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ORIGINAL PAPER

Communication Deficits and the Motor System: Exploring Patterns of Associations in Autism Spectrum Disorder (ASD)

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Abstract Many children with autism spectrum disorder (ASD) have notable difficulties in motor, speech and language domains. The connection between motor skills (oral-motor, manual-motor) and speech and language deficits reported in other developmental disorders raises important questions about a potential relationship between motor skills and speech-language deficits in ASD. To this end, we examined data from children with ASD ($n = 1781$), 2–17 years of age, enrolled in the Autism Speaks—Autism Treatment Network (AS-ATN) registry who completed a multidisciplinary evaluation that included diagnostic, physical, cognitive and behavioral assessments as part of a routine standard of care protocol. After adjusting for age, non-verbal IQ, Attention Deficit Hyperactivity Disorder (ADHD) medication use, and muscle tone, separate multiple linear regression analyses revealed significant positive associations of fine motor skills (FM) with both expressive language (EL) and receptive language (RL) skills in an impaired FM subgroup; in contrast, the impaired gross motor (GM) subgroup showed no association with EL but a significant negative association with RL. Similar analyses between motor skills and interpersonal relationships across the sample found both GM skills and FM skills to be associated with social interactions. These results suggest potential differences in the contributions of fine versus gross motor skills to autistic profiles and may provide another lens with which to view communication differences across the autism spectrum for use in treatment interventions.

Keywords Autism · Motor deficits · Language · Social interactions

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Introduction

The connection between motor skills and communication deficits in neurodevelopmental disorders has been a growing area of interest over the past decade. Studies have shown that deficits in language are correlated with deficits in oral-motor and manual-motor skills in autism spectrum disorder (ASD) (Gernsbacher et al. 2008) and that the relationship between them may be genetic (Bishop 2001) or neurological (Hill 2001) in origin. However, much of this research has excluded individuals with ASD to preclude the effects of co-morbidities like rigid and repetitive behaviors and poor social skills. Additionally, the bulk of the speech and language research in ASD has been with infants and younger children and those with higher IQs. The question that continues to challenge the field is whether any specific factor drives the onset of the communication deficits and maintains them across development in ASD.

A growing body of evidence suggests that oral-motor skills, manual motor skill development and basic perceptuo-motor impairments may impact both social and speech and language skills in individuals with ASD (Bhat et al. 2011; Gernsbacher et al. 2008; MacDonald et al. 2013). In fact, motor delays at 18 months have been found to be highly predictive of the severity of ASD at 3 years of age in at-risk toddlers with a family history of ASD (Brian et al. 2008). Though the vast majority of these infants at risk show no signs at 6 months, there is a set of social communicative, motor and attention behaviors at 12 months that together can predict later diagnosis (Tager-Flusberg 2010). Interestingly, the correlation of motor deficits with expressive language and receptive language skills helps to differentiate children with ASD from their typically developing peers and separate these children into language ability subgroups (highly, moderately, and minimally fluent) (Gernsbacher et al. 2008). Despite this, the only motor abnormalities currently included in the diagnostic criteria for ASD are stereotypical, repetitive motor movements (The Diagnostic and Statistical Manual of Mental Disorders—5th ed.; DSM-5; American Psychiatric Association 2013). These repetitive behaviors include motor stereotypies such as body rocking, hand flapping, and finger/hand mannerisms, though motor issues are not limited to these actions (McCleery et al. 2013). Difficulties with postural control, fine and gross motor coordination and gait abnormalities also frequently co-occur with ASD (Leary and Hill 1996; Provost et al. 2007; Bhat et al. 2011). In a comprehensive review of the autism high-risk infant literature, Rogers (2009) concluded that motor issues are a consistent finding in this population. Importantly, she observed that some of the subtle repetitive movements appear to emerge early in development even before social and communication impairments. A growing body of evidence thus seems to implicate the motor system in ASD (Fournier et al. 2010).

