DOI: 10.26387/bpa.2023.00009

Cognitive, language and motor development in toddlers with Down syndrome: Beyond the floor effect of Italian BSID–III scores

Lina Pezzuti¹, Rosa Ferri¹, Elvira Caramuscio¹, James Dawe¹, Anna Scala²

¹ Department of Dynamic and Clinical Psychology and Health, Sapienza University of Rome, Italy ² Aipd – Associazione Italiana Persone Down, Sezione Roma Onlus, Italy

lina.pezzuti@uniroma1.it

. ABSTRACT. La valutazione dello sviluppo delle abilità cognitive, linguistiche e motorie di bambini con sindrome di Down è spesso caratterizzata da profili piatti, con i punti di forza che sono oscurati dai predominanti punti di debolezza. Nel presente studio è stato indagato, su un campione di 144 bambini con sindrome di Down, un modo alternativo di convertire i punti grezzi in punti ponderati della *Bayley Scales of Infant and Toddlers Developmental–III (BSID–III)*. I risultati hanno evidenziato due punti di forza: le abilità comunicative espressive e ricettive.

• SUMMARY. Down syndrome is the most common genetic cause of intellectual disability but there are few studies on the development of toddlers with this syndrome. Considering the importance of early interventions aimed at improving the abilities of this population, the purpose of the present study is to assess the cognitive, language, and motor development of a group of 144 Italian toddlers with Down syndrome aged 12–36 months (individually matched for gender, chronological age, and parental education level with typical development toddlers) through the use of the Bayley Scales of Infant and Toddlers Development–III (BSID–III). The assessment of cognitive, language, and motor development of people with low abilities is far from being easy because it is very frequent to find flat profiles, where strengths are obscured and weaknesses are predominant. So, an alternative method of converting raw scores to weighted scores was used to overcome the floor effect of BSID–III scores. Results showed an increase in cognitive and communicative delay evident as early as the second and third years of age and a developmental profile of toddlers with DS characterized by strengths in expressive and receptive communication skills. Results were discussed in reference to the literature in recent years.

Keywords: Down syndrome, BSID–III, Cognitive development, Language development, Motor development, Toddlers

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INTRODUCTION

Down syndrome (DS) is the most common genetic cause of intellectual disability and it is a clinical manifestation of chromosome abnormality, known as trisomy 21, which is characterized by a range of physical, intellectual, and clinical symptoms (Contestabile, Benfenati & Gasparini, 2010; Ferreira-Vasques & Lamônica, 2015; Grieco, Pulsifer, Seligsohn, Skotko & Schwartz, 2015; Roizen & Patterson, 2003). Despite several individual differences (Ferreira-Vasques & Lamônica, 2015; Karmiloff-Smith et al., 2016; Roizen & Patterson, 2003), the literature points to an atypical general development profile, involving cognitive, language, motor, self-care and personal-social domains (de Lima Freire, de Melo, Hazin & Lyra, 2014; Ferreira-Vasques & Lamônica, 2015; Karmiloff-Smith et al., 2016 Roizen & Patterson, 2003). The cognitive profile of DS population is characterized by an IQ between 30 and 70 with a mild to severe delay and which seems to increase with age (Aoki, Yamauchi & Hashimoto, 2018; Godfrey & Lee, 2018; Grieco et al., 2015; Karmiloff–Smith et al., 2016; Patterson, Rapsey & Glue, 2013; Robert & Richmond, 2015; Wester Oxelgren et al., 2018; Yang, Conners & Merrill, 2014).

Although DS has been extensively studied in the past, what we know about the syndrome mainly concerns the adult population, while there are few studies on samples of school–age children, particularly pre–school children (Karmiloff–Smith et al., 2016; Grieco et al., 2015; Naranajo & Robles–Bello, 2020; Needham, Nelson, Short, Daunhauer & Fidler, 2021; Patterson et al., 2013). For what concern cognitive development of toddlers with DS, Milojevich and Lukowski (2016) suggested the presence of cognitive delay as early as preschool age. In agreement with this study a longitudinal research by Aoki et al. (2018) suggested that cognitive, language, and motor developmental delays are evident as early as preschool age and consist of a slower rate of development in children with DS than that in children with typical development (TD).

