: 7.30 to 13.64]; Pseudowords: 9.84 [CI: 7.34 to 12.43]; Word Attack: 12.56 [CI: 9.27 to 15.84]; all *p*'s < 0.01). For all other comparisons, RD = MD = MDRD.

The KeyMath Interpreting Data Subtest (Henceforth, *KeyMath-ID*)

Mean scores and standard errors for the KeyMath-ID are shown in **Figure 3**. The two-way ANOVA did not reveal a significant interaction between math disability and reading disability ($F < 1$, $p = 0.73$, $\eta_p^2 = 0.01$). The one-way ANOVA identified a significant effect of learning disability group on the level of performance [$F_{(3, 356)} = 12.23$, $MSE = 238.550$, $p = 0.0001$, $\eta_p^2 = 0.093$]. Tukey comparisons revealed that the mean scores of the TA group were significantly higher than all other disability groups (TA> MD, TA > RD, and TA > MDRD (all *p*'s < 0.05). However, there were no other significant differences among the mean scores of the groups of individuals with disabilities, that is, RD = MD = MDRD (For more details see Note in **Figure 3**).

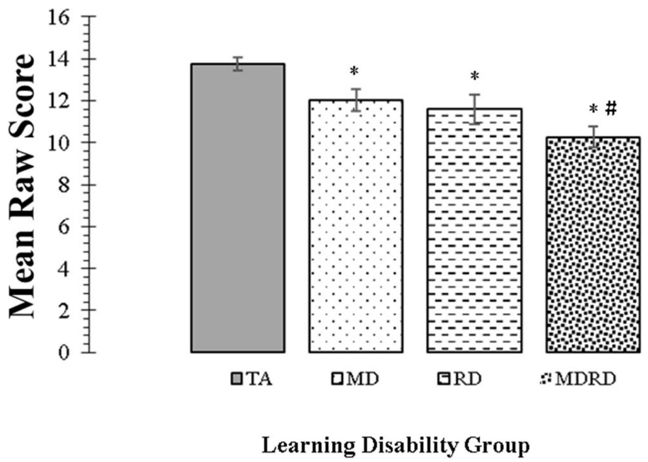

FIGURE 3 | Mean scores for the KeyMath Interpreting Data subtest. Bars represent one standard errors. The symbol "*" indicates a significant difference from the typical achievement (TA) control group at $p < 0.05$ on *post hoc* Tukey test. The symbol "#" indicates a marginally significant trend at $0.05 < p < 0.10$ on *post hoc* Tukey test. TA > MD (1.72, [CI: 0.07 to 3.36], $p = 0.04$). TA > RD (2.16, [CI: 0.25 to 4.07], $p = 0.02$). TA > MDRD (3.47, [CI: 1.95 to 4.99], $p < 0.01$). RD = MD. RD = MDRD. # MD > MDRD (1.75, [CI: −0.08 to 3.59], $p = 0.068$).

Tests of Intellectual Functioning

Mean scores on the tests of intellectual functioning (WAIS-R Vocabulary, Digit Span, and Block Design) are shown in the panels of **Figure 4**. The two-way ANOVA did not reveal a significant interaction on any of these subtests [$F_{(1, 356)} < 1.33$, $p > 0.25$, $\eta_p^2 \leq 0.02$]. The one-way ANOVA identified a significant effect of learning disability group on the mean scores of the Vocabulary subtest [$F_{(3, 356)} = 50.14$, $MSE = 383.92$, $p < 0.0001$, $\eta_p^2 = 0.30$], the Block Design subtest [$F_{(3, 356)} = 24.23$, $MSE = 225.95$, $p < 0.0001$, $\eta_p^2 = 0.17$], and the Digit Span subtest [$F_{(3, 356)} = 37.75$, $MSE = 240.86$, $p < 0.0001$, $\eta_p^2 = 0.24$].

For the Vocabulary subtest, *post-hoc* tests showed that TA had higher mean scores than all the disability groups (TA > MD, TA > RD, and TA > MDRD (all *p*'s < 0.01). The mean difference between MD and MDRD was also significant ($p < 0.01$). For the Block Design subtest, only the mean differences TA > MD, and TA > MDRD were significant ($p < 0.01$).

On the Digit Span subtest, TA had higher mean scores than all the disability groups (TA > MD, TA > RD, and TA > MDRD, all *p*'s < 0.01). MDRD had significantly lower mean scores than MD and RD all *p*'s < 0.01) (For more details see Note in **Figure 4**).

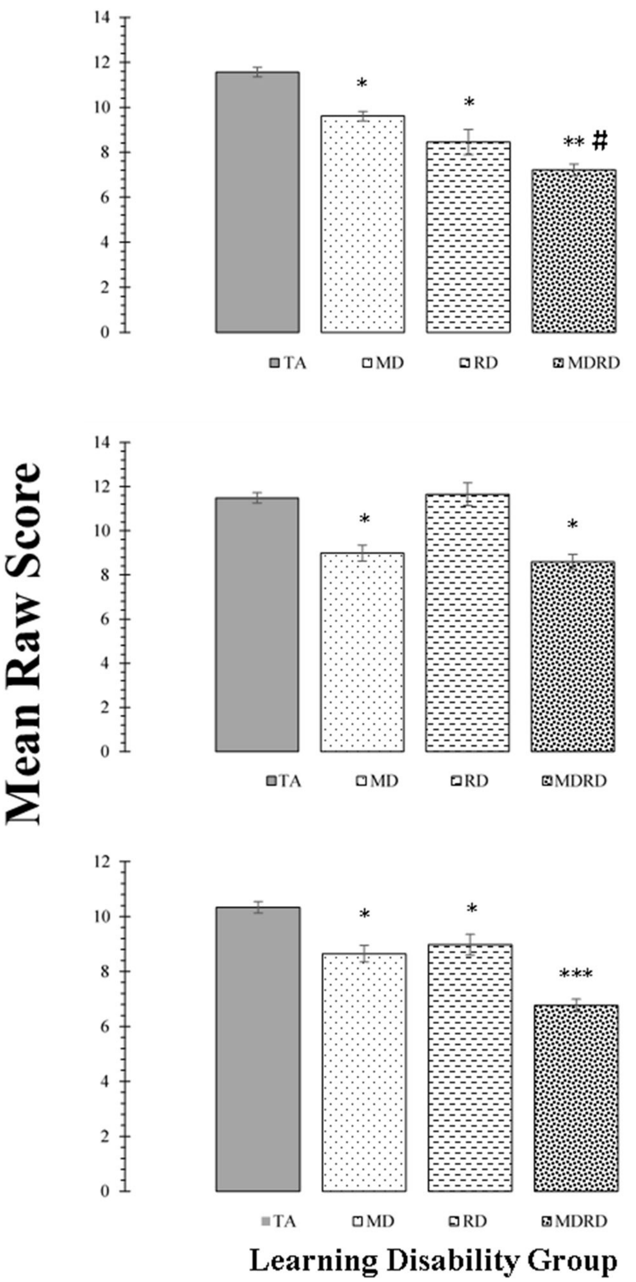

FIGURE 4 | Top Panel: Mean scores for the WAIS-R Vocabulary subtest. Middle Panel: Mean scores for the WAIS-R Block Design subtest. Bottom Panel: Mean scores for the WAIS-R Digit Span subtest. For all panels, bars represent one standard errors. Asterisks summarize the results of *post hoc* Tukey test comparisons: "*" difference from the typical achievement (TA) control group at $p < 0.05$; "**" significant difference from the mathematical disability (MD) group at $p < 0.05$; "***" significant difference from the reading disability (RD) group at $p < 0.05$. The symbol "#" indicates a marginally significant trend at $0.05 < p < 0.10$ on *post hoc* Tukey test. For the Vocabulary subtest: TA > MD (1.95, [CI: −0.92 to 2.98], $p < 0.01$); TA > RD (3.10, [CI: 1.90 to 4.30], $p < 0.01$); TA > MDRD (4.34, [CI: 3.39 to 5.29], $p < 0.01$). MD > MDRD (2.39, [CI: 2.39 to 3.54], $p < 0.01$). # RD > MDRD (1.24, [CI: −0.06 to 2.54], $p = 0.069$). For the Block Design subtest: TA > MD, (2.50, [CI: 1.36 to 3.64], $p < 0.01$); TA > MDRD (2.89, [CI: 1.84 to 3.94], $p < 0.01$). For the Digit Span subtest: TA > MD (1.68, [CI: 0.74 to 2.62], $p < 0.01$);

(Continued)

FIGURE 4 | TA > RD (1.36, [CI: 0.26 to 2.45], $p = 0.01$); TA > MDRD (3.57, [CI: 2.69 to 4.44], $p < 0.01$). MD > MDRD (1.88, [CI: 0.83 to 2.93], $p < 0.01$). RD > MDRD (2.21, [CI: 1.02 to 3.40], $p < 0.01$).

Systematic Review Report: Mapping Reading and Mathematical Disability Deficits to Neuroanatomical Correlates of Dyslexia and Dyscalculia

Table 3 lists all the studies recognized in the systematic review which identified neuroanatomical correlates of dyslexia and/or dyscalculia. It included studies published since 2004 which used either (a) functional neuroimaging to examine the cerebral blood oxygenation in individuals with dyslexia and dyscalculia, who performed during phonological or numerical magnitude tasks; (b) structural neuroimaging to examine white or gray matter tractography in individuals with dyslexia or dyscalculia; (c) lesion-symptom mapping in individuals who suddenly developed alexia (acquired dyslexia) or acalculia (acquired dyscalculia) after suffering a stroke or recovering from surgery to remove a brain tumor. The neuroanatomical correlates of dyslexia and the neuroanatomical correlates of dyscalculia are summarized in **Table 4**.

Neuroanatomical Correlates of Reading Disability Deficits in Dyslexia

The systematic review identified a total of 22 different brain regions whose dysfunction or abnormal development has been associated with impaired reading among individuals with dyslexia and/or alexia. Of those 22 brain regions, only three were identified as neuroanatomical correlates of reading disorders in all four types of research studies surveyed. That is, multiple functional neuroimaging, structural neuroimaging, functional connectivity, and lesion-symptom mapping studies revealed that abnormal activity or development in the *left inferior frontal gyrus*, the *left fusiform gyrus* and the *left angular gyrus* likely contribute to impaired reading.