Among the earliest developing motor-related behaviors linked to communication development are vocal imitation and the imitation of facial expressions, which emerge in the first 6 months of life (Iverson and Fagan 2004). This is when infants begin to engage in coordinated routines with a parent/caregiver that serve as the building blocks of social reciprocity and precursors of interpersonal interaction (Colonnesi et al. 2012). Yirmiya et al. (2006) found evidence of reduced communication synchrony during mother-infant interactions in high-risk infant siblings of children diagnosed with autism compared with low-risk infants without a family history of autism. In a separate study, Iverson and Wozniak (2007) found that whereas the rate of rhythmic arm movements increased from pre-babble to babble-onset stages in both high-risk and low-risk infants, the increase was lower in the high-risk group; delays in reduplicative babbling and first word onset have also been evident in high-risk infants. Taken together, these findings provide further support for a potential link between speech-language, social and motor skills in ASD.

Evidence for a role for the motor system in speech perception comes from a variety of studies (Galantucci et al. 2006; Liberman and Mattingly 1985). In one such study, Fadiga et al. (2002) demonstrated using Transcranial Magnetic Stimulation (TMS), that during speech listening there is an increase in motor evoked potentials recorded from the listeners' tongue muscles when the pronunciation of the presented words strongly evoked tongue movements. Phonetic perception appears then to activate left hemisphere areas involved in speech production, namely, Broca's area, cerebellum, premotor cortex, and anterior insula, in addition to auditory areas such as superior temporal gyrus (STG) (Benson et al. 2001; Price et al. 1996; Zatorre and Binder 2000). This is consistent with the idea that producing speech entails a generation of internal motor models of speech which are compared with incoming data. More recently, Kuhl et al. (2014) found that infants as young as 11–12 months of age show evidence of sensitivity to motor speech gestures and draw on these during phonetic perception. Studies also indicate that non-linguistic oral-motor skills appear to contribute to children's nonword repetition ability, a clinical measure of phonological processing. These findings suggest that aspects of language learning and consequent language deficits may be rooted in the ability to perform complex sensorimotor transformations (Krishnan et al. 2013). Additionally, children identified on the basis of language impairment also show significant motor co-morbidity (Robinson et al. 1991; Webster et al. 2006). Not surprisingly, then, in a subgroup of children with ASD, communicative deficits appear to stem from basic motor and aural-motor issues (Leary and Hill 1996). In children with normal non-verbal IQ, vocal and imitation skills in early years predict language skills at 5 years of age better than early joint attention. To the extent that motor difficulties in autistic individuals can range from more basic skills such as pointing to more refined skills such as articulation and imitation, Belmonte et al. (2013) recommend that motor skills be assessed across this entire range in individuals with ASD. Based on their findings, the present study further distinguishes fine from gross motor deficits in ASD, in keeping with the precise articulatory gestures involved in speech production.

In summary, early motor abilities appear to set the stage for the development of social communication skills, and the degree of intellectual disability in combination with social, motor, and language impairments may prove to be an aggregate marker that best predicts ASD and intervention outcomes (Mody and Belliveau 2012). One should note that although IQ is implicated in ASD and IQ is related to language, disparities in IQ do not fully account for the heterogeneity in the language ability of children with ASD. Kjelgaard and Tager-Flusberg (2001) found that there were children with ASD who had low IQ but language skills within the normal range, as well as ASD children with high IQ and impaired language ability. Thus, language and cognitive ability may be dissociable in children with ASD. However, controlling for IQ remains essential because low cognitive capacity might skew the results by contributing to poorer performance among the low functioning participants. The heterogeneity among children and adults with ASD appears to suggest alterations in developmental trajectories across multiple domains, with the disorder emerging from an aggregate of risk factors rather than any single marker.

Insofar as a developing infant discovers the world by exploring their environment through physical and vocal play, early motor deficits observed in high-risk infants may have cascading effects on the child's development of social and communication behaviors (Bhat et al. 2012). It remains unclear whether motor deficits account for the communication deficits that are characteristic of individuals with ASD. The ATN database provides an opportunity for exploring this question. Through secondary analyses of select measures of language, social interaction and motor skills, we aim to identify relationships that could further inform our understanding of communication deficits in ASD. We hypothesize that fine motor skills but not gross motor skills will be associated with language given the fine-grained articulatory-acoustic mapping required to master speech; social behaviors, however, draw on motor interactions with peers during activities, at home, on the playground or in the classroom and as such may be associated with fine and gross motor skills. The pattern of associations can have important implications for developing motor-based interventions targeting language and social communication in this population.