Concerning language development delay, it would become more evident with the onset of the complex language acquisition phase (Abbeduto, Warren & Conners, 2007; Levy & Eilam 2013). Furthermore, non-verbal communication skills and receptive language would be less compromised than expressive language. (Abbeduto et al., 2007; Bello, Onofrio & Caselli, 2014; Caselli et al., 1998; Eggers & Van Eerdenbrugh, 2018; Ferreira–Vasques & Lamônica, 2015; Galeote, Sebastian, Checa, Rey & Soto, 2011; te Kaat-van den Os, Volman, Jongmans & Lauteslager, 2017; Mason–Apps, Stojanovik, Houston–Price, Seager & Buckley, 2020; Næss, Lyster, Hulme & Melby–Lervåg, 2011; Patterson et al., 2013; te Kaat–van den Os, Jongmans, Volman & Lauteslager, 2015; Witecy & Penke, 2017; Zampini & D'Odorico, 2011).

Regarding motor skills, the developmental trajectories of fine and gross motor skills of children with DS are similar to children with typical development, but progress is much slower (Aoki et al., 2018; Cardoso, de Campos, Dos Santos, Santos & Rocha, 2015; Ferreira-Vasques & Lamônica, 2015; Kim, Kim, Jeon & Jung, 2017; Malak, Kostiukow, Krawczyk-Wasielewska, Mojs & Samborski, 2015; Tudella, Pereira, Basso & Savelsbergh, 2011). DS is characterized by a general impairment in motor skills with difficulties in motor planning and coordination, difficulties in fine motor skills, and a delay in achieving developmental milestones (Alesi & Battaglia, 2019; Ferreira-Vasques & Lamônica, 2015; Frank & Esbensen, 2015). A recent systematic review by Needham et al. (2021), with the aim of analyzing early developmental characteristics of fine-motor skills in children with Down syndrome, revealed important individual differences in the development of these skills that, combined with the low number of studies present in literature, makes further research necessary to obtain more complete information about developmental trajectories and to plan appropriate interventions (Needham et al., 2021).

The most commonly used standardized instrument for the assessment of the development of toddlers aged between 0 and 42 months is the Bayley Scales of Infant and Toddlers Development - Third Edition (BSID-III; Bayley, 2006; Italian adaptation by Ferri, Orsini, Rea, Stoppa & Mascellani, 2015). This instrument is widely used in clinical settings to identify young children with developmental delay and to assist the specialist in intervention planning. However, few studies in literature used the BSID-III to investigate clinical populations and only some scales of BSID-III often are used (i.e. Cardoso et al., 2015). This, perhaps, because the American BSID-III (Bayley, 2006) tends to underestimate developmental deficits due to the mixed sample used for the standardization of the instrument (characterized not only by toddlers with typical development but also by premature toddlers, toddlers with Down syndrome, cerebral palsy, pervasive developmental disorders, language disorder and toddlers with atypical developmental risks), which leads to lower normative averages, increased standard deviation scores and decreased accuracy of the assessment (Anderson & Burnett, 2017). In contrast, the Italian normative sample of BSID–III (Ferri et al., 2015) is characterized entirely by children with typical development and this could explain the flattening of the Italian BSID–III scores (Ferri, Carleschi, Mascellani, Coatti & Stoppa, 2005), a fairly common issue when using standardized instruments to assess clinical populations (i.e. Laghi et al., 2022; Pezzuti et al., 2018). However, the BSID–III scales also show some strengths, as they allow to assess toddler's functioning through direct observation of his/her behavior and to draw up a developmental profile characterized by possible strengths and weaknesses, which is essential for planning personalized interventions, focused on the real needs of children (Ferri et al., 2015).

