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Reliability and agreement of ages and stages questionnaires®: Results in late preterm and term-born infants at 24 and 48 months

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ABSTRACT

Aim: To evaluate the reliability of the Ages and Stages Questionnaires (ASQ-3) 24 and 48 month intervals translated to Spanish by Brookes Publishing, and the agreement between both questionnaires, comparing late preterm (LPI) and term-born infants (terms).

Methods: Two cohort samples of healthy LPI and terms that were born in a private hospital in Barcelona, Spain. Internal consistency was analyzed by Cronbach's alpha scores and Pearson product- moment correlation between the domain scores and the overall score. The agreement was analyzed using Pearson's correlations between the two questionnaires, and the odds ratio (OR) for positive screening at 48 months, given a positive screen in 24 month assessment.

Results: A total of 473 evaluations were analyzed, representing 331 children. Cronbach's alpha scores for the motor domains on both intervals were low, but acceptable compared with the overall score; a strong positive correlation between the domain and overall score were obtained in the majority of the domains. The correlation between the 24 and 48 month total scores were positive, especially for LPI. Having at least 2 domains in the referral zone at 24 months was associated with an OR of 140 [95% CI 14.85; 3575.65] for positive screen at 48 months.

Conclusion: The Spanish ASQ-3 24 and 48 month intervals appear to be a reliable for developmental screening and for the follow-up of children, especially for LPI. Having two or more domains in the referral zone at 24 months screening is a significant predictor of developmental delay risk at 48-month questionnaire.

1. Introduction

Early childhood neurological development is a determining factor in behavior and learning throughout life [1]. There is evidence suggesting that early detection of developmental delay (DD) allows for timely, effective intervention [2,3]. Early intervention can allow improving the learning and behavior alterations that LPI can present, modifying the natural history of these alterations [4,5]. The identification of children with subtle DD represents a real challenge for pediatricians, as it has been shown that clinical impression is subjective and not sensitive to this target [6]. For this reason, the American Academy of Pediatrics (AAP) recommends the application of standardized developmental screening tools during the health monitoring of infants and preschool children at specific ages (9, 18, and 30–36 months), and especially when certain well-known risk factors are at play, such as premature birth, and genetic or metabolic problems [7]; while in European countries, like the Healthy Child Programme of England, recommend the application of screening tests and developmental surveillance, by the time the child is one year old and between two and two-and-a-half years old [8].

Despite the existence of health monitoring policies that emphasize the importance of standardized screening tools for developmental assessment, fewer than 50% of pediatricians in the United States (U.S.) routinely use them [9]. A number of development screening tools have been developed; they may be distinguished by the conditions they assess, their cultural adaptation, and their means of application. Of note among these are the parent-completed developmental screening tools, which have seen increasing use given their low cost, their ease of use, and the empowerment they provide to the adults responsible for the care and stimulation of the children [10,11].

One of the parent-completed developmental screening tool in wide use in the U.S, is the *Ages and Stages Questionnaires* (ASQ) which was

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developed at the University of Oregon in the 1980s and then revised in 2009, as the ASQ3 [12,13]. These questionnaire has been adapted for use in several countries [14,15,16,17,18,19,20] including the region of Galicia in Spain, were the ASQ completed by the educators was studied [21]. The ASQ has been shown to be valid and reliable both in screening children born at full term and in the monitoring of children with biological risk factors [22,23].

In primary care in Spain, the pediatrician is responsible for monitoring child development in health controls, following clinical protocols [24], as a result of which there is a risk of under-diagnosis of children with subtle problems of development, as it has been noted in published reports6. The late preterm infant (LPI) follow-up group of the Spanish Neonatology Society (SEN34-36) has proposed the use of simplified neurodevelopment questionnaires, with the use of ASQ-3 at two years of age [25].