Methods

This study was conducted as part of the research activities of the Autism Treatment Network (ATN), a registry collecting data on children with ASD from across 17 academic health centers in the US and Canada. All participants completed a multi-disciplinary evaluation that included diagnostic, physical, cognitive and behavioral assessments. Parents or legal guardians of all participants provided written consent for participation and the institutional review board at each participating site reviewed and approved this study.

Participants

This study examined data from 6708 children enrolled in the ATN at the time of the database closure for the analyses undertaken (December 19, 2013) who had age at enrollment available. We aimed to leverage the strength of the entire database, males and females, by incorporating all ASD diagnoses [autistic disorder, Asperger's disorder, pervasive developmental disorder—not otherwise specified (PDD-NOS)], confirmed by DSM-IV-TR (fourth edition, text revision; APA 2000) and administration of the Autism Diagnostic Observation Schedule (ADOS; Lord et al. 2000), the full age range available (2–17 years), and all IQ levels. The data were collected as routine standard of care of ASD, though not all collaborating centers used the same instruments. Additionally, we controlled for select variables including, age, non-verbal IQ, and use of medication for Attention Deficit Hyperactivity Disorder (ADHD), insofar as these variables and a history of ADHD all affect language and may confound our findings. We opted to use ADHD medication status than the Childhood Behavior Checklist (CBCL) for controlling for ADHD symptoms as we did not have CBCL data on all the subjects. The ADHD medication usage served as a proxy for clinically significant attention issues. We also adjusted for muscle tone, based on neurological examination, as children with low tone are more likely to show motor impairments. Thus, we tried to exclude these confounding effects likely to influence the receptive and expressive language skills, social interaction skills, and fine and gross motor skills, key variables assessed using the Mullen Scales of Early Learning (Mullen 1995) and Vineland Adaptive Behavior Scales-II (Sparrow et al. 2005). After adjusting for age, non-verbal IQ, ADHD medication use, and muscle tone, while maximizing the number of eligible participants for inclusion (approx. 1781 children), the actual sample ranged in age between 2 and 15.5 years. Table 1 below lists the variables and standardized measures used in the analyses.

There were other measures of language [e.g., Preschool Language Scale (PLS-IV and V), Clinical Evaluation of Language Fundamentals (CELF-IV, CELF-P2), Oral and Written Language Scales (OWLS)] and IQ (e.g. Stanford Binet, 5th Edition) in the database; however, the samples were too small to justify using them in this large scale analysis. It is important to note that we used raw scores in the analyses as the truncated range of scores of the population makes it difficult to derive meaningful standard score interpretations. Additionally, the use of the Mullen Scales with participants who fell outside the age range, is fairly well justified. It is a developmental measure that would be appropriate for individuals with impaired functioning who cannot complete an age-level measure. Finally, the GM Scale of the Mullen was not a required part of the original

Table 1 Study variables and corresponding assessment measures

Measure/assessment	Subtest/test section	Abbreviation	Score type
Motor			
VABS-II[a]	Gross motor subdomain	VABS-II GM	Raw scores, (v-scale score ≤10 used only for defining impairment)
Mullen[b]	Fine motor scale	Mullen FM	Raw scores, T-scores (T-score ≤40 used only for defining impairment)
Language			
Mullen[b]	Expressive language scale	Mullen EL	Raw scores
Mullen[b]	Receptive language scale	Mullen RL	Raw scores
Social interactions			
VABS-II[a]	Interpersonal relations subdomain	VABS-II IR	Raw scores
ADOS[c]	Quality of social overtures	ADOS QSOV[d]	0–3 rating 0 = no problem 2 = poor
Nonverbal IQ			
Mullen[b]	Visual reception scale	Mullen VR	Raw scores

[a]Vineland Adaptive Behavior Scale-Second Edition (VABS-II): Parent Caregiver Rating Form (Sparrow et al. 2005)

[b]Mullen Scales of Early Learning (Mullen 1995)

[c]Autism Diagnostic Observation Schedule (ADOS; Lord et al. 2000)

[d]Quality of Social Overture Variable (ADOS; Lord et al. 2000)

ATN protocol; as such there was not enough data for analysis from this measure, so the Vineland was used as a proxy.