Individuals with dyslexia exhibit less activation than normal readers at the left inferior frontal gyrus (106–108), left superior temporal gyrus (107, 109–111, 164), left fusiform gyrus (114, 115), and the left angular gyrus (109, 110, 118, 119) when performing the same phonological tasks used to assess in individuals with reading disabilities.

Structural neuroimaging studies have also shown that individuals with dyslexia exhibit reduced gray matter in the left inferior frontal gyrus, fusiform gyrus, and angular gyrus (115). Boets et al. (138) determined that normal readers store phonological representations of words in the left auditory cortex (Brodmann Area 41 and 42), and that a functional connection between the auditory cortex and the left inferior frontal gyrus allows normal readers to access these representations to read more fluently.

Boets and colleagues also used diffusion tensor imaging to show that the left arcuate fasciculus—the bundle of axons that connect the inferior frontal gyrus to the auditory cortex—has

TABLE 3 | Systematic review of the Neuroanatomical correlates of dyslexia and the neuroanatomic correlates of dyscalculia.

Method	Neuroanatomical correlates of dyslexia	Neuroanatomical correlates of dyscalculia
Functional Neuroimaging Studies (fMRI, PET)	**Hypoactivation during phonological tasks**: • Left superior frontal gyrus (105) • Left middle frontal gyrus (105) • ***Left inferior frontal gyrus*** (106–108) • Left superior temporal gyrus (107, 109–111) • Left superior temporal sulcus (107, 109–112) • Left middle temporal gyrus (113) • Left inferior temporal gyrus (108, 109, 114) • ***Left fusiform gyrus*** (112, 114–116) • Left superior parietal cortex (117) • Left inferior parietal cortex (117) • ***Left angular gyrus*** (105, 109, 110, 118, 119) • Left supramarginal gyrus (120, 121) • Left middle occipital gyrus (112) **Hyperactivation during phonological tasks:** • Right medial prefrontal cortex (113) • Left primary motor cortex (109, 113) • Left anterior insula (109, 110, 113) • Left caudate nuclei (109, 110) • Lobule VI of the Left cerebellum (122) • Precuneus (112) No significant differences in activation between individuals with dyslexia and controls • Cerebellum (123) **Null Findings:** No significant differences in activation between individuals with dyslexia and controls during phonological tasks (129)	**Hypoactivation during numerical magnitude tasks**: • Left superior frontal gyrus (124) • Left medial prefrontal cortex (125) • Right fusiform gyrus (124, 125) • ***Right intraparietal sulcus*** (125) • ***Bilateral intraparietal sulci*** (124, 126, 127) **Hyperactivation during numerical magnitude tasks**: • Right superior frontal gyrus (124) • Left postcentral gyrus (124) • ***Left angular gyrus*** (124, 126, 128) • Bilateral supramarginal gyrus (124) **Null findings**: No significant differences in activation between individuals with dyscalculia and controls during numerical magnitude tasks (129)
Structural Neuroimaging Studies (MRI)	**Lower gray matter volume vs. controls**: • Right middle frontal gyrus (130) • ***Left inferior frontal gyrus*** (115, 131) • Left inferior temporal gyrus (115) • ***Left fusiform gyrus*** (115, 132) • ***Left angular gyrus*** (115, 132) • Occipitotemporal cortex (133) **Greater cortical thickness vs. controls**: • Right superior temporal gyrus (134) • ***Left fusiform gyrus*** (134) No difference in gray matter volume between individuals with dyslexia and controls in the following regions • Cerebellum (135)	**Lower gray matter volume vs. controls**: • Left fusiform gyrus (132) • ***Left angular gyrus*** (132) • ***Right intraparietal sulcus*** (136) **Null findings**: No difference in matter volume between individuals with dyslexia and controls (135)
Functional Connectivity Studies (DTI)	**Lower functional connectivity in the following white matter tracts vs. controls**: • Right superior longitudinal fasciculus (137) • Left arcuate fasciculus, connecting ***the left inferior frontal gyrus*** and the left auditory cortex (138) • Reduced connectivity between the ***left inferior frontal gyrus*** and multiple left posterior temporal areas, including the ***left fusiform gyrus***, left inferior temporal gyrus, left middle temporal gyrus, and left superior temporal gyrus (139) • White matter tracts between the right parahippocampal gurus the ***left fusiform gyrus*** (140) • White matter tracts between the ***left angular gyrus*** and left lingual gyrus, as well as the ***left angular gyrus*** and the left cerebellum (105) • Left auditory thalamus and the left planum temporale (141) • ***Left angular gyrus*** and left superior temporal gyrus (123) **Greater functional connectivity between the following structures among individuals with dyslexia vs. controls**: • Left cerebellum and the left supramarginal gyrus (122) • Thalamus and the inferior parietal cortex (117)	**Lower functional connectivity in the following white matter tracts vs. controls**: • Inferior fronto-occipital fasciculus and inferior longitudinal fasciculus, connecting the right fusiform gyrus and ***right intraparietal sulcus*** (145) • Bilateral posterior superior longitudinal fasciculus (146) **Greater functional connectivity between the following structures among individuals with dyscalculia vs. controls**: • ***Bilateral intraparietal sulci*** and the left superior frontal gyrus (147) • ***Bilateral intraparietal sulci***, the right superior temporal gyrus, and the right supramarginal gyrus (148) • Primary visual cortex and inferior occipital cortex (129) • Primary visual cortex and fusiform gyrus (129) **Null findings**: No significant differences between individuals with dyslexia and typical readers in the following white matter tracts: • Bilateral arcuate fasciculus (142) • Corona radiata (142)

(Continued)

TABLE 3 | Continued

Method	Neuroanatomical correlates of dyslexia	Neuroanatomical correlates of dyscalculia
	Null findings: No significant differences between individuals with dyslexia and controls in the following white matter tracts: • Bilateral arcuate fasciculus (142) • Corona radiata (142–144)	
Lesion-Symptom Mapping Studies	**Alexia associated with damage to**: • ***Left inferior frontal gyrus*** (115, 149, 150) • Right Posterior middle temporal gyrus (151) • Right fusiform gyrus (152) • ***Left fusiform gyrus*** (149, 153–155) • ***Left angular gyrus***, via the posterior cerebral artery (156, 157) • Left supramarginal gyrus (158)	**Acquired dyscalculia associated with damage to**: • Left thalamus (159, 160) • ***Left angular gyrus*** (161, 162) • ***Left intraparietal sulcus*** (163)

The ***bolded and italicized*** *brain regions were identified as a neuroanatomical correlate of dyslexia or dyscalculia in all four types of studies in included in the systematic review (functional neuroimaging studies, structural neuroimaging studies, functional connectivity studies, and lesion-symptom mapping studies).*

significantly lower white matter in individuals with dyslexia than in controls, while the structure of the auditory cortex itself was left intact. The authors suggested that individuals with dyslexia develop below average ready fluency in part due to impaired access to phonological representations of words—even words that they are familiar with, exactly as in individuals with reading disabilities. More recent studies have shown the that individuals with dyslexia exhibit lower functional activation than normal readers in the white matter tracts between the left angular gyrus and left lingual gyrus (105) as well as the tracts between the left angular gyrus and left superior temporal gyrus (123).

Finally, lesions to the left fusiform gyrus can result in alexia (50, 149, 153, 154) or from damage to the left posterior cerebral artery, which supplies blood to the left angular gyrus (156, 157). Lesions to the *pars opercularis* section (Brodmann area 44) of the left inferior frontal gyrus can cause alexia, primarily in the form of abrupt deficits in decoding pseudowords (115, 149), whereas lesions to the *pars triangularis* section (Brodmann area 45) are well-known cause in Broca's aphasia (46).

Neuroanatomical Correlates of Mathematical Disability Deficits in Dyscalculia

The systematic review identified a total of 17 different brain regions whose dysfunction or abnormal development has been linked to impaired mathematical cognition among individuals with dyscalculia and/or acalculia. Of these 17 brain regions, converging evidence from multiple sources of evidence suggests that there are only two key neurological structures whose dysfunction may produce distinct and pervasive difficulties with mathematical cognition in individuals with dyscalculia: the *left angular gyrus* and the *bilateral intraparietal sulci*.

Atypical hypoactivation at the right intraparietal sulcus and atypical hyperactivation at the left angular gyrus have been associated with specific aspects of impaired mathematical cognition in individuals with dyscalculia. Individuals with dyscalculia exhibit less activation than controls at the bilateral intraparietal sulci (124–127) when performing variants of the non-symbolic magnitude comparison tasks used to assess the quantitative reasoning skills of individuals with mathematical disabilities. They exhibit greater activation than controls at the left angular gyrus when performing these same magnitude comparison tasks (124, 126).

TABLE 4 | Summary of the neuroanatomical correlates of dyslexia and the neuroanatomical correlates dyscalculia.

Type of research study	Neuroanatomical correlates of dyslexia	Neuroanatomical correlates of dyscalculia
Functional Neuroimaging Studies	Left angular gyrus Left inferior frontal gyrus Left fusiform gyrus	Left angular gyrus Left intraparietal sulcus Right intraparietal sulcus
Structural Neuroimaging Studies	Left angular gyrus Left inferior frontal gyrus Left fusiform gyrus	Left angular gyrus Right intraparietal sulcus
Functional Connectivity Studies	Left angular gyrus Left inferior frontal gyrus Left fusiform gyrus	Left angular gyrus Left intraparietal sulcus Right intraparietal sulcus
Lesion-Symptom Mapping Studies	Left angular gyrus Left inferior frontal gyrus Left fusiform gyrus	Left angular gyrus Left intraparietal sulcus

The brain regions listed here have been characterized as neuroanatomical correlates of dyslexia or dyscalculia in all four types of studies included in this systematic review (functional neuroimaging studies, structural neuroimaging studies, functional connectivity studies, and lesion-symptom mapping studies), with the exception of the intraparietal sulci. The review did not identify any structural neuroimaging studies that characterized the left intraparietal sulcus as a neuroanatomical correlate of dyscalculia, nor did it identify any lesion-symptom mapping studies that characterized the right intraparietal sulcus as a neuroanatomical correlate of dyscalculia.

Few structural neuroimaging studies have investigated the neuroanatomical correlates of dyscalculia. There is some evidence, however, that individuals with dyscalculia exhibit lower gray matter volume than controls in the right intraparietal sulcus (136) as well as in the left fusiform gyrus and left angular gyrus (132).