In the Hospital de Barcelona-SCIAS, the ASQ-3, translated into Spanish by Brookes Publishing, was introduced for use in 2013 in an LPI cohort at 24 and 48 months and for term-born infants. We selected the age of 2 years based on the recommendation other European countries, like England [8], and the policies of SEN34–36 in Spain [25]. The follow up at 48 months was chosen based in fact that this age is considered the last step for the detection of subtle delays that can benefit from intervention [26]. In our previous studies it was shown that the ASQ-3 allowed for the identification of those at risk [27,28], however, no reliability studies for the ASQ-3 had been carried out in this population. In addition, few studies have analyzed the effects of biological risk factors, such as premature birth, on the psychometric properties of the test and on the developmental pathways of the children.

The aim of the present study, then, was to evaluate the reliability of the Ages and Stages Questionnaires (ASQ-3) 24 and 48 month intervals translated to Spanish by Brookes Publishing, and the agreement between both questionnaires, comparing LPI and terms, in an upper middle class sample in Barcelona, Spain.

2. Methods

2.1. Population

A cohort study was carried out including LPI (GA of $34^{0/7}$ to $36^{6/7}$ weeks) and term-born infants (GA of $37^{0/7}$ to $41^{6/7}$ weeks) born in the private hospital of a healthcare insurance company with a neonatal intensive care unit, in Barcelona, Spain. The socio-economic level of the participants was considered middle-high due to the social characteristics of the population analyzed, assuming that this population could afford an expensive private healthcare center.

We followed two cohort samples: the first of children born from January 1 to December 31, 2009, and the second of children born from January 1 to December 31, 2011. Inclusion criteria were: LPI and term born in the period whose parents were locatable and, after phone contact, agreeable to participation. For the term group, we selected a sample of children born in the hospital at full- term gestational age matched by date of birth with LPI. We included only apparently healthy term-born infants who were followed up by pediatricians belonging to our insurance group.

Exclusion criteria were children with malformative syndromes and with known genetic or metabolic diseases and, in the term group, we also excluded those who were admitted to Neonatal Intensive Care Unit during the neonatal period.

2.2. Measures

Ages and Stages Questionnaires® Third Edition translated to Spanish by Brookes Publishing (ASQ-3) [13] is a validated, parent-completed developmental screening tool. Twenty-one questionnaires are available from 1 to 66 months of age. Parents answer 30 questions covering five domains of development, including communication, gross motor, fine motor, problem solving, and personal-social domain. Each domain contains six questions that can be answered with a 'yes' (10 points), 'sometimes' (5 points), or 'not yet' (0 points); which are summed for a domain total.

Infants were one standard deviation below the mean in any domain were considered on the monitoring zone and if they have two standard deviations below the mean in any domain have positive screen and were considered in referral zone, or at risk of DD, in accordance with the ASQ manual [13].

Parents were instructed to try activities with their children to facilitate accurate evaluation and were offered the option of completing the questionnaire by e-mail or with a home visit by a person other than a healthcare professional. The healthcare professional brought the document to parents and provided advice as needed.

Parents of included preschoolers completed the ASQ-3 48 month interval for the first cohort, and the 24 and 48 month intervals for the second cohort.

All the parents signed an informed consent form. The project was approved by the hospital's teaching and ethics commission.

2.3. Statistical analysis

For descriptive analysis we first assessed the background characteristics of the study samples. We compared the demographic characteristics of the LPI and term groups using central tendency measures and proportions.

We compared mean scores and cutoff scores of ASQ-3 24 and ASQ-3 48 months with those from the U.S. normative data [13]. A difference of > 5 points was considered significant, considering that the ASQ-3 is a range scale with scores increasing in intervals of five [16].

For reliability analysis, internal consistency was measured with the Cronbach alpha coefficient for each of the five domains and for the overall test. Criteria for Cronbach's alpha included: > 0.9 Excellent; > 0.8 Good; > 0.7 Acceptable; > 0.6 Questionable; > 0.5 Poor; and, < 0.5 Unacceptable [29]. Additionally, the Pearson product-moment correlation coefficients between the overall score and the total scores for each domain were analyzed.

The agreement between the two interval questionnaires was measured separately for the LPI and terms; we analyzed the correlations of the scores between both intervals separately, and, finally, we examined the capacity of positive screen at 24 month to predict poorer performance at 48 months, using logistic regression analysis.