The distributions of the primary variables in the sample were roughly normal, the mean and median not extremely far off from each other and the Q–Q plots (quantile–quantile) falling along a line at a 45° angle. Fine and gross motor scores though correlated ($r = 0.49$, $p < 0.0001$) were examined separately, based on a hypothesized difference in their contribution to autistic profiles. To this end, we divided participants into motor skill subgroups based on VABS-II v-scale score and Mullen T-score cutoffs corresponding to poor or impaired versus unimpaired GM and FM skills (see Table 1) to further explore the impact of an impaired motor system on speech and language in ASD. As noted earlier, raw score were used in the analysis. Data were analyzed using multiple linear regression models to examine the association between motor skills and language, as well as between motor skills and social interactions, controlling for age, nonverbal IQ, ADHD medication use and muscle tone.

Results

The results are organized under two main headings: (a) the relationship between motor skills (FM and GM) and language abilities (RL and EL) in ASD, and (b) the relationship between motor skills and social interactions in ASD.

Relationship Between Motor Skills and Language Abilities in ASD

Across Entire Sample

Multiple linear regression models built separately for expressive language (EL; Mullen raw score) and receptive language (RL; Mullen raw score) as dependent variables and gross motor (GM; VABS-II raw score) and fine motor (FM; Mullen raw score) as independent variables yielded significant associations between language and motor abilities. After adjusting for age, non-verbal IQ, ADHD medication use, and muscle tone, both EL (mean = 17.3, SD = 9.0; $\beta = 0.4$, std error = 0.04, $p < 0.0001$; $n = 1562$) and RL (mean = 18.3, SD = 9.5; $\beta = 0.3$, std error = 0.04, $p < 0.0001$; $n = 1562$) were found to be positively associated with FM (mean = 24.7, SD = 6.7). In contrast, RL showed no association with GM (mean = 18.2, SD = 9.4; $\beta = 0.02$, std error = 0.02, $p = 0.3034$; $n = 1443$), whereas EL was positively related (mean = 17.3, SD = 9.0; $\beta = 0.1$, std error = 0.02, $p < 0.0001$; $n = 1449$) with GM (mean = 56.2, SD = 10.3).

Within Impaired Motor Skills Subgroups

The results were further examined as a function of impaired motor skills. Table 2 presents standardized assessment raw scores of the four subgroups with impaired and unimpaired fine motor skills and gross motor skills within the ATN sample.

Specifically, using T-scores and v-scale scores for determining impaired versus unimpaired motor skill grouping cut-offs but raw scores in the analysis, and adjusting for age, non-verbal IQ, ADHD medication use, and muscle tone, the impaired FM group (based on Mullen T-score ≤40) showed significant positive association with both EL (β = 0.39, std error = 0.05, $p < 0.0001$; $n = 1297$) and RL (β = 0.25, std error = 0.04, $p < 0.0001$; $n = 1292$); the impaired GM group (based on VABS-II v-scale score ≤10), however, showed no association with EL (β = 0.02, std error = 0.05, p = 0.67; $n = 334$) but a significant negative association with RL (β = −0.09, std error = 0.04, p = 0.04; $n = 332$). The effect sizes of these findings were bigger for the impaired FM (0.028, 0.046) than impaired GM (−0.010, 0.002) associations with RL and EL, respectively. In the unimpaired FM group (Mullen t-score ≥50) FM was not significantly associated with EL (β = 0.60, std error = 0.39, p = 0.14; $n = 47$) or RL (β = 0.18, std error = 0.40, p = 0.65; $n = 47$); in the unimpaired GM group (Vineland v-scale score ≥13) GM skill was positively associated with EL (β = 0.12, std error = 0.04, p = 0.006; $n = 568$) but was unrelated to RL (β = 0.07, std error = 0.04, p = 0.10; $n = 567$). See Table 3 for complete details including the effect sizes of the associations between the variables.