The literature states that we are far from being able to delineate a typical developmental profile of DS: we can describe a general impairment, but how the delay is manifested, at different stages of development and within different developmental domains, is still a debated issue (Karmiloff-Smith et al., 2016; Needham et al., 2021; Patterson et al., 2013). So, given the small number of studies on the developmental characteristics of DS toddlers and the importance of early intervention to improve the development and quality of life of this population, the main objective of the present study is to investigate the cognitive, language, and motor development of toddlers with DS aged 12-36 months, assessed by the Italian version of the Bayley Scales of Infant Development - Third Edition (BSID-III; Ferri et al., 2015). To overcome the floor effect of BSID-III scores, an alternative method of converting raw scores to weighted scores proposed by Hessl et al. (2009) and extended by Orsini, Pezzuti and Hulbert (2015) was used, which recovered intra- and interindividual variability in scores.

The present study attempts to test the following hypotheses:

- Since the literature has shown that developmental delay increase with age, (Aoki et al., 2018; Grieco et al., 2015; Patterson et al., 2013), we assume that this can also be observed in the first three years of life, assuming BSID– III scores of DS toddlers aged 12–24 months to be higher than those obtained by DS toddlers aged 25–36 months.
- The developmental profile of toddlers with DS could be characterized not only by deficits and weaknesses, but also by strengths, which could be identified in gestural communication skills and comprehension language (Abbeduto et al., 2007; Mason–Apps et al., 2020).

MATERIALS AND METHODS Participants

Participants were recruited from the Italian Association of Down People of Rome Onlus (Associazione Italiana Persone Down, AIPD, Roma Onlus) and parents of toddlers gave their authorization through an informed consent process. The clinical group included 144 Italian full-term toddlers with DS diagnosis (82 males and 62 females), without serious health problems (i.e. leukemia and heart disease requiring surgery) aged 12-36 months (Mean = 23.98, SD = 7.35). They were compared with a group of toddlers with typical development (TD; Control Group), who were part of the Italian BSID-III standardization sample, individually matched for gender, chronological age, and years of education parents (we consider the higher number of years of education between the two parents) (see Table 1). So, the two groups (DS and TD) included the same numbers of males and females and they were perfectly matched for age in months and days (Mean age_{Down group} = 23.98, SD age_{Down group} = 7.35; Mean $age_{Control group} = 24.01$, SD $age_{Control group} = 7.27$; $t_{(286)} = .035, p = .972$, Cohen's d = .00) and years of education parents (Mean $edu_{Down group} = 15.33$, SD $edu_{Down group} =$ 2.57; Mean edu_{Control group} = 15.33, SD edu_{Control group} = 2.57; $t_{(286)} = .000, p = 1.000$ Cohen's d = .00). The full sample was divided into two age groups to observe any developmental differences between 2nd year (12-24 months) and 3rd year (25-36 months) of life. The study was approved by the local ethical committee at Sapienza University of Rome.

Instrument

 Bayley Scales of Infant and Toddlers Development – Third Edition (BSID–III; Bayley, 2006; Italian adaptation by Ferri et al., 2015). The Italian standardization of BSID–III was carried out on a sample of 1,050 typically developing toddlers (544 males and 506 females) aged between 12 months and 15 days and 42 months and 14 days (Ferri et al., 2015). The psychometric reliability of the five subscales of the Italian BSID–III (Cognitive, Expressive communication, Receptive communication, Fine–motor, and Gross–motor subscales) vary between .87 (Fine– motor) and .94 (Expressive communication). The reliability

Crowns		Age (me	onths)	Parental education (years)			
Groups	N	М	SD	М	SD		
Total toddlers with TD	144	24.01	7.28	15.33	2.57		
Females with TD	62	24.37	6.97	15.50	2.52		
Males with TD	82	23.74	7.53	15.20	2.62		
Total toddlers with DS	144	23.98	7.35	15.33	2.57		
Females with DS	62	24.39	6.99	15.42	2.52		
Males with DS	82	23.68	7.64	15.26	2.62		

Table 1 – Sample size, mean, and standard deviation of age and parental education of TD and DS groups and gender groups

of composite scores varies between .94 (Linguistic composite scores) and .92 (Motor composite scores). These values were similar to the USA standardizations.