3. Results

1) General information

The first cohort included 179 children who were assessed with ASQ-3 at the age of 48 months. In the second cohort a total of 152 children were assessed at 24 months; of these, 142 were followed up and re-assessed at the age of 48 months. The total, then, consisted of 473 evaluations made of 331 children (see Fig. 1).

The bio-demographic characteristics of the sample are presented in Table 1. In the LPI group the median gestational age was 36 weeks (range 34–36) while in the term group it was 39 weeks (range 37–41). There were significant differences in birth weight; 61.9% of the LPI group were hospitalized in the Neonatal Intensive Care Unit while no children from the term group were admitted (in accordance with the exclusion criteria for the study), and the percentages of caesarean sections and twin births were significantly higher for the LPI group compared to the term group. The 84,5% of mothers of the term sample and 77,3% of the LPI sample report university education.

2) Comparison of study sample ASQ-3 mean scores with published U.S. normative data

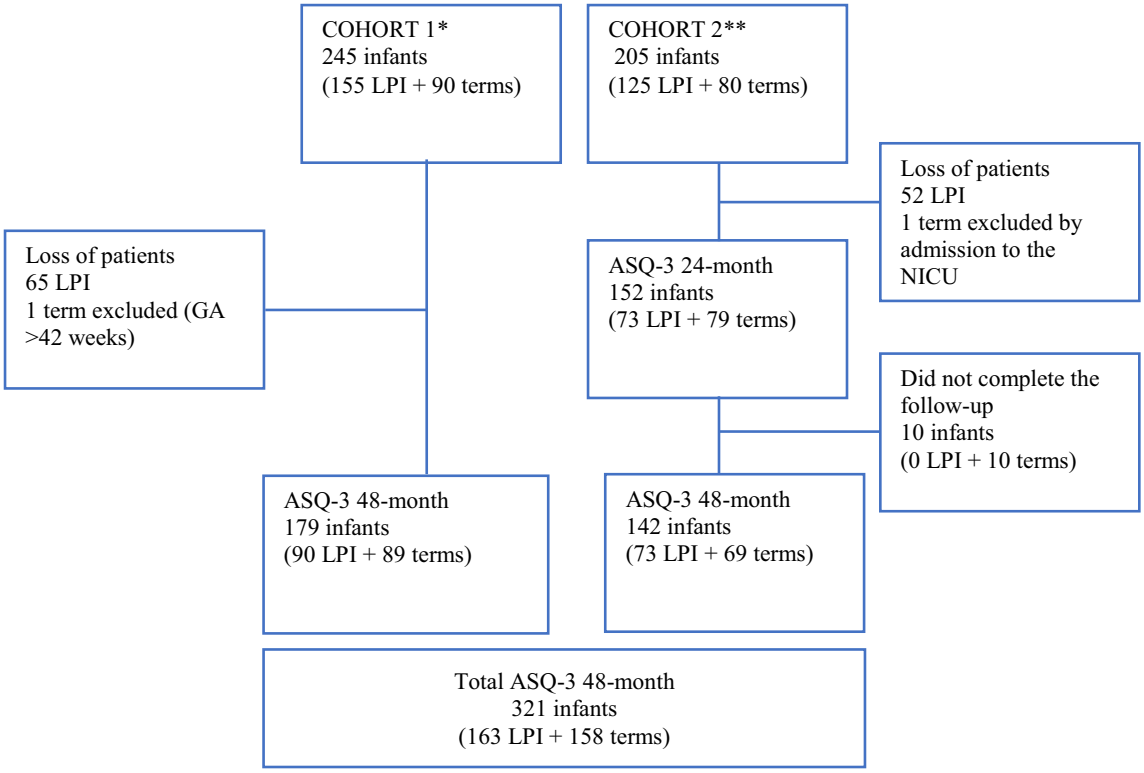

Fig. 1. Flowchart showing the number of eligible children in the final study population.
*COHORT 1: infants born at term and late preterm between 1st January and 31st December 2009.
**COHORT 2: infants born at term and late preterm between 1st January and 31st December 2011.
LPI: late preterm infants; terms: term-born infants; NICU: Neonatal Intensive Care Unit.