Table 2 Descriptive statistics of the adjusted regression analysis samples for impaired and unimpaired GM and FM subgroups: assessment raw scores

Label (raw scores)	Variable	Impaired		Unimpaired	
		Mean	SD	Mean	SD
Fine motor subgroups					
Mullen expressive language	EL	16.6	8.5	29.9	9.5
Mullen receptive language	RL	17.3	9.0	31.6	9.1
Mullen fine motor	FM	23.7	5.8	36.9	7.7
Mullen visual reception	IQ	25.4	8.0	38.7	8.6
Gross motor subgroups					
Mullen expressive language	EL	15.6	8.9	18.1	9.1
Mullen receptive language	RL	17.0	9.3	19.0	9.5
Vineland gross motor	GM	49.7	10.0	61.0	8.8
Mullen visual reception	IQ	25.0	8.5	26.8	8.4

Relationship Between Motor Skills and Social Interactions in ASD

Multiple linear regression models with social interactions (VABS-II Interpersonal Relationships) as the dependent variable, and age, non-verbal IQ, ADHD medication use, and muscle tone as independent variables built separately for GM (VABS-II raw score) and FM (Mullen raw score) found both GM and FM skills to be significantly associated with social interaction skills. That is, for each unit increase in gross or fine motor raw score, Vineland Interpersonal Relationships raw score increased by about 0.3 (β = 0.3, std error = 0.02, $p < 0.0001$; $n = 1602$) for GM (mean = 56.34, SD = 10.28), and also 0.3 (β = 0.3, std error = 0.05, $p < 0.0001$; $n = 1621$) for FM (mean = 24.77, SD = 6.73). In contrast, a similar analysis using adjusted multinomial logistic regression with motor skills and Quality of Social Overtures Variable (QSOV), a measure of social skills from the ADOS (see Table 1), revealed that after adjusting for age, non-verbal IQ, and muscle tone, there was a significant negative association between QSOV scores and FM raw scores but not GM raw scores. Specifically, for each unit increase in FM raw score, the odds of having a QSOV score of 2 (i.e., poor quality overtures lacking integration into context and/or social quality) versus 0 (no problem) decreased by 10 % (OR 0.90, 95 % CI 0.82–0.99, p = 0.0226; $n = 1494$); there was no significant association of QSOV scores with GM (OR 0.97, 95 % CI 0.93–1.0, p = 0.05872; $n = 1383$).

Table 3 Associations between Motor (GM, FM) and Language (RL, EL) raw scores in Impaired and Unimpaired Gross and Fine Motor subgroups

Analysis group	Estimate	SE	T value	p-value	Effect size
Impaired GM & EL ($n = 334$)	0.02	0.05	0.43	0.67	0.002
Impaired GM & RL ($n = 332$)	−0.09	0.04	−2.07	0.04*	−0.010
Impaired FM & EL ($n = 1297$)	0.39	0.05	8.25	<0.0001*	0.046
Impaired FM & RL ($n = 1292$)	0.25	0.04	5.66	<0.0001*	0.028
Unimpaired GM & EL ($n = 568$)	0.12	0.04	2.77	0.006*	0.013
Unimpaired GM & RL ($n = 567$)	0.07	0.04	1.64	0.10	0.007
Unimpaired FM & EL ($n = 47$)	0.60	0.39	1.51	0.14	0.063
Unimpaired FM & RL ($n = 47$)	0.18	0.40	0.45	0.65	0.020

*$p < 0.05$

Discussion

Results from the current study are consistent with findings in the literature and support the hypothesis that motor problems frequently seen in children with ASD appear to be associated with their language and social behaviors (Bhat et al. 2011; Gernsbacher et al. 2008). After adjusting for age, non-verbal IQ, ADHD medication use and muscle tone, multiple linear regression analyses with the ATN sample revealed that as the Mullen fine motor score increased, Mullen receptive and expressive language scores improved. Interestingly though, Vineland gross motor skills revealed a positive association with Mullen expressive language but no association with Mullen receptive language, raising the possibility of a potential difference in the contributions of gross versus fine motor deficits to language outcomes in ASD. Research in the field of phonological development may in fact support this view insofar as speech is a sequence of finely-tuned articulatory gestures and both speech perception and production entail sensitivity to this articulatory-acoustic mapping (Studdert-Kennedy 2000). Results from the additional analyses of language abilities in GM-impaired versus FM-impaired subgroups appear to be consistent with this notion.