Individuals with dyscalculia show lower functional connectivity than controls between the right intraparietal sulcus and the right fusiform gyrus (145), and between the parahippocampal gyrus and left fusiform gyrus (140). More recent evidence suggests that individuals with dyscalculia show greater functional connectivity than controls between the bilateral intraparietal sulci and the left superior frontal gyrus (147, 148) as well as between the primary visual cortex and the fusiform gyrus (129).

Finally, lesions to the left intraparietal sulcus (163) or to the left angular gyrus (161, 162) can produce symptoms of acquired dyscalculia. Furthermore, direct damage to left angular gyrus from strokes typically results in *Gerstmann's syndrome*, a disorder characterized by a sudden inability to write (agraphia), an inability to recognize ones' own fingers (finger agnosia), left-right disorientation, and a severe impairment performing mathematical tasks (165).

In summary, the evidence concerning the role of the intraparietal sulci is mixed. The bilateral intraparietal sulci have been reported as correlates of impaired mathematical cognition in multiple functional neuroimaging and functional connectivity studies. However, the right intraparietal sulcus alone has been linked to dyscalculia in structural neuroimaging studies, whereas the left intraparietal sulcus alone has been linked to acalculia in lesion-symptom mapping studies. The left angular gyrus is the only neuroanatomical structure where abnormalities in physiological activation, white matter volume, white matter tractography, and lesions have been consistently identified in individuals with dyscalculia and/or acalculia. Furthermore, the left angular gyrus is the only neuroanatomical structure identified in this systematic review whose dysfunction is also linked to impaired reading in people with dyslexia or alexia.

DISCUSSION

The Cognitive Profiles of Mathematical Disability, Reading Disability, and Comorbid Math and Reading Disability

Table 5 presents a summary of the cognitive profiles of MD, RD, and MDRD participants in this study, organized by their psychoeducational domain. In the reading domain, the RD and MDRD groups demonstrated impaired phonological processing. Similarly, in the math domain, the MD and MDRD groups demonstrated impaired quantitative reasoning on the KeyMath-ID subtest. However, unexpectedly, the RD group also demonstrated a deficit in quantitative reasoning, equal in magnitude to the two math-disabled groups. In the domain-general tests, the MD and RD groups demonstrated impaired verbal semantic memory and working memory; the MD group also demonstrated impaired visuospatial reasoning. Meanwhile, the MDRD group demonstrated additional severe impairments in verbal semantic memory and working memory.

TABLE 5 | The cognitive profiles of the MD, RD, and MDRD participants.

Domain	MD group	RD group	MDRD group
Reading	N/A	• Impaired phonological processing	• Impaired phonological processing
Math	• Impaired quantitative reasoning	• Impaired quantitative reasoning*	• Impaired quantitative reasoning
Domain-general	• Impaired verbal semantic memory • Impaired verbal working memory • Impaired visuospatial reasoning	• Impaired verbal semantic memory • Impaired verbal working memory	• Impaired verbal semantic memory+ • Impaired verbal working memory+ • Impaired visuospatial reasoning

*The symbol *indicates an unexpected finding. The symbol + indicates an additive deficit, where the MDRD group demonstrated a significantly greater deficit than either the MD or RD group.*

The Additive Hypothesis of Cognitive Deficits in Comorbid Math and Reading Disabilities

The present analysis did not reveal an interaction between reading disability and math disability on any of the tests used. Therefore, the results do not support a synergistic or the antagonistic deficit hypothesis, echoing previous findings from children (90) and adults (74). Instead, these results lend further support to the additive hypothesis of cognitive deficits: comorbid learning disabilities are the sum result of separate, specific, underlying cognitive deficits. Importantly, although cross-sectional, the results are similar in children and adult groups. A remaining issue is to understand the mixed bag of domain-specific or domain-general effects.

Domain-Specific Deficits in Comorbid Mathematical and Reading Disabilities

The contrast patterns determined in the univariate analysis only partially correspond with the domain-specific view of learning disabilities. On the three phonological tests, the RD and the MDRD participants both had lower performance scores than the TA participants, exhibiting a considerable and specific deficit in phonological processing. Meanwhile, the MD participants did not exhibit a phonological deficit. This result follows the well-established predictions from a long history of research in dyslexia; it has been shown for several decades that individuals with reading disabilities alone or comorbid reading-and-math disabilities consistently exhibit deficits in phonological processing, frequently making mistakes in applying phoneme-grapheme correspondence rules (28, 29, 31). It is this specific deficit in phonological processing that underlies poor word recognition, and thereby underlies poor reading fluency (6, 23). Consistent with the domain-specific hypothesis, both individuals with reading disabilities and those with the comorbid condition exhibited distinct impairments in phonological processing, but the impairment in individuals with comorbidity was no more severe than in individuals with single disabilities.

However, the results from the KeyMath-ID are not compatible with the domain-specific view of learning disabilities. Against the prediction of that view (70, 166), in our study individuals with reading disabilities showed significant deficits on tests of quantitative reasoning. The results do not support the additive hypothesis of cognitive deficits either, because the deficit demonstrated by the MDRD group did not equal the sum of the deficits in the MD and RD groups. A plausible account for the pattern of results is that successful completion of the

KeyMath-ID relies on higher-level, general-domain skills such as verbal and linguistic reasoning (a conclusion supported by the fact that the KeyMath-ID correlates with tests such as the Kaufman Test of Educational Achievement, the Iowa Tests of Basic Skills, the Measures of Academic Progress test, and Group Math Assessment and Diagnostic Evaluation test).

It must be noted that the KeyMath-ID is not finely tuned enough to measure the very basic ("bottom-up") mechanisms involved in numerical processing. Rather, it can validly identify shared linguistic, comprehension and reasoning deficits in comorbid mathematical and reading disabilities. We acknowledge the limitation in the completeness of our study, since performance on this subtest may not accurately reflect the role played by latent deficits in non-symbolic and symbolic numerical processing. However, despite these limitations, our findings clearly show all three learning disability groups demonstrated challenges in successfully solving the written word problems included in the KeyMath-ID. Importantly, a partial correlation analysis controlling for age and all demographic variables of our sample reveals a strong correlation with the WRAT-R arithmetic subtest (partial $r = 0.78$; $p < 0.0001$), indicating 60% of shared variance with the KeyMath-ID. In addition, cognitive research (167–169) has shown the WRAT arithmetic (which builds up from basic to progressively more complex numerical skills, i.e., from enumeration to pre-algebra) and more in general, arithmetic tasks, indeed tap onto symbolic quantitative skills, partially dependent on symbolic numerical processing [for critical discussion of these relationships, see (75, 78, 79)]. These converging elements therefore indicate, although indirectly, the plausible underlying constraint of number processing on the quantitative reasoning abilities we measured.

Domain-General Deficits in Comorbid Reading and Mathematical Disability

The contrast patterns determined in the univariate analysis of the WAIS-R/WISC-III, Block Design, Vocabulary, Digit Span subtests correspond to a greater extent with the domain-general view of specific learning disabilities. On the Block Design subtest, the MD and MDRD groups both performed far below the typical achievement level, suggesting that individuals with mathematical disabilities experience a pervasive deficit in visuospatial reasoning. When performing a test of visuospatial reasoning such as the Block Design task, the participants make visual approximations about a group of shapes and how they can be rearranged to match a two-dimensional geometric pattern. Such approximations may be negatively influenced by impaired access to the approximate number system—the cognitive mechanism that allows to make estimations about objects and other non-symbolic quantities, and manipulate them in mathematical operations (63). This cognitive system is persistently impaired in individuals with mathematical disabilities (68, 69) and is correlated with impaired perceptual reasoning skills (72). Particularly, neuroimaging studies using the Corsi Blocks task (a test of visuospatial pattern recognition) reported a strong correlation between deficits in visuospatial ability and math disability (73, 170).

In contrast to the MD group, the RD group did not demonstrate a deficit in visuospatial reasoning; remarkably, they scored higher on average than participants in typical achievement range. Individuals with reading disabilities have previously shown to match the performance of non-learning-disability participants on various iterations of this visuospatial reasoning task (44, 90). It has been proposed that reading-disabled individuals may compensate for deficits in phonological processing by relying on visuospatial reasoning to learn, recognize, and articulate words (42, 43). However, while RD individuals have consistently shown faster response times (but not greater accuracy) in identifying impossible figures and manipulate complex shapes with blocks, there is insufficient evidence that they possess an advantage in spatial processing (171).

The most noteworthy findings of the current study were obtained from the WAIS-R Vocabulary and Digit Span subtests. On the Vocabulary subtest, the MD group and the RD group performed at a statistically equivalent level; both groups scored significantly lower on average than the typical achievement range, implying that both groups experienced a specific difficulty in articulating age-appropriate word definitions. This was an unexpected finding for the MD group in particular; individuals with math disabilities are not known to demonstrate difficulty in word recognition, especially when tested with familiar words. The difficulty demonstrated by the RD group was also unexpected, because the Vocabulary subtest does not directly assess phonological processing. The participants did not need to read any of the items (they were read by a test administrator), nor were any of the words considered to be too irregular for their age-appropriate lexicon (the WISC included common words such as "clock," or "alphabet"). Given that the between-group differences in education rating were insignificant and none of the participants had an estimated IQ within the clinically-critical range, these findings were unexpected.

It was originally hypothesized that individuals with comorbid math and reading disabilities would not exhibit additive deficits on the Vocabulary subtest because it does not exclusively assess domain-specific phonological processing or arithmetic abilities. The MDRD participants performed at an even lower level than both the MD and the RD group. While the mean difference between the RD and MDRD was marginally significant ($p = 0.069$), the difference in mean scaled scores between the TA and MDRD groups (4.34) was nearly the sum of the difference between the TA and MD groups (1.95) and the difference between the TA and RD groups (3.10), corresponding to the additive hypothesis. In stark contrast, the individual deficits shown by the MD and RD groups and the additive deficits exhibited by the MDRD group suggest that an impairment to some other domain-general cognitive system mediates their ability to articulate definitions of common words.