Table 1
Analysis of bio-demographic and perinatal variables of the sample.

	Terms	LPI	P
N	168	163	
Birth weight (g) X ± SD	3260.8 ± 421.0	2464.71 ± 420.2	< 0.0001
Gestational age (w) (median and rank)	39,2 (37–41)	36 (34–36)	< 0.0001
Male gender (%)	96 (57.1)	96 (58.9)	NS
Mother's age (y) X ± SD	37.2 ± 3.5	38.2 ± 4.3	0.03
Father's age (y) X ± SD	38.6 ± 5.2	39.6 ± 5.2	0.08
University mother (%)	142 (84.5)	126 (77.3)	NS
Single parent (%)	3 (1.8)	5 (3.1)	NS
Caesarean section (%)	71 (42.3)	100 (61.3)	0.0004
Twins (%)	3 (1.8)	64 (39.3)	< 0.0001
Neonatal Intensive Care Unit admission (%)	0 (−)	101 (61.9)	< 0.0001

Terms: term-born infants. LPI: late preterm infants. P = p-value.
X = mean; SD = standard deviation.

Table 2 presents the scores and the cutoff for ASQ-3 24 and 48 months for the Barcelona sample in comparison to the U.S. normative data [13]. The scores for ASQ-3 at 24 months are similar to those for the U.S., except for the fine motor domain, which were lower. In contrast, at 48 months the scores for the fine motor domain were higher than those reported in the U.S. normative data. There were no differences in the other domains. However, important differences in cutoff scores were found in fine motor domain at 24 months and in all domains at 48 months, due to the lower standard deviation in Barcelona sample compared with U.S. sample. Cutoff scores were higher in Barcelona sample at 48 months.

It will be noted that in both samples the scores between the 24 and 48 month questionnaires showed a rising trend, except for the gross motor domain in the study sample, which remained stable, and the fine motor domain in the U.S. normative data, in which the scores decreased.

3) Reliability: Cronbach's alpha coefficient

The Cronbach alpha coefficient for the sample's ASQ-3 24 and 48 month scores was computed for each domain and for the overall score. Item deletion did not improve alpha coefficients in any domain. Scores ranged from 0.35 (gross motor at 48 months) to 0.68 (communication at 24 and 48 months). These results indicate that internal consistency ranged from unacceptable to questionable in all domains. But when the overall analysis was made, we obtained Cronbach alphas of 0.78 and 0.79, which indicate acceptable-to-good internal consistency (Table 3).

4) Reliability: Pearson product- moment correlation coefficients between the domain scores and the overall score for each questionnaire

Fig. 2 shows the Pearson product-moment correlation coefficients between the overall and the total scores for each domain, for both ASQ-3 at 24 and 48 months. A strong positive correlation between the domain scores and overall score was obtained, except for fine and gross motor, where the correlations were moderate. The correlations between the various domains was significant, but weakly so. We did not find differences in the correlation pattern between domains and the overall score when we reviewed the ASQ-3 24 and 48 months form separately.

5) Agreement: Correlational analysis at 24 and 48 months

The correlation between the scores for ASQ-3 24 and 48 months was positive and significant for the overall score and for the majority of the

Table 2
Comparison of study sample ASQ-3 mean scores and cutoff scores at 24 and 48 months with US normative data.