The Italian BSID–III test results are expressed through standardized cognitive, language, and motor development scores with a mean of 10 and a standard deviation of 3. Because of the global developmental delay that is typical of toddlers with DS, we started the BSID– III test with the items that were usually appropriate for children younger than the chronological age of our subjects (Ferri et al., 2005).

Data analysis

- BSID-III traditional weighted scores. We computed *t*-tests for the independent groups and the standard differences (i.e., effect sizes) to compare clinical and control groups on each dependent variable of BSID-III and, to compare age-groups, separately, for DS and TD groups. The effect size (e.g. Cohen's d) and statistical significance (*p*-value) are reported. For the interpretation of Cohen's *d*, we used Hyde's (2005) guidelines: small effect (.11<*d*<.35); moderate effect (.36 <*d*<.65), large effect for (.66<*d*<1.00), or very large effect (*d*>1.00). Repeated measures ANOVA tests (with η^2 as a measure of effect size: .01 small effect, .06 medium effect and .14 for large effect) with Bonferroni correction are calculated in order to compare the subjects within the clinical group with DS and TD group on 5 BSID–III subscales, separately.

New weighted scores (Hessl et al., 2009; Orsini et al., 2015). Using the method proposed by Hessl et al. (2009) and extended by Orsini et al. (2015), the raw scores of subtests, obtained by the group with DS, that correspond to a weighted score (ws) of 1 were transformed into z-points using means and standard deviations of the raw scores of the subtests for each age group. All other weighted scores higher than 1 were transformed into z-points using the inverse formula of their composition in the standardization process: z = (ws - 10)/3. In this way, the resulting measures of the weighted scores are expressed in Z scores, and therefore, the floor effect presented by a minimum score of 1 was overcome. For more detailed information about this statistical method, see Orsini et al. (2015) and Pezzuti et al. (2018). For example, a toddler of 24 months of age gets the raw scores reported in column A of Table 2 to the 5 BSID-III subscales (Cognitive, Expressive, Receptive, Finemotor and Gross-motor). Using the traditional method, such raw scores would be all converted to a weighted score of 1 (see column B) corresponding to a normalized z point of -3 (see column C); conversely, using the means and standard deviations of the age-relevant reference sample of the subject (see column D), the new z points and new weights score are lower (see column E and F respectively)

	Α	В	С	D	Е	F
Subscales	Raw score (<i>x</i>)	Range of raw score corresponding to a weighted score (ws) of 1	z normalize d scores = (ws - 10)/3	Mean (M) and standard deviation (SD) of the raw scores of the subtests on group of 24 months of age (N = 1050)	<i>z</i> new points (zn) with Orsini et al. (2015) method: z = (x - M)/SD	New weighted scores (zn*3)+10
Cognitive	45	0-45	-3	64.0 (6.8)	-2.79	-8
Receptive	13	0-13	-3	29.0 (4.3)	-3.72	-1
Expressive	2	0–16	-3	28.0 (5.8)	-4.48	-3
Fine-motor	20	0-31	-3	40.0 (3.0)	-6.67	-10
Gross-motor	35	0–45	-3	57.0 (3.5)	-6.29	-9

Table 2 - Example of computing new standard scores on data of toddlers of 24 months s of age

and we can reasonably conclude that the most impaired abilities in this toddler are motor skills.

We computed *t*-tests for the independent groups and the standard differences (i.e., effect sizes) to compare the age in the group with DS, and repeated measures ANOVA tests (with η^2 as a measure of effect size) with Bonferroni correction are calculated in order to compare the subjects within the clinical group with DS on 5 BSID–III subscales.