The groups' performances on the Digit Span subtest followed a similar pattern to the one seen for the Vocabulary subtest. The MD and RD groups performed at a statistically-even level—both groups significantly lower than the typical achievement range—indicating that these participants experienced a specific deficit in verbal working memory. Just as in the Vocabulary subtest, the

MDRD group performed at even lower level on average than the single-disability groups. In support of the additive hypothesis, the mean difference in scores between the TA and MDRD groups (3.57) was slightly more than the sum of the difference between the TA and MD groups (1.68) and the difference between the TA and RD groups (1.57); as described in section The Cognitive Profiles of Mathematical Disability, Reading Disability, and Comorbid Math and Reading Disability, this over-additivity was not the result of a significant interaction between math disability and reading disability, and therefore does not fit the synergistic hypothesis. These results suggest that individuals with math disabilities or reading disabilities alone present a specific, persistent impairment to their verbal working memory system, and that this impairment is even more pronounced in individuals with comorbid math and reading disabilities in a manner that is both statistically and clinically significant.

The verbal working memory deficit reported in the RD and MDRD groups was consistent with evidence from previous studies; individuals with reading disabilities consistently demonstrate impaired working memory (33, 81, 172, 173). The deficit demonstrated by the MD group, however, was not expected. Previous investigations about the relationship between impaired mathematical cognition and impaired working memory have yielded mixed results. In some studies, individuals with math disabilities exhibit deficits in working memory only when performing tasks that assess working memory and visuospatial reasoning simultaneously (68, 170); the Block Design subtest we used does not test these constructs simultaneously, because the prompt remains visible to the participant at all times (thereby remaining accessible in the participant's short-term sensory memory).

In other studies, individuals with math disabilities do not exhibit any significant deficits relative to controls when performing forward or backward recall tasks (174); but when wider selection criteria are used to classify participants into control and MD groups (i.e., below the 30th percentile on a standardized arithmetic test), the MD participants lag behind controls on memory span (175, 176). Given this contradicting evidence, we did not expect that the MD participants in the current study would demonstrate a specific working memory impairment; instead, the MD group did demonstrate pervasive difficulties in performing the working memory task.

Furthermore, the MDRD group—who was expected to exhibit a working memory deficit as seen in individuals with dyslexia—performed significantly poorer than both the MD and RD groups. Reminiscent of the pattern seen on the WAIS-R Vocabulary subtest, the difference in mean scaled scores between the TA and MDRD groups (2.21) was approximately even to the combined sum of the difference between the TA and MD groups (1.88) and the difference between the TA and RD groups (0.83). This provides further support to the additive hypothesis. An additive working memory deficit in MDRD participants has been previously reported in adults (74).

In contrast, Landerl et al. (90) reported that children with a math disability or reading disability alone did not exhibit significant deficits in working memory, but those with comorbid math and reading disabilities did—only on the backward-recall trials (and not the forward-recall trials) of the WISC-III Digit Span subtest. They found a significant interaction between math disability and reading disability on the comorbid group's mean scores on the backward Digit Span subtest, a result they interpreted as supporting the synergistic deficit hypothesis, but one that was not replicated in the current study. While it is unlikely that a deficit in working memory is the mechanism that underlies all forms of impaired numerical magnitude processing (67), again, the current results show that such is the case in linguistically-mediated math disability, since two separate, independent impairments to numerical cognition and reading ability contribute to a persistent, severe working memory deficit unique to individuals with comorbid math and reading disabilities.

A Neuroeducational Model of Comorbid Mathematical and Reading Disabilities

The performance of the MDRD participants on the psychoeducational tasks in this study revealed two specific cognitive functions that are more severely impaired in comorbidity than in the single disability (see Bottom Panel of **Figure 5**). As expected, the MDRD group presented domain-specific deficits equal to those shown by the single-disability groups; their scores were on par with the RD group on two of the three phonological tasks, and also matched the performance of the MD group on the test of quantitative reasoning. More importantly, the MDRD participants exhibited domain-general deficits in verbal semantic memory and verbal working memory. In both the semantic memory task and the verbal working memory task, the magnitude of the MDRD group's deficit was approximately the sum of the separate deficits demonstrated by the MD and RD groups. These findings lend strong support to the additive deficit hypothesis of an independent math disability and an independent reading disability combined to produce a greater deficit in semantic memory and a larger deficit in working memory—deficits that were greater in magnitude than those observed in the participants with a single disability.

The model proposed in **Figure 5** suggests that the left angular gyrus may be the key neurological structure that mediates the cognitive deficits uniquely expressed in both math and reading disabilities in both children and adults (see Top Panel of **Figure 5**). Previous studies have shown that atypical function of the left and angular gyrus is associated with deficits in phonological processing, numerical cognition, and working memory—all of which were exhibited by the MDRD participants in the current study.

Five strands of evidence form the basis for this hypothesis. First, functional neuroimaging studies have consistently identified the left angular gyrus as a pivotal structure in mediating word recognition, word decoding, and reading comprehension in normal readers (110, 177, 178). Individuals with reading exhibit less activation than normal readers at the left angular gyrus during tasks that involve effortful word decoding (such as the Word Attack subtest in this study), and also show decreased activation during semantic processing of visual and auditory words (179–181). In comparison to controls, individuals with dyslexia show atypical hypoactivity at the left angular gyrus while performing phonological tasks (55, 56) as well in individuals with dyscalculia while performing tasks that

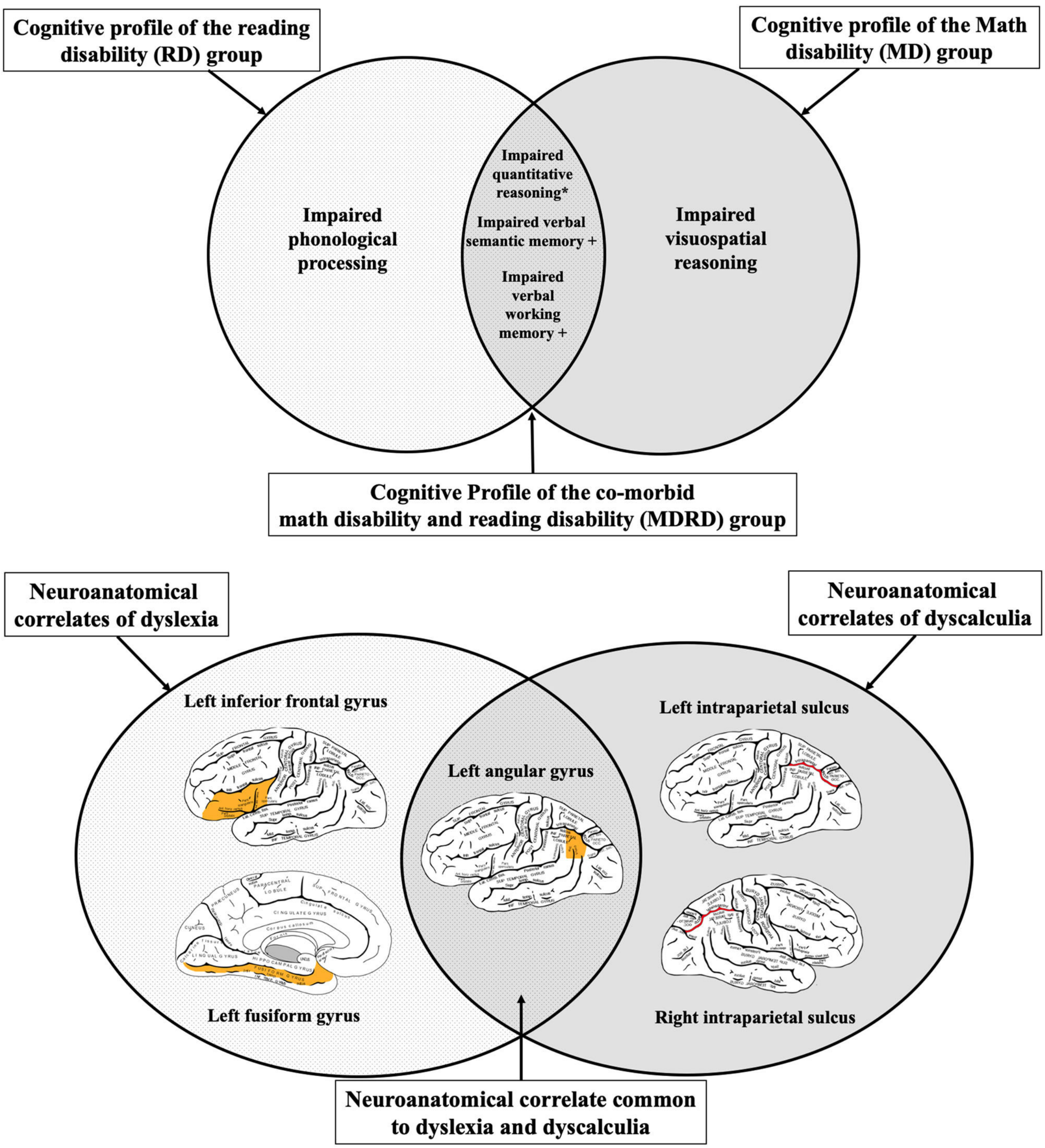

FIGURE 5 | A neuroeducational hypothetical model of dyslexia-dyscalculia comorbidity. Top Panel: The neuroanatomical correlates of dyslexia and dyscalculia as determined by the systematic review. The left angular gyrus—a brain region where atypical physiological function has been reported in both individuals with dyslexia and individuals with dyscalculia. Bottom Panel: The cognitive profiles of individuals with reading disability (RD), math disability (MD), or comorbid math and reading disability (MDRD) in the current sample. The symbol "+" indicates an "additive deficit", where the MDRD group demonstrated a significantly greater deficit than either the MD or RD group. The symbol "*" indicates an unexpected finding with novel implications for RD and MDRD; see Discussion. Brain diagrams were adapted from (45). Anatomy of the Human Body. Retrieved from https://www.bartleby.com/107/189.html.

test the approximate number system (84, 182). Second, lesions to the posterior cerebral artery (the blood supply of the left angular gyrus) produces symptoms of alexia (157), while lesions to the gyrus itself can produce acquired dyscalculia, alongside other symptoms of Gerstmann's syndrome (183).

Third, the left angular gyrus has been shown to mediate the two aspects of memory retrieval: the retrieval of phonetic representations of familiar words (138) and the retrieval of arithmetic facts (85). The latter demonstrates a contrast in the type of calculations which involve differentially the right and left hemisphere; the right intraparietal sulcus is associated with greater activation for processing inexact quantities for calculations involving the approximate number system, while the left angular gyrus and left intraparietal sulcus are involved in retrieving exact quantities for calculations that require a single exact solution. In addition, the left angular gyrus has shown low functional connectivity with the inferior frontal and left fusiform gyri during phonological processing tasks (177, 184).