Our findings revealed a strong positive association between fine motor skills and EL and RL in the impaired FM group. However, the lack of association of GM skills with EL, combined with its significant negative relationship with RL in the impaired GM subgroup may reflect a trade-off between GM and language skills in individuals with ASD. Based on a limited capacity model, we speculate that the demands of an impaired gross motor system may leave the child on the autism spectrum with fewer resources for developing joint attention abilities. This could account for deficits in receptive language and consequent failure to attend to speech (Ceponiene et al. 2003). If borne out by future studies, these findings may be important for the development of phenotypes for use in treatment intervention (Belmonte et al. 2013). However, the GM–RL result while significant was only a small effect. In fact, based on the relative effect sizes of the associations between motor and language skills, the relationship between FM and EL/RL may be a more robust one than that between GM and EL or RL.

Our data also revealed a significant association of both gross and fine motor skills with social interaction skills (VABS-II IR) on a broad level, across the entire sample. However, further probing using the QSOV measure from the ADOS once again revealed different patterns of association with gross and fine motor skills. Whereas an increase in fine motor scores was associated with reduced probability of scoring poorly in the social domain (QSOV), gross motor and QSOV scores were unrelated. We did not correct for multiple comparisons given the exploratory nature of the analyses aimed at developing new hypotheses. Additionally, some researchers do not believe it is necessary to correct for multiple comparisons (Rothman 1990).

In summary, the analysis of the ATN registry data yielded some interesting patterns of association between motor skills and language and social behaviors. The results highlight the importance of assessing and monitoring gross and fine motor skills in individuals with ASD. This distinction may help provide another lens through which to view the differences in core social interaction and language deficits across the autism spectrum for potential use in phenotype research in ASD. Additionally, it may help distinguish between profiles under the control of known genes (e.g., FOX P2; CNTNAP2) and explain the patterns of overlap with other developmental disorders.

As with any secondary analyses of this nature, there are limitations to the current study. One shortcoming was the limited number of language measures to select from, with more comprehensive language measures like the CELF, PLS, and OWLS being available only for a small number of the subjects. Furthermore, a measure of oral-motor abilities (e.g., test of apraxia of speech, nonword repetition) would have allowed for a more direct examination of the association between fine motor and language abilities in the present study. This is especially important given the presence of motor apraxia in this population. In the face of growing evidence of apraxia of speech in autistic individuals (Tierney et al. 2015), findings from the present study serve to further emphasize the role of the motor system in ASD. Additionally gross motor skills were based on the Vineland (VABS-II), a parent-report measure, whereas the Mullen provides a direct assessment of fine motor skills. The lack of an examiner-administered assessment of gross motor skills is a limitation of the database and sample we are working with, in that the GM Scale of the Mullen was not a required component of the original ATN protocol. Hence, the different pattern of association with gross versus fine motor skills should be viewed with some caution pending confirmation in future studies using a direct measure of GM skills. It is worth mentioning, though, that studies have found Mullen scores on the gross and fine motor subtests to be significantly related to corresponding scores on the VABS-II (Lloyd et al. 2013).

That motor skills may mediate oral language development, opportunities for social interactions and joint attention warrants further research on potential links between manual-motor (e.g., sign language) and oro-motor (e.g. speech) gestures in minimally verbal individuals on the autism spectrum (Shield 2014). Finally, a prospective data model would have been better suited to answer the questions under study. Longitudinal data would have allowed for meaningful interrogation of interaction effects between age, non-verbal IQ, motor and language variables for characterizing ASD developmental trajectories and profiles; in its absence, we chose to adjust for age and non-verbal IQ to examine the relationship between motor, language, and social variables. The study, however, was undertaken with these limitations in mind and as such represents a pilot effort to explore the impact of motor skills on language and social interactions in ASD. Despite the shortcomings, the present study yielded some interesting results that may contribute to the generation of new hypotheses and the development of more comprehensive models of ASD for use in treatment studies. Intervention by AAC (Augmentative-Alternative Communication) and assistive technology (AT) specialists and occupational therapists supporting the development of writing and typing skills may serve to stimulate the development of print as an alternative form of communication especially in minimally verbal individuals on the spectrum with normal cognitive function. An important first step, however, would be the development of a comprehensive cognitive, sensorimotor and communication assessment battery specifically designed for use with this population to help determine their candidacy for various types of intervention.

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Authors' Contributions MM, JOR, CMD conceived of the study, MM, LAN, JOR participated in its design, AMS performed the statistical analysis, SBG, CF participated in its coordination, MM drafted the manuscript.

Conflict of Interest The authors declare that they have no conflict of interest.

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