		n	Communication		Gross motor		Fine motor		Problem solving		Personal-social	
			Mean/SD	Monitoring zone/ referral zone	Mean/SD	Monitoring zone/ referral zone	Mean/SD	Monitoring zone/ referral zone	Mean/SD	Monitoring zone/ referral zone	Mean/SD	Monitoring zone/ referral zone
ASQ 24 months												
Barcelona sample	ASQ-3 Spanish	152	50 ± 11.4	38.6/27.2	53 ± 7.6	45.4/37.8	**45.5 ± 10**	**35.5/25.5**	48.8 ± 9.6	39.2/29.6	48.8 ± 9.6	39.2/29.6
USA 2009 [13]	ASQ-3	1443	51.2 ± 13	38.2/25.2	54.7 ± 8.3	46.4/38.1	**51.7 ± 8.2**	**43.4/35.2**	49 ± 10	39.6/29.8	51 ± 10	41.3/31.5
ASQ 48 months												
Barcelona sample	ASQ-3 Spanish	321	57.2 ± 6.5	**50.7/44.2**	54.4 ± 7	**47.4/40.4**	**52.4 ± 8.7**	**43.7/35**	56.1 ± 6.6	**49.5/42.9**	55.1 ± 6.6	**48.5/41.9**
USA 2009 [13]	ASQ-3	672	52.9 ± 11.1	41.8/30.7	52.7 ± 9.9	42.7/32.8	45.3 ± 14.7	30.6/15.8	52.7 ± 10.7	42.0/31.3	50.3 ± 11.8	38.5/26.6

n = sample size; ASQ-3 = Ages and Stages Questionnaire Third Edition.
SD = standard deviation.
Bold numbers indicate differences greater than 5 points.

domains. In the LPI group these correlations ran from moderate to high (ranging between 0.51 and 0.72) except for the personal-social domain in which the correlation was not significant. Meanwhile, in the term group the correlations were low (communication, gross motor, personal-social, and overall score) or not significant (fine motor and problem solving) (Fig. 3).

6) Agreement: Risk of developmental delay at 48 months according to risk at 24 monthss

Having a positive screening at ASQ-3 24 months, was not associated with a greater risk of positive screen nor with lower mean scores at 48 months; nevertheless, having two or more domains in the referral zone was associated with the risk of DD at 48 months (OR 140 [95% CI 14.85; 3575.65]) as well as a lower ASQ-3 scores (Table 4 and Fig. 4). We did not find differences between the domains, but rather in the sum. It is of note that there were only children in the LPI cohort with two or more domains below the cut-off on the 24 month interval.

4. Discussion

The scores obtained on the ASQ-3 in this Barcelona sample were similar to those reported in the U.S. normative data. The exceptions to this were in the fine motor domain, with children's scores at 24 months lower than those reported for the U.S. by Squires et al. [13]. By contrast children's scores at 48 months, were higher. In the other domains, the differences were generally small and not clinically relevant. However, the cutoff scores were higher in all domains at 48 months for Barcelona sample, but lower for fine motor at 24 months.

On reviewing the reliability of the ASQ-3 we found questionable internal consistency in the analysis of domains, as reported as well by Lopes et al. in Portugal and Alvik et al. in Norway [14,30]. Although these results might cast a shadow of doubt on the reliability of the test, we should stress that the size of alpha depends on the number of items in the scale, and each domain consists of only 6 questions [31]. Additionally, the items in each domain may ask for very different skills, and would not be expected to be highly correlated such as items in the personal social domain[13.] By the same token, if we consider the overall score, with thirty questions, Cronbach's alpha rises to 0.78/0.79, which is in the acceptable-to-good reliability. In a systematic review of ASQ-3 2–2.5 years, Velikonja et al. demonstrated that the English version of the questionnaire, based on the medians of the five ASQ-3 domains, had positive values for internal consistency (Cronbach's alpha > 0.70), with variability in the results of the domain analysis and less reliability in the translated/adapted versions of the ASQ-3 [32].

Another way of analyzing the reliability of a questionnaire is by the Pearson product- moment correlation between the domain scores and the overall score. With this analysis we found correlations between the domains and the overall score to be moderate-to-high, as was found by Lopes et al. in the validation of ASQ in Portugal [14], while the correlation among the domains was low, which is to be expected, given that they mark out separate domains.

In our study we analyzed a cohort of children who were assessed at 24 and 48 months of age, and we found that the scores obtained at 48 months were higher than those at 24 months. Correlations were significant between overall scores of both questionnaires, with greater correlation for the LPI than for the term. Few studies have examined the developmental trajectories of children using ASQ, and the methodologies employed have been diverse. Valla et al. [33] found, using ASQ in Norway, that a majority of children showed positive and stable normative developmental pathways from 4 to 24 months of age, while a small percentage of children had decreasing scores or other patterns of development.