Fourth, impairments to subcomponents of working memory can be mapped to the disruption frontal-lobe-to-parietal-lobe association fibers, converging on the left angular gyrus. According to Baddeley's model of Working Memory (185), a subcomponent of working memory called the phonological loop facilitates the short-term storage encoding verbal information into long-term memory. In Positron Emission Tomography (PET) studies with normal readers performing pseudowords tasks similar to the psychoeducational tests used here, researchers have shown that the angular gyrus is directly active in facilitating the short-term storage and retrieval of unfamiliar phonemic sequences (173, 186).

Lastly, applying TMS (Transcranial Magnetic Stimulation) to the left angular gyrus has been shown to increase the accuracy of semantic memory when pairing stimuli for classical conditioning (187), but can also cause deficits in visuospatial reasoning (mainly right-left disorientation). This collection of evidence from functional imaging, lesion-symptom analysis, and neuropsychology research provide valuable insight into the unique role of the left angular gyrus in mediating the cognitive deficits shared in math and reading disabilities.

The MDRD participants demonstrated additive domain-general deficits in verbal working memory and verbal semantic memory, which suggests a functional relationship between the verbal, expressive component of these deficits, and the retrieval of symbolic and semantic representations of words from long-term memory. A possible explanation is that a pervasive deficit to the verbal working memory system (specifically the phonological loop subcomponent in Baddeley's model) impairs both the short-term storage of basic verbal information and the retrieval of semantic information from long-term storage. The model shown in the Bottom Panel of **Figure 5** proposes that a pervasive deficit in the temporary storage of verbal information is an additional core deficit unique to individuals with comorbid dyslexia-dyscalculia, associated with atypical function of the left angular gyrus.

CONCLUSIONS

In summary, converging evidence from neuroimaging and psychoeducational research suggests that impaired phonological, numerical, semantic, and working memory processes may be related to dysfunction of the left angular gyrus in individuals with dyslexia and individuals with dyscalculia. Individuals with dyslexia exhibit atypical hypoactivation at the inferior frontal gyrus when performing the same psychoeducational tests that are used to assess phonological processing deficits in individuals with reading disabilities. Individuals with dyslexia also exhibit atypical hypoactivation at the visual word form area of the left fusiform gyrus (located on the sagittal surface of the temporal lobe) when viewing written words, and consistently demonstrate impaired word recognition. These cognitive profiles are similar to those seen in the present study, where individuals with reading disabilities demonstrated significant deficits on tests of vocabulary and reading fluency. Similarly, individuals with dyscalculia exhibit atypical hypoactivation of the bilateral intraparietal sulci when performing numerical magnitude processing tasks, akin to the psychoeducational tests used to assess the function of the approximate number system in individuals with mathematical disabilities.

Multiple sources of evidence show that individuals with dyslexia and individuals with dyscalculia both exhibit dysfunction of the left angular gyrus. The left angular gyrus is the only neuroanatomical region where abnormalities in physiological activation, white matter volume, and white matter tractography have been reported among individuals with dyslexia and individuals with dyscalculia in functional neuroimaging, structural neuroimaging, and functional connectivity studies. Individuals with dyslexia exhibit atypical hypoactivation at the left angular gyrus when performing word definition recall tasks, demonstrating the same deficits in semantic memory that are seen in individuals with reading disabilities when performing the WAIS-R Vocabulary task. Individuals with dyscalculia exhibit atypical hyperactivation at the left angular gyrus when retrieving arithmetic facts, presenting similar difficulties as individuals with math disabilities when performing psychoeducational tests that assess basic math fluency. Furthermore, the left angular gyrus is the only neuroanatomical region where lesions have been identified in individuals with alexia as well as individuals with acalculia. The individuation of such area as the main overlapping brain structure associated with reading-math comorbidity makes sense anatomically since meta-analytic evidence suggests its role is to specifically associating language with other types of information and could be regarded as a language processing marginal area participating in an "extended Wernicke's area" or "Wernicke's system" (188).

Based on past and present evidence, we conclude that children and adults with comorbid math and reading disabilities demonstrate domain-specific deficits equivalent to single-disability individuals. However, individuals with comorbidity also demonstrate additive domain-general deficits in verbal working memory and verbal semantic memory. According to the evidence gained from the exhaustive review of the literature, the domain-specific reading deficits may correspond to developmental differences in the left inferior frontal and fusiform gyri, and mathematical deficits can be traced to developmental differences in the bilateral intraparietal sulci. The current model proposes that domain-general, additive deficits in semantic memory and verbal working memory—two pervasive impairments that are unique to neuropsychological profile of individuals with comorbid math and reading difficulties—may be the result of atypical development of the left angular gyrus.

Looking ahead, most recent advances in the emerging field of neuroimaging genetics have revealed bilateral interplay between genetic profile and neurological function and sometimes even a trilateral interaction between the latter and neuroanatomy. These studies have primarily focused on dyslexia (189–192), but similar evidence is also emerging for dyscalculia (193–195) and first evidence has appeared for their comorbidity (132). Although exhaustive review is beyond the scope and space of this paper (for very recent reviews see (193, 196), our model and research synthesis is complementary and could be further probed with neuroimaging genetics, specifically, in the pursuit of precise early detection of comorbid reading and math disability well before they emerge in formal educational settings. While genetic profile is insufficient to fully explain a complex condition such as dyslexia-dyscalculia comorbidity, some genetic constellations, or endophenotypes, can clearly help in early diagnosis (197). Because early intervention might be crucial to address this complex severe condition, genetics might be an instrument helping to close or at least narrow the diagnostic gap in the early years of life, before schooling. The present synthesis and model could be a starting point for the timely identification of reliable endophenotypes linked with early development of the angular gyrus and later psychoeducational diagnosis of reading-math disability comorbidity.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available upon reasonable request in a form being coherent to all institutional, national and international ethical standards (e.g., anonymization), including intellectual property guidelines.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Research Ethics Board of The University of British Columbia. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

JG curated database, designed and conducted main analyses, and wrote the basis of the first draft. LS designed and led the prospective cohort study and database, obtained funding, and revised draft. AD'A collected data, helped design the study, helped with database curation and management, helped with data analysis, obtained further funding, oversaw writing of the final manuscript from first to final draft, and edited into final manuscript. All authors contributed to the article and approved the submitted version.

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Impact of a Combined Philosophy and Mindfulness Intervention on Positive and Negative Indicators of Mental Health Among Pre-kindergarten Children: Results From a Pilot and Feasibility Study

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Background: Fostering greater resiliency to stress, optimal psychosocial development and promoting better mental health and well-being in youth is an important goal of the Canadian and American elementary school systems (1, 2). Recent research on mindfulness and philosophy for children (P4C) has yielded promising results regarding innovative interventions that may be implemented in elementary school settings to foster greater child resiliency and well-being (3–5).

Goal: The goal of this feasibility study was to pilot a new intervention, which combines mindfulness meditation and P4C activities, with the goal of improving mental health in pre-kindergarten children, assessed with positive (i.e., social skills and adaptability) and negative (i.e., internalized symptoms, comprises depression, anxiety, inattention; and hyperactivity) indicators.

Methods: A randomized cluster trial with a wait-list control group was employed to evaluate the impact of the combined MBI and P4C intervention on child mental health. Two classrooms of pre-kindergarten children ($N = 38$, mean age $= 4.6$ years old) took part in this study and were randomly allocated to the experimental or wait-list control conditions. Teachers completed pre- and post-intervention questionnaires.

Results: ANCOVAs did not reveal a significant effect of condition on internalized symptoms, controlling for baseline levels. Sensitivity analyses indicated that for the whole sample, internalized symptom scores were statistically significantly lower at post-intervention, when compared to pre-intervention scores. No impact of group on levels of hyperactivity was found, however, sensitivity analyses indicated that for both the experimental and control groups, hyperactivity scores were statistically significantly lower at post-intervention, when compared to pre-intervention scores. Finally, no impact of group on levels of social skills and adaptability were found. Sensitivity analyses conducted using paired t-tests did not indicate statistically significant pre-to-post changes in scores for both variables.

Conclusion: These preliminary results suggest that mindfulness and philosophy for children may not be the most effective intervention to foster short-term resiliency, well-being and better mental health in children. Yet, group differences were often small and past research suggested the effectiveness of this type of intervention. Further research considering the impact of moderators such as age or baseline levels of psychopathology, using longer time frames and comparing the effectiveness of this combined intervention with other types of school-based interventions with similar aims (such as, e.g., P4C or MBI alone) is warranted, to evaluate if mindfulness and P4C interventions have an added value compared to other types of interventions implemented in school settings.

Keywords: preschool, school psychology, mental health, mindfulness-based interventions (MBIs), philosophy for children (P4C)

INTRODUCTION

Fostering greater resiliency to stress, optimal psychosocial development and promoting better mental health and well-being in youth is an important goal of the Quebec (Canadian province) and American elementary school systems (1, 2). In Canada, 20% of elementary school children have received a diagnosis of a psychological disorder (6). However, only a fifth of youth who need help actually have access to mental health services (7). Given the high prevalence of psychological difficulties in elementary school children, it appears crucial to develop interventions that can help them foster greater resiliency, better mental health and well-being. Resiliency is comprised of two essential components, which are (1) the presence of a significant risk/adversity (e.g., for one's own mental health); and (2) a positive adaptation in spite of this risk/adversity (8). Resiliency is considered to be a dynamic process, which results from interactions between personal attributes (e.g., emotional regulation skills, well-being, social skills, perseverance) and environmental risk and protective factors (8, 9).

In order to fully understand the concept of mental health, researchers have recently suggested to conceptualize it as the result of both positive (e.g., well-being and positive affect) and negative indicators (e.g., symptoms pertaining to psychopathology and negative affect) (10). Developing prevention strategies and interventions has been recommended as a potential solution to reach a greater number of students and to prevent psychological disorders, while fostering greater well-being (11). Recent research on mindfulness and philosophy for children has put forth promising results regarding innovative school interventions that may foster greater resiliency and well-being in elementary school children (3–5).