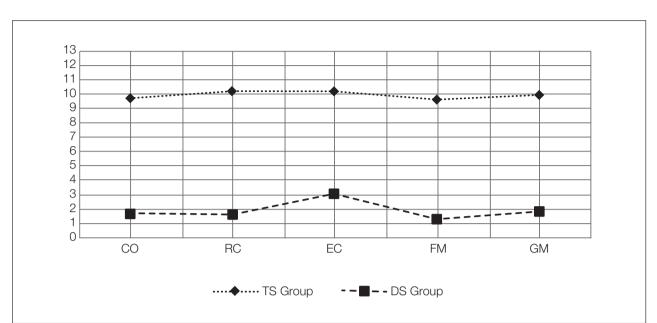
RESULTS

Results using the traditional BSID-III method of converting raw scores into weighted scores (traditional method). In comparison with the Typical Development (TD) group (n = 144), the Down syndrome (DS) group (n = 144) had considerably lower traditional weighted scores in all measures of BSID-III (see Table A in Supplementary material and Figure 1).

We then compared the scores of each BSID-III measure

with the other four measures in the DS group. The ANOVA effect–sizes (η^2) of results reported in Table 3 show a large effect ($\eta^2 > .14$) for the following comparisons: Cognitive and Expressive communication subscales with the first tending to be lower; Receptive communication and Expressive communication subscales with the former tending to be lower; Expressive communication and Fine–motor subscales with the former tending to be higher. A medium effect ($.06 < \eta^2 < .14$) emerged for the comparison of Expressive communication and Gross–motor subscales with the former tending to be higher. In sum, for the group with DS the Expressive communication subscale tends to be higher than other ones, while Fine–motor subscales tend to be lower than Expressive communication.

Table B (reported in Supplementary material) and Figure 2 show the comparison between two age cohorts in all BSID–III subscales for both the TD and DS groups. In particular, the results highlight: a higher score for the TD group aged 12–24 months than for the TD group aged 25–36 months on the Gross–motor (GM) subscale; and a



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Legenda. CO = Cognitive scale; RC = Receptive communication; EC = Expressive communication; FM = Fine-motor; GM = Gross-motor.

Comparison between pairs of 5 BSID–III _ subscales	[]	1]	[/	2]	Repeated measures Anova test and group mean comparisons			
	M_{1}	SD ₁	<i>M</i> ₂	SD ₂	Diff.	<i>F</i> _(1,143)	Р	η^2
CO _[1] <i>vs</i> RC _[2]	1.68	1.21	1.63	1.18	.05	.23	.630	.04
CO _[1] <i>vs</i> EC _[2]	1.68	1.21	3.03	1.58	-1.35	96.17	<.001	.19
CO _[1] <i>vs</i> FM _[2]	1.68	1.21	1.31	.81	.37	11.85	<.001	.03
CO _[1] vs GM _[2]	1.68	1.21	1.81	1.52	13	.88	.349	.00
RC _[1] vs EC _[2]	1.63	1.18	3.03	1.58	-1.40	97.75	<.001	.20
RC _[1] vs FM _[2]	1.63	1.18	1.31	.81	.32	9.33	.003	.02
RC _[1] vs GM _[2]	1.63	1.18	1.81	1.52	17	1.48	.226	.00
EC _[1] vs FM _[2]	3.03	1.58	1.31	.81	1.72	144.48	<.001	.32
EC _[1] vs GM _[2]	3.03	1.58	1.81	1.52	1.22	59.98	<.001	.13
FM _[1] vs GM _[2]	1.31	.81	1.81	1.52	49	14.22	<.001	.04

Table 3 – Profiles of the group with DS: comparisons between pairs of five BDSI–III scales using traditional weighted scores

Legenda. CO = Cognitive scale; RC = Receptive communication scale; EC = Expressive communication scale; FM = Fine-motor scale; GM = Gross-motor scale.

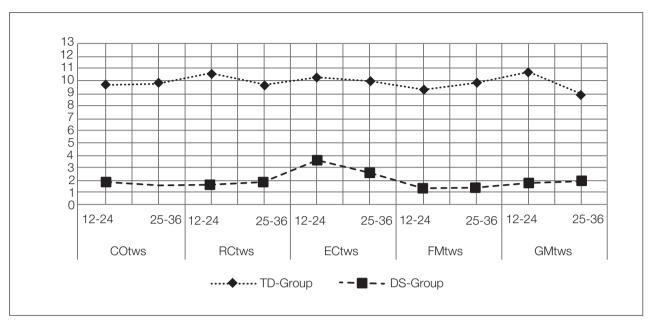


Figure 2 - BSID-III age-profiles of DS and TD groups using traditional weighted scores (tws)

Legenda. CO = Cognitive scale; RC = Receptive communication; EC = Expressive communication; FM = Fine–motor; GM = Gross–motor.

higher score for the children with DS aged 12–24 months than for the group with DS aged 25–36 months on the Expressive communication (EC) subscale.