Our study was not designed to establish development pathways, as there are only two measurements made by the ASQ-3. Nonetheless, it is possible to assess the stability of the risk of DD, with a very significant

Table 3
Reliability analysis for each domain and overall score using Alpha Cronbach.

	Communication	Gross motor	Fine motor	Problem solving	Personal-social	Overall score
24-month (n = 152)	0.68 (0.60–0.74)	0.43 (0.31–0.56)	0.37 (0.24–0.51)	0.37 (0.22–0.53)	0.51 (0.39–0.63)	0.79 (0.74–0.84)
48-month (n = 321)	0.68 (0.63–0.73)	0.35 (0.25–0.46)	0.54 (0.46–0.61)	0.5 (0.42–0.58)	0.46 (0.37–0.54)	0.78 (0.75–0.82)

n = sample size.

OR when there were two or more domains in the referral zone on ASQ-3 at 24 months, which only occurred in the LPI group. In this regard, we did not find differences between the domains.

Previous studies have reported an increase in the psychometric properties of the test in higher biological risk samples [22], which may be explained in part by the effect of extreme values and the increased stability of the delays. Hornmann et al. [34] found that overall development of moderately-late preterm children had stability patterns comparable to term-born children at 4 to 5 years of age, probably because when the children advance in their age, the DD have achieved greater stability. Studies have shown that ASQ-3 has a high negative predictive value; that is, if a child does have a negative screen, they are less likely to manifest risk in development over the medium and long term. The positive predictive value, however, is low [35,36,37], probably because the more subtle problems in development may be resolved with time while there is a lesser likelihood of other problems appearing, especially with increasing age in the child.

The strongest point of this study is that it analyzed two child cohorts of terms and LPI—with high levels of and patient participation and adherence. However, one of the limitations of the study is that it did not consider the possible interventions undergone by the children. Another limitation is that the follow-up sample was small, increasing the possibility of false negative results. For these reasons it seems opportune to continue the follow-up of children for a longer time period in order to learn more about the developmental pathway and about the impact of early detection of subtle developmental problems on learning and school performance. In addition, these studies should be replicated with larger more economically diverse families.

5. Conclusions

The ASQ-3 24 and 48 months in Spanish appears to be a reliable tool for developmental screening and follow-up of children between 2 and 4 years of age, especially in the LPI group. A positive agreement was found for the two questionnaires analyzed. Having two or more domains in the referral zone on the 24 months screening is a significant predictor of the risk of DD risk at 48 months of age.