MINDFULNESS-BASED INTERVENTIONS FOR YOUTH

In recent years, mindfulness-based interventions (MBIs) specifically designed for youth have received significant research attention (5, 12–14). Mindfulness can be defined as the process by which we bring our attention to the experience unfolding in the present moment, intentionally and without judgement (15). Results have shown that MBIs can significantly alleviate psychological distress and foster greater well-being and resiliency, namely in school settings (16–20). As such, MBIs could represent an interesting intervention to implement in school settings, namely because of their ease of implementation (21). MBIs typically consist of short exercises that are easily integrated within the classroom routine. These exercises are meant for students to develop their ability to pay attention and become aware of their sensory experiences and to recognize their thoughts and emotions, in order to become better at regulating them. Past research on MBIs in school settings has shown encouraging results with regards to decreasing various internalized (e.g., anxiety, depression, inattention) and externalized (e.g., aggressiveness, conduct disorders, hyperactivity) symptoms, while increasing well-being and resiliency both in regular education classrooms and special education classrooms (5, 12–14, 16–20, 22).

SELF-DETERMINATION THEORY AND MINDFULNESS

To date, there is a paucity of theoretical models explaining why and how MBIs have a positive impact on youth's well-being (21). In recent years, researchers have suggested that mindfulness and its effects on well-being could be best understood through self-determination theory [SDT; (23)]. SDT is a macro-theory of well-being and functioning that posits that we all have three basic psychological needs that are essential to well-being: the need for autonomy (the need to act with volition and in accordance with our interests and values), the need for competence (the need to have an impact on our environment) and the need for affiliation (feeling connected to others and loved by others). When measuring children's well-being, it is important to take into account the evaluations of relevant stakeholders in children's lives, but also children's own evaluations, and aspirations regarding their own lives. Basic psychological need satisfaction is a widely researched determinant of well-being among children. More than three decades of research with children show that the satisfaction of the basic psychological needs of autonomy, competence and relatedness in children is key for children's academic achievement, perseverance and life satisfaction, all of which have been included in various conceptualizations of child well-being (24). Hence, as SDT research will detail, during this early developmental period supports for relatedness, autonomy, and competence are required for infants and young children to be intrinsically motivated, to attach to others and form secure social bonds, and to integrate social regulations into their self-regulatory capacities, all processes essential to adaptation and thriving in "cultural animals" such as humans (25). Yet their importance goes beyond these early developmental issues, to bear on wellness, relationship qualities, experience, and quality of behavior in virtually every domain and at all ages across the lifespan.

The need for autonomy (also referred to as self-determination) is a cornerstone of SDT, and, as such, research has placed significant importance on the need for youth and adults alike to act volitionally and in accordance with their values. Research with adults has shown that mindfulness is positively correlated with self-determined motivation, which can be defined as doing an activity for the sake and pleasure of it or because it's personally meaningful and related to one's core values. Recently, mindfulness has been proposed as an antecedent of self-determined motivation (26), and evidence from correlational and priming studies shows that people who are more mindful report more self-determination in their day-to-day activities and choose to act in more self-determined ways (27–29). Several studies have even identified self-determination as a key explanatory mechanism in the relationship between mindfulness and well-being (29–32). Moreover, (33) showed that dispositional mindfulness increased well-being through the mediating role of self-determined goal motivation. Furthermore, initial research on students indicates that dispositional mindfulness was positively associated with self-determined motivation (34). However, these studies were mainly correlational and only focused on dispositional mindfulness as a predictor.

Recently, results from pilot studies using experimental designs have shown encouraging results as to how MBIs could be useful tools to foster self-determination in youth (35). Specifically, MBIs could help youth better identify and accept their personal values, strengths and weaknesses, thus leading to the construction of a more realistic identity and higher levels of self-determination and well-being (36). Furthermore, decades of research in SDT have indicated that higher self-determination is linked to perseverance and academic achievements in elementary and high school students (37, 38). However, results from (35, 39) study cited above, although encouraging, also indicated that the MBI was not more useful than a social skills development program to increase self-determination, which suggests that a MBI alone may be necessary, but not sufficient to significantly and undoubtfully improve basic psychological needs satisfaction in children. As such, we posit that the combination of mindfulness techniques with existential psychology (through philosophy for children activities, detailed below) could have a stronger impact on basic psychological needs satisfaction in elementary school students, particularly self-determination. Further explanation is provided below.

EXISTENTIAL PSYCHOLOGY

Issues pertaining to self-determination, such as living a life in accordance with one's beliefs and values, are also central to existential psychology (40, 41). Existential psychology is a branch of clinical psychology in which individuals are encouraged to explore and discuss core existential questions and struggles that are shared by all humans. One goal of existential therapy is to bring the client to re-focus on the present and to notice what is important to them in the present. This renewed awareness is thought to help individuals become more self-determined and motivated to act in accordance with their values (42). Although existential psychology is different from SDT, within both theoretical frameworks, an individual's self-determination is central and can be achieved through increased awareness of one's own strengths, weaknesses, and other factors that can help or hinder self-determination, all of which can be facilitated through MBIs (43). Indeed, according to (43), SDT and existential psychology (as well as existential philosophers) share common grounds with regards to ideas related to authentic and inauthentic actions. Authentic actions can hence be seen as "endorsed by the self" and congruent with one's values and sense of self. Furthermore, Deci and Ryan (43) establish links between Kierkegaard's view of the self and SDT: "…to achieve a self is to be committed to relate the self to the self, of taking responsibility for ever reevaluating what one believes, and then acting in accord with that best synthesis. In this view, a genuine human being is "infinitely interested in his existence," and what one does is the current best synthesis of all that one truly believes, knows, and feels."

However, unlike research on SDT and mindfulness, evidence-based research within existential psychology for youth is almost inexistent (44). To our knowledge, no study to date has been published detailing the association and possible impact of an existential psychology intervention for children on well-being. Thus, one goal of this study was, to develop and present an adaptation of an intervention based on existential psychology for children, while integrating it with the SDT and mindfulness research frameworks in school settings. This was done in order to provide children with an intervention that is better suited to foster their self-determination. Indeed, as MBIs help individuals become more aware of their values through an increased awareness of everyday thoughts, emotions and sensations, existential psychology (through the implementation of philosophy activities for children) was added so that children could directly and actively reflect on their personal values, facilitating the process of identifying them. Integrating mindfulness and existential psychology could thus provide children with a more effective intervention to foster self-determination, as defined by SDT. More concretely, by practicing mindfulness exercises, a child could start noticing that their throat gets tight when they are in the presence of a classmate whom they call a friend, or that they feel warm and tingly when they think about their pet. This could help them become more aware of what is important to them (e.g., animals, being with people they love). In turn, this knowledge could foster a much deeper reflection and integration for them when they engage in the philosophy activities.

PHILOSOPHY FOR CHILDREN (P4C)

The roots of existential psychology can be traced back to philosophy, which has aimed for centuries to provide answers to humans' need to make sense of the world (42). Similarly, children, as much as adults, strive to make sense of the world surrounding them (45). Philosophy for children (P4C), centered on the practice of dialogue, aims to develop children's capacity to think by and for themselves and to become more self-determined, providing some answers to questions children may ask themselves (46–48). As such, P4C aims primarily to provide an educational context in which children are asked to think by and for themselves about topics that are of interest to them, fostering greater intellectual and emotional autonomy, and consequently, higher self-determination.

Such aims converge toward the prospective and utopic functions of philosophical dialogue as a practice, broadly defined as the capacity for students to understand themselves as subjects acting upon the world. Some educational researchers link this to the concept of agency or to the idea that attitudes, methods, and cognitive processes subsumed by the philosophical dialogues contribute to pupils' recognition of the power of individual and collective agency to act upon the present and to mold the future. They are likely to lead pupils to see themselves as the subjects of a complex history, influenced, at least partly, by their thoughts and actions through the practice of dialogue (49, 50).

P4C is becoming increasingly popular in school settings, namely to foster greater well-being in youth (47). Its practice aims to foster greater awareness of thoughts and emotions, and, as such, shares common goals with mindfulness (51, 52). During P4C activities, students are presented with an existential or philosophical theme (while reading a story, viewing a short video, or choosing specific themes amongst themselves) (53). Following the initial philosophical primer, students are invited to identify, as a group, topics they wish to discuss (53–56). In this philosophical process, pupils are expected to learn to question, conceptualize and problematize natural or human phenomena, as well as to deconstruct, rationally weigh, take position on and tolerantly discuss thoughts related to existential, ethical, esthetical, epistemological, etc., controversies.

There is a paucity of evidence-based, experimental results on the impact of P4C activities on mental health in children (52). Available correlational and quasi-experimental results indicate a positive association between P4C, cognitive and academic skills (57, 58), self-esteem, and the ability to identify one's own emotions and thoughts in elementary school children (59). Upon researching the literature on this topic, only two quasi-experimental studies documenting the association between P4C techniques and children's well-being were found. One study among children displaying various emotional and behavioral disorders (including, but not restricted to, autism spectrum disorders, and communication disorders) in a special education curriculum reported benefits in self-regulatory (namely turn-taking and patience) and overall participation in class in these participants (60). Further work by the same group of authors showed similar results on self-regulation and rule adherence among children with severe behavioral disorders receiving special education in secure settings (61). While these correlational results provide preliminary evidence for the use of P4C activities in classrooms, to our knowledge, no experimental study has been published on the impact of P4C on mental health and well-being in children. Furthermore, there is no available, published evidence of the influence of P4C on self-determination and basic psychological needs satisfaction. One goal of this study was thus to implement an experimental design to document the impact of P4C in pre-kindergarten children using a randomized controlled trial and to evaluate if P4C had an impact on self-determination in children.

Present Study

Based on the complementarity between existential psychology and mindfulness interventions for youth, we designed an intervention combining a MBI and P4C activities for pre-kindergarten children. The main objective was thus to conduct a pilot feasibility study exploring the effectiveness of an intervention combining mindfulness-based and P4C activities. We evaluated the impact of this combined intervention on positive (i.e., social skills and adaptability) and negative (i.e., internalized symptoms: depression, anxiety, inattention; and hyperactivity) indicators of well-being using an experimental cluster design with a wait-list control condition.

Hypotheses

Based on previous findings from the literature on MBIs in youth and preliminary literature on the impact of P4C, we hypothesized that the combined intervention would have a beneficial impact on participants' psychological health and well-being. Specifically, we hypothesized children from the experimental group would experience increases in positive indicators of well-being (i.e., better social skills and adaptability) from pre-to-post intervention. These increases would be greater than those observed in children from the control group. We also hypothesized that children from the experimental group would experience decreases in negative indicators of psychological health (i.e., internalized symptoms) from pre-to-post intervention, and that these decreases would be greater than those observed in children from the control group.

METHODS

Two classrooms of pre-kindergarten children ($N = 38$, mean age = 4.6 years-old) from a private elementary school in the Eastern Townships area (Quebec, Canada) took part in this study and were randomly allocated to an experimental ($n = 19$ students) or a wait-list control condition ($n = 19$ students).

TABLE 1 | Descriptive statistics and sample distribution.

	N	Mean age	Condition
Experimental group	19	4.6	MBI and P4C
Control group	19	4.7	Wait-list
Total sample	38	4.68	
Girls	23		
Boys	15		

Descriptive statistics can be found in **Table 1**. A randomized cluster trial with a wait-list control group was implemented to evaluate the impact of the combined MBI and P4C intervention on positive and negative indicators of well-being. Teachers completed measures at pre-intervention and immediately post-intervention. There was no attrition in this study. After the intervention was completed with the experimental group, wait-list controls were offered the intervention. Informed consent was obtained from all parents of students taking part in this study, as well as from both teachers participating. All parents gave consent for their child to participate in the project.

Procedure

A 5-weeks intervention was implemented as part of this project. Specifically, each weekly session comprises a mindfulness opening activity, followed by a P4C activity. Each session lasted ~1 h. Mindfulness activities comprised in this intervention were: (1) an introductory activity on mindfulness meditation and related psychoeducational content about thoughts, physical sensations and emotions; (2) mindful eating; (3) mindful pausing; (4) mindful listening; and (5) mindfulness and gratitude. The mindfulness activities were adapted from the *Mission Meditation* manual, an evidence-based intervention adapted and tailored to fit elementary school children's developmental needs and attention span (4). P4C activities comprised in this intervention were based on themes of: (1) happiness; (2) normal vs. not normal; (3) making mistakes; (4) sadness and anger; and (5) separation and death. These themes were selected as they were most closely related to existential psychology and were thought to foster self-determination more directly in children. Students were presented with short stories, simple questions (often accompanied by an image or poster to illustrate the question), short comic strips and video clips. The intervention was led by two undergraduate students in psychology with extensive training in MBIs and P4C. Both were supervised on a weekly basis by the research team throughout the course of this project.

Measure

Given the age of the participants in this study, teachers were asked to complete the pre-intervention and immediately post-intervention questionnaires. Specifically, teachers were asked to report on children's anxiety (three items, e.g., Worries about things that cannot be changed), depression (three items, e.g., Is sad), inattention (three items, e.g., Has a short attention span), hyperactivity (three items, e.g., Has trouble staying seated), social skills (three items, e.g., Encourages others to do their best), and adaptability (three items; Adjusts well to changes in routine;

TABLE 2 | Means and standard deviations for negative and positive indicators of well-being (scores on the BASC-III subscales).

	Control group		Experimental group	
Dependent variable	**Pre-test (SD)**	**Post-test (SD)**	**Pre-test (SD)**	**Post-test (SD)**
Negative indicators				
Internalized symptoms	9.29 (3.84)	7.88 (4.74)	9.61 (2.03)	8.55 (2.61)
Hyperactivity	3.37 (1.77)	2.26 (2.08)	4.08 (1.78)	3.55 (1.64)
Positive indicators				
Social skills	2.11 (2.35)	3.16 (2.17)	4.16 (0.94)	3.66 (1.07)
Adaptability	3.21 (1.47)	3.74 (1.33)	4.89 (0.81)	5.00 (1.27)

Is easily calmed when angry; Seems to take setbacks in stride) using the respective subscales of the *Behavior Assessment Scale for Children* (BASC III) (62). The teacher-report form was used to evaluate the presence of internalized and externalized symptoms in children. Teachers were asked to rate their agreement on a 4-point Likert-type scale (1–never to 4–always) and subscale scores were summed. For the purposes of the present study, depression, anxiety and inattention were grouped to represent overall changes in internalized symptoms. Internal consistency was acceptable to good for the majority of subscales in this sample ($\alpha_{\text{adaptability}} = 0.78$; $\alpha_{\text{social skills}} = 0.69$; $\alpha_{\text{hyperactivity}} = 0.80$; $\alpha_{\text{internalized symptoms}} = 0.81$).

Data Analysis

Hypotheses were tested using ANCOVAs, as they have been recommended to increase statistical power in randomized controlled trials (63). In this study, ANCOVAs allowed the comparison of post-intervention scores across groups, while controlling for pre-intervention scores. *Post-hoc* analyses were conducted using paired t-tests, to evaluate pre-to-post differences in scores within each group and examine changes over time. Effect sizes were also computed in order to assess the magnitude of the observed effects.

RESULTS

Preliminary analyses using independent *t*-tests first showed that the two groups differed at pre-intervention on measures of social skills [$t_{(37)} = -3.60$, $p = 0.001$] and adaptability [$t_{(37)} = -4.49$, $p < 0.001$]. In other words, children from the experimental group had higher levels of social skills and adaptability than the children from the control group. Descriptive results can be found below and in **Tables 2**, **3**. Data assumptions for normality (64), homogeneity of variance (65), and sphericity were all met in this sample.

TABLE 3 | Results of ANCOVA for internalized symptoms, hyperactivity, social skills and adaptability.

Variable	df	F	p	Partial η^2
Internalized symptoms	1	2.08	0.15	0.05
Hyperactivity	1	2.81	0.10	0.07
Social skills	1	1.07	0.32	0.02
Adaptability	1	0.49	0.48	0.01

Main Analyses

Negative Indicators of Mental Health

ANCOVAs did not reveal a significant effect of condition on internalized symptoms [$F_{(1, 33)} = 2.08$, $p = 0.15$, partial $\eta^2 = 0.05$], controlling for baseline levels. Sensitivity analyses were conducted using paired *t*-tests. These analyses indicated that for the whole sample, internalized symptom scores were statistically significantly lower at post-intervention [$t_{(35)} = 2.45$, $p = 0.01$; 95% CI = 0.23, 2.53], when compared to pre-intervention scores. Hence, participants from both groups displayed lower internalized symptom scores from pre-to-post intervention, descriptive statistics showing a similar decrease in scores for participants from both groups.

We found no impact of group on levels of hyperactivity [$F_{(1, 37)} = 2.81$, $p = 0.10$, partial $\eta^2 = 0.07$; please refer to **Table 3**). Sensitivity analyses were conducted using paired *t*-tests. These analyses indicated that for both the experimental and control groups, hyperactivity scores were statistically significantly lower at post-intervention [$t_{(37)} = 3.24$, $p = 0.002$; 95% CI = 0.30, 1.32], when compared to pre-intervention scores. Hence, participants from both groups displayed lower hyperactivity scores from pre-to-post intervention, descriptive statistics showing a similar decrease in scores for participants from both groups.

Positive Indicators of Mental Health

We found no impact of group on levels of social skills [$F_{(1, 37)} = 1.01$, $p = 0.32$, partial $\eta^2 = 0.02$] and adaptability [$F_{(1, 37)} = 0.49$, $p = 0.48$, partial $\eta^2 = 0.01$; please refer to **Table 3**].

Change Over Time

Negative Indicators of Mental Health

To document change over time, we conducted paired *t*-tests to examine within-group changes in pre-to-post intervention scores. For internalized symptoms, paired *t*-tests revealed a statistically significant change within participants in the experimental group [$t_{(18)} = 2.19$, $p = 0.04$] and the wait-list control group [$t_{(16)} = 2.29$, $p = 0.03$]. Participants from both groups showed a decrease in internalized symptoms from pre-intervention ($M_{pre\ experimental} = 9.61$; $M_{pre\ control} = 9.29$) to post-intervention ($M_{post\ experimental} = 8.55$; $M_{post\ control} = 7.88$). Similarly, for hyperactivity, paired t-tests revealed statistically significant change within participants in the in the experimental group [$t_{(18)} = 2.45$, $p = 0.02$] and the wait-list control group [$t_{(18)} = 2.44$, $p = 0.02$]. Participants from both groups showed a decrease in internalized symptoms from pre-intervention ($M_{pre\ experimental} = 4.08$; $M_{pre\ control} = 3.37$) to post-intervention ($M_{post\ experimental} = 3.55$; $M_{post\ control} = 2.26$).

Positive Indicators of Mental Health

Paired t-tests did not reveal statistically significant pre-to-post changes for adaptability (experimental: $t_{(18)} = -0.36$, $p = 0.71$; control: $t_{(18)} = -1.88$, $p = 0.70$) for participants in the experimental and wait-list control groups. For social skills, paired t-tests did not reveal statistically significant pre-to-post changes in participants from the experimental group [$t_{(18)} = 1.52$, $p = 0.14$). In contrast, paired t-tests were significant for participants from the control group [$t_{(18)} = -2.4$, $p = 0.02$]. Hence, participants in wait-list control group showed an increase in social skills scores from pre-intervention ($M_{pre} = 2.11$) to post-intervention ($M_{post} = 4.16$), whereas scores remained similar among participants in the experimental group ($M_{pre} = 3.16$; $M_{post} = 3.66$).

DISCUSSION

The goal of the present study was to evaluate the impact and feasibility of a combined MBI and P4C intervention on negative and positive indicators of mental health. Overall, results from this pilot feasibility study show that the combined MBI and P4C intervention was not more useful in decreasing internalized symptoms, hyperactivity symptoms, social skills and adaptability as the passage of time, as can be seen by similar improvements in scores of the control group participants and small effect sizes. These preliminary results suggest that mindfulness and P4C may not be more effective as an intervention to foster short-term resiliency, well-being and mental health in preschool children as the passage of time.

As detailed in the introduction, the initial goal of this pilot feasibility study was to explore the impact of a combined mindfulness and P4C intervention on positive and negative indicators of mental health in pre-kindergarten children, with the overarching goal of developing an adaptation of the existential psychology framework for youth. The present findings are surprising given that previous research had shown a positive impact of both interventions with elementary school children (5, 57, 58, 60).