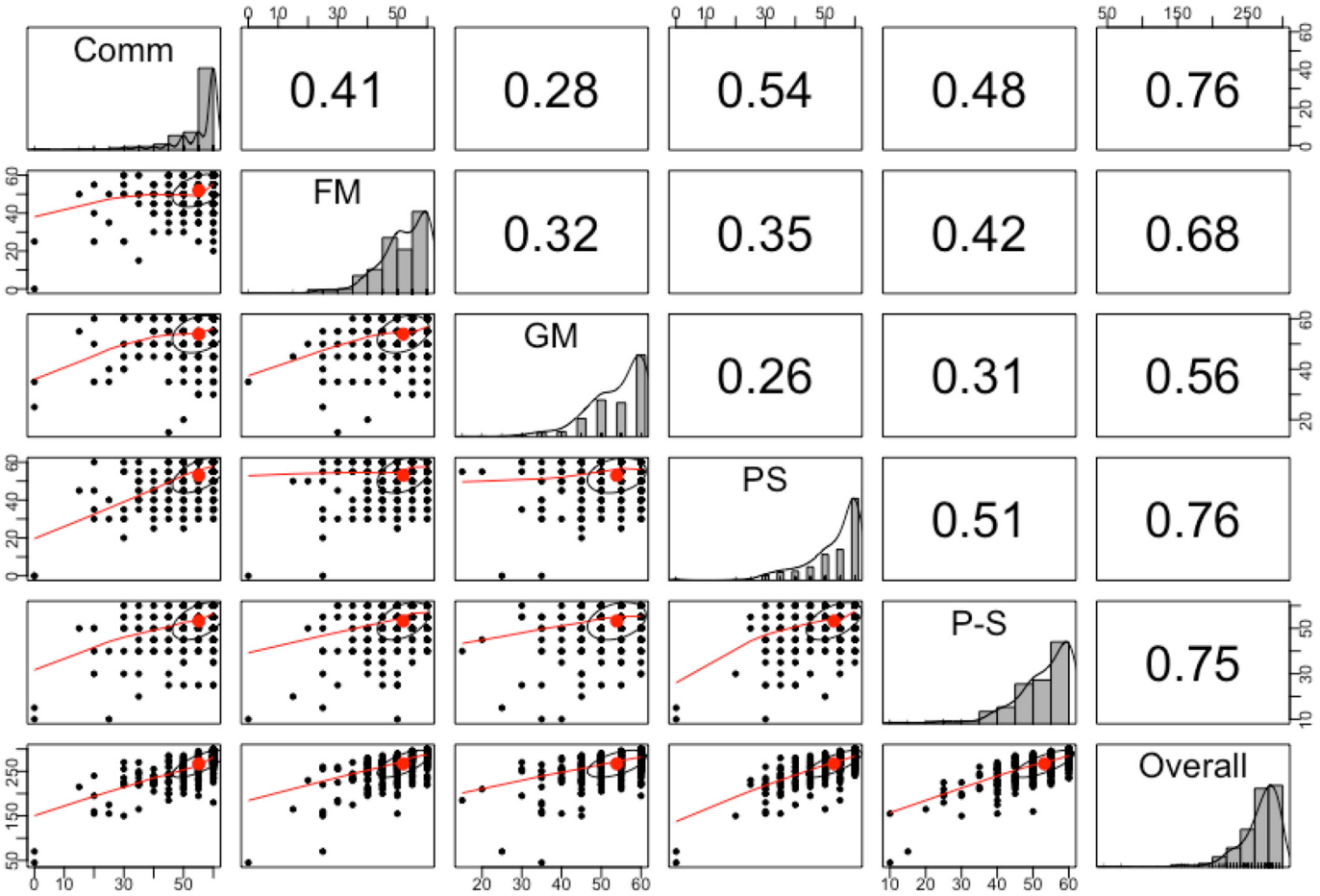

Fig. 2. Pearson product- moment correlation matrix between the domain scores and the overall score on 24- and 48-month questionnaires.
All correlations are significant at $p < 0.001$.
Comm: communication; FM: fine motor; GM: gross motor; PS: problem solving; P-S personal-social.

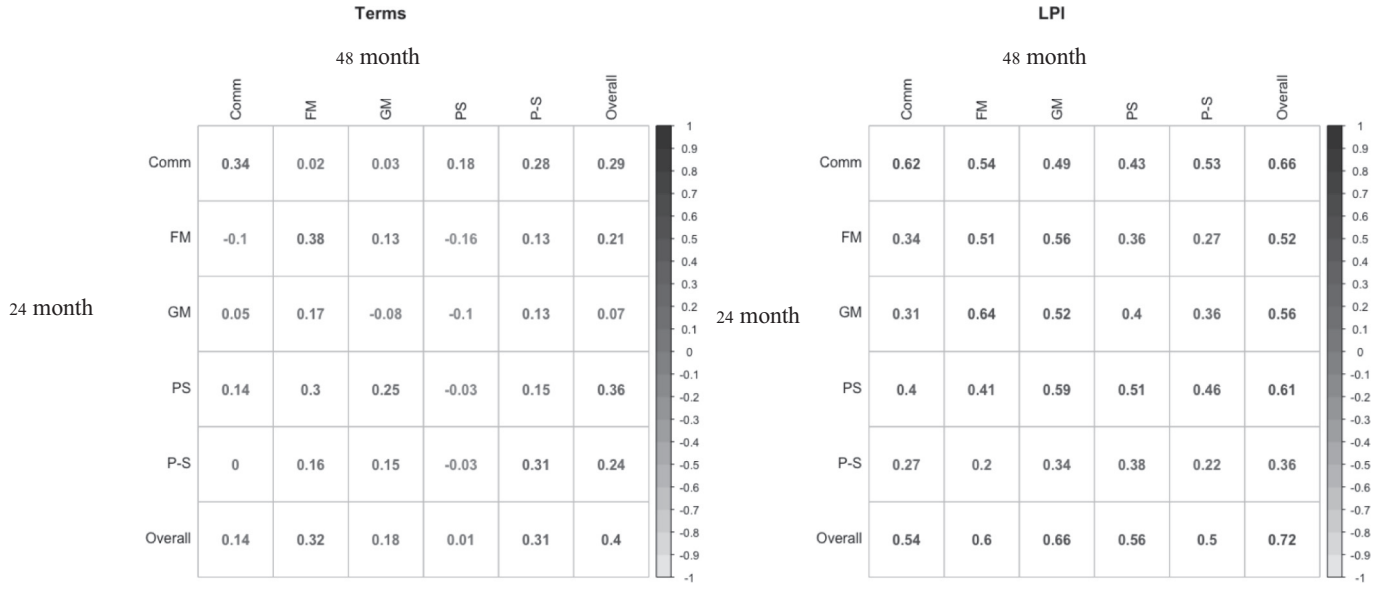

Fig. 3. Correlation for domains and overall scores at 24 and 48 months in term-born and late preterm infants.
Comm: communication; FM: fine motor; GM: gross motor; PS: problem solving; P-S personal-social; terms: term-born infants; LPI: late preterm infants.

Table 4
Odds ratio for risk on developmental delay at 48 month, according 24 month assessment, in term-born and late preterm infants.

	Terms	95% CI	LPI	95% CI
1 domain in referral zone	5.89	[0.68; 43.32]	5.60	[0.24; 69.85]
2 or more domains in referral zone			140.00***	[14.85; 3575.65]

Developmental delay risk = at least 1 domain in referral zone.
Referral zone: domain score > −2 SD below the mean score.
LPI: late preterm infants; terms: term-born infants; CI: confidence interval.
In the footnote is the definition for the asterisk: *p > 0.05.

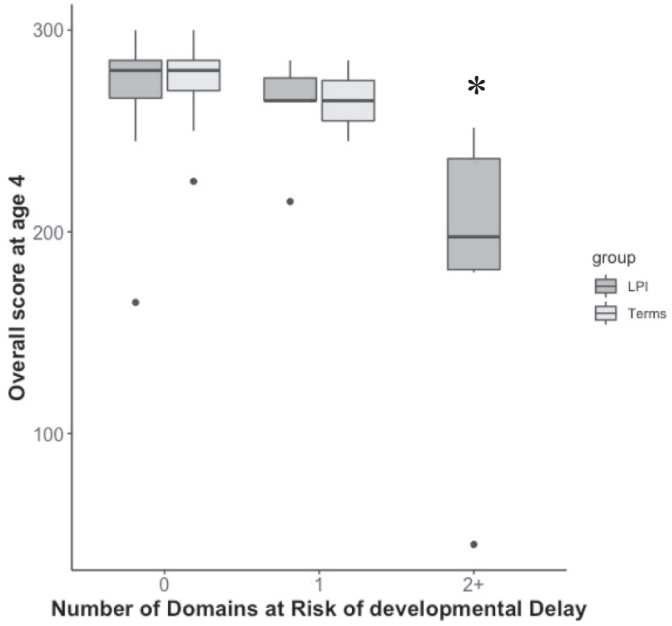

Fig. 4. Comparison of ASQ-3 48-month overall score according to number of domains under cut-off point at 24 months, in term-born and late preterm infants.
0: no domains in referral zone; 1: one domain in referral zone; 2+: two or more domains in referral zone.
*p < 0.05. LPI: late preterm infants; terms: term-born infants.

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