Given the paucity of evidence-based research pertaining to the usefulness of P4C on mental health, results from this pilot are difficult to explain in relation to the previous literature. Indeed, upon researching the literature on this topic, one quasi-experimental study documenting the impact of P4C on mental health in children (aged between 9 and 12 years-old) with severe behavioral disorders was found and benefits with regards to self-regulation were reported (60). Work by the same group of authors also show improvements in self-regulation and rule adherence following P4C in teenagers with severe psychiatric disorders in juvenile prison settings (61). Authors argue that, although encouraging dialogue and argumentation in juvenile prisons may be perceived as counterintuitive, the use of P4C for these teenagers may be helpful in fostering a sense of control and self-determination in an environment in which typically there is none. As such, P4C may show promise in fostering greater autonomy and self-determination in this specific population. Self-determination theory would also allow for the empirical study of themes pertaining to existential psychology with youth.

Caution is warranted in drawing conclusions between the studies mentioned above, implemented with older elementary school students, and the present one, implemented with pre-kindergartners, because of the important difference in ages and levels of cognitive, social and emotional development. Nonetheless, it is worth mentioning that P4C activities chosen for this study were initially developed for pre-kindergartners and adapted accordingly to their cognitive, social and emotional levels of reasoning. Furthermore, past research in philosophy has also been published on this practice for pre-kindergartners and kindergartners alike (51, 66), suggesting P4C is adequate and may be beneficial with younger children as well.

An appreciable amount of research describing the positive impact of MBIs on positive and negative indicators of mental health in clinical and non-clinical samples of elementary school children has been published previously (5, 12, 67). Although research with pre-kindergarteners remains scarce, previous work by Flook et al. (68) has described benefits of mindfulness training with regards to social skills, social-emotional development and overall academic achievement for this population. On the other hand, recent work by Thierry et al. (69), failed to find statistically significant impacts of mindfulness training on prosocial behaviors in prekindergartners, although authors did report benefits with regards to executive functioning (working memory, inhibition and flexibility). Overall, these results seem to indicate that MBIs are a suitable intervention for preschoolers, although the present study does not show the same results.

We did not find similar results in the present study and the reason for this remains unclear, although certain explanations warrant attention. Some studies on mindfulness have documented ceiling effects in non-clinical populations in school settings (70), which may partly explain the present findings. Speaking to the low-level baseline of psychopathology in this sample, the available margins for change and for overall improvement from pre-to-post intervention were lower, which could explain the non- statistically significant results and small effect sizes that were obtained. Indeed, a recent meta-analysis on the impact of mindfulness-based interventions in youth showed much stronger effect sizes among clinical populations (0.50 vs. 0.20 for non-clinical populations), as there is more "room to improve" among these children (67). Nonetheless, it is important to note that participants from both the control and experimental groups showed improvements from pre-to-post intervention.

This being said, a recent report on Canadians' mental health has recommended that more research be devoted to developing evidence-based interventions that can be universally implemented in classroom as prevention strategies to foster better mental health and to prevent the development of psychological disorders in elementary school students (71). Cost-benefit analyses of the best mental health prevention programs for youth in the United States have also shown that these programs yield a minimum of 8$ for each dollar invested in mental health prevention. Thus, investing in developing prevention programs is a viable and effective solution to decrease future potential mental health issues and psychological disorders in school-based settings (72).

Furthermore, some methodological issues may have influenced the results. Noticing internalized symptoms (and subsequent changes in these symptoms) in students is known to be a difficult task for teachers, given that they are not as disruptive as externalized symptoms and do not tend to impact the overall classroom climate as much (17). Teachers were also not blind to both conditions in this study, which could have biased their way of reporting pre and post intervention scores for their students. In order to correct for these important issues, adding parent-reported data in future studies of this sort could help in providing a clearer picture of the overall evolution of internalized symptoms in preschool children participants. It could also be interesting for future research to include behavioral measures of constructs such as altruism (e.g., to give children a number of stickers and ask them how many they would keep and share with others), in order to reduce shared method variance.

Another reason that could explain our results is the duration of our intervention. Implementing a longer intervention, such as the 10-weeks intervention detailed in the abovementioned studies, could also perhaps yield different results, as the "dosage" would be stronger. Lasting only 5 weeks, this combined intervention was considered as brief and therefore may not have been "strong" enough to provoke changes among the children. Finally, it is possible that philosophical activities are a source of stress in very young children, which may not have developed the cognitive ability to reflect on existential questions. It is also possible that these manifestations of stress are short-lived and are worth it for future gain. Anecdotal evidence provided by participants and their teachers alike tend to support the latter. Indeed, teachers did not report an increase in distress in their students, and students reported liking to participate in the P4C activities. They were eager for the next visit from session leaders and asked frequently their teachers when they would be back in class.

From a feasibility standpoint, results from this study and anecdotal evidence collected from teachers taking part in this project and from session leaders show the intervention was positively received and enjoyed by students and teachers alike. Themes were understood by the participants, who actively participated in the MBI and P4C activities. Visual and video content was preferred to stories and comic strips, as children could easily and rapidly understand the existential themes that were presented to them. When stories were used, session leaders reported they often had to reword the content of the story and remind students of the theme during the discussion. In such instances, participants did not share amongst themselves as much, which made for less lively conversations. Perhaps the use of philosophically-oriented stories may be more adequate for older elementary school children. Overall, however, the project was well received by children and the school and thus, shows good acceptability and feasibility. This underlines the need for more research, as anecdotal evidence and empirical evidence brought forward by the data, such as the small effect sizes, are contradictory in this study.

Strengths and Limitations

This study counts multiple strengths. First, the experimental design implemented in this project represents a notable strength. Indeed, both classrooms were similar at pre-intervention, with the exception of the level of social skills and adaptability, and were randomly assigned to each condition. With regards to social skills and adaptability, given pre-intervention scores from students in the experimental group were statistically and significantly higher than those of students in the control group, it is possible the control group simply "caught up" with the other group over time. However, with regards to social skills and adaptability, although children in the experimental group were substantially higher than those in the control group prior to the intervention, at post intervention, when looking at the confidence intervals, both groups overlapped, and thus were not really different from each other. Second, the absence of attrition in this study further represents a major strength, as it ensures pre-to-post differences in scores and changes in time are not due to changes in the sample. Finally, this study provides preliminary data on the impact of P4C sessions (combined with the MBI) from the rigorous standpoint of an experimental, randomized cluster trial. Given almost-to-none evidence-based data is available on the impact of P4C in preschool and elementary school children in psychology, this study contributes to strengthening the knowledge on this increasingly implemented practice in classrooms across North America. Finally, this study was innovative in its aim to document the adaptation of the existential psychology framework for children, although it could not provide preliminary evidence showing its promise in fostering greater mental health and well-being. Future work will be needed to establish if such benefits are observed in school settings.

Despite these strengths, the small sample size represents a noticeable limitation of this project. As it remains possible that both classrooms may have been different, a larger randomized cluster trial would have provided more robust and generalizable results, and should thus be planned as future steps in this line of research. This would also provide higher statistical power. Implementing a longitudinal trial would also strengthen future studies of this sort, as results from the present study only speaks to immediate, post-intervention effects of the intervention on children's mental health. Past mindfulness research in youth has shown patterns where benefits become apparent only after a 3 or 6-months follow-up (35). Similar patterns could emerge in our participants, although this remains a hypothesis at the moment. Administering a more important number of items in future studies may also be helpful in providing a clearer, more complete portrait of the situation. Indeed, as only a few selected items of each subscale were administered in the current project, this reduced the sensitivity of our measures to changes in our participants. Perhaps new statistically significant pre-to-post changes would be detected with a larger number of administered items. We note, however, that with this sample size and this number of items, we were still able to detect changes in internalized symptoms, hyperactivity and social skills. Furthermore, all questionnaires in one group were filled out by one teacher, which may have further biased our results. Finally, given improvements that were observed in participants from both conditions, it remains a strong possibility that the documented effects were due to the simple passage of time.

Suggestions for Further Research

Adding a longitudinal component to future studies of the sort would allow for a more detailed portrait of the impact of such a combined intervention on mental health in youth. Furthermore, using age as a moderating variable in larger sample size studies would also allow us to better grasp if P4C is most useful with younger or older elementary school children. Based on previous preliminary research in P4C with youth from clinical populations and our own justification of the relevance of combining a MBI and P4C, measuring self-determination in future studies is also recommended, as, theoretically, a combined mindfulness and P4C intervention should help increase self-determination in children. Furthermore, as P4C activities are often led by teachers or completed in the presence of teachers, future studies could incorporate observational methods to control for teacher autonomy supportive behaviors (73), as these behaviors have been linked to higher reported levels of basic psychological needs satisfaction in children and overall greater self-determination (74, 75).

Similarly, future studies of this sort could incorporate observational methods to evaluate changes in altruism, as similar methods have been successfully previously employed with preschoolers in the mindfulness literature (68). Implementing a multi-method approach by combining observational and teacher-reported measures would increase the scope of variables being evaluated and strengthen the study design. Including parents and caregivers reported data would also be useful in gaining better insight of the impact this combined intervention has in pre-kindergarten children.

Finally, a further step would also be to compare the effectiveness of the combined intervention to another intervention aimed at fostering greater mental health and self-determination in children, within an experimental design with an active control group. Meanwhile, publishing any evidence-based data on the impact of P4C would also help in providing a better sense of its usefulness (or lack thereof) on psychological health and well-being in preschoolers youth.

CONCLUSION

Overall, results from this pilot feasibility study showed that the MBI and P4C intervention was not more useful in decreasing anxiety and hyperactivity symptoms as the passage of time, as can be seen by similar or greater improvements in scores of the control group participants. These preliminary results suggest that mindfulness and philosophy for children may not be the most effective intervention to foster short-term resiliency, well-being and better mental health in children. Further research using longer time frames and comparing the effectiveness of this combined intervention to other types of school-based interventions with similar aims (such as, for example, P4C or MBI alone) is warranted, to evaluate if mindfulness and P4C interventions have an added value compared to other types of interventions implemented in school settings.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Bishop's University Research Ethics Board. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin and by the teachers taking part in this study.

AUTHOR CONTRIBUTIONS

CM-H and DL conceptualized and coordinated the study, adapted the intervention and trained the session leaders involved in this study, performed data analysis, and drafted the manuscript. GM and GT contributed extensively to data interpretation and revision of the manuscript. CD helped in data collection and coordination of the study, while contributing to revision of the manuscript. M-AE and MG contributed to the design of the study and revision of the manuscript. All authors contributed to the article and approved the submitted version.

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Conflict of Interest: CM-H has released a manual on the mindfulness-based intervention described and used in this study (Midi Trente Publishers).

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