 Results with an alternative method of converting raw scores into weighted scores (Hessl et al., 2009; Orsini et al., 2015). Using the traditional transformation of the raw scores according to the conversion tables of the BSID–III Italian standardization (Ferri et al., 2015), a very high percentage of subtests with a weighted score of 1 at the five BSID–III subscales, occurred in the sample. In details, there were subscales that, more than others, had a weighted score of 1, such as Fine–motor (84.0%), followed by Receptive communication (68.8%), Gross–motor (69.4%), Cognitive (69.4%), and Expressive communication (25.0%).

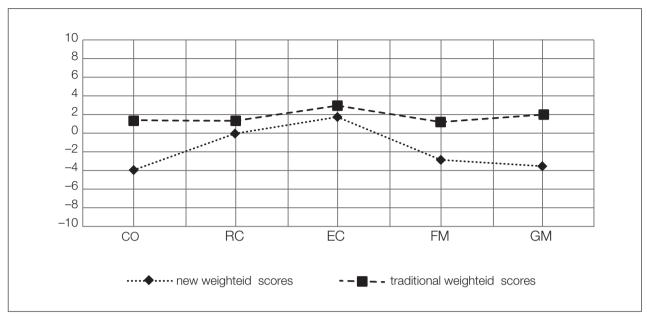
Thus, a second and new transformation of the raw scores was performed using the method proposed by Hessl et al. (2009) and extended by Orsini et al. (2015), following the steps shown in Table 2. Figures A–E reported in Supplementary material show the weighted scores according to the two methods (traditional and new

transformation) for the sample of 144 toddlers on the five BSID–III subscales. Compared with the traditional method, the new method increased intra– and inter– individual variability of scores.

– Results of the study of BSID–III profiles using the new method on DS sample. Figure 3 shows the BSID–III profiles of the DS sample with two kinds of scores and while the profile of the traditional weighted scores is fairly flat, the profile of the new weighted scores shows more variability between the five BSID–III measures, highlighting strengths (e.g. in EC and RC) and weaknesses (e.g. in CO and GM).

From repeated measures ANOVA tests on new weighted scores (see Table 4), to compare all scores of each measure with the other four BSID–III measures, a medium (.06< η^2 <.14) and large (η^2 >.14) effect emerged for the following comparisons between: Cognitive and Receptive communication subscales with the former subscale tends to be lower; Expressive communication and Cognitive subscales with the former tends to be higher; Receptive communication and Expressive communication





Legenda. CO = Cognitive scale; RC = Receptive communication; EC = Expressive communication; FM = Fine–motor; GM = Gross–motor.

Comparison between pairs of 5 BSID–III subscales	[2	1]	[2	2]	Repeated measures Anova test and group mean comparisons			
	M_{I}	SD ₁	M_2	SD ₂	Diff.	<i>F</i> _(1,143)	р	η^2
CO _[1] <i>vs</i> RC _[2]	-3.80	6.20	25	3.07	-3.55	79.5	<.001	.12
CO _[1] <i>vs</i> EC _[2]	-3.80	6.20	2.01	3.61	-5.81	152.04	<.001	.25
CO _[1] vs FM _[2]	-3.80	6.20	-2.43	4.36	-1.37	7.49	.007	.02
CO _[1] vs GM _[2]	-3.80	6.20	-3.75	6.23	05	.01	.927	.00
RC _[1] vs EC _[2]	25	3.07	2.01	3.61	-2.26	66.00	<.001	.10
RC _[1] vs FM _[2]	25	3.07	-2.43	4.36	2.18	34.11	<.001	.08
RC _[1] vs GM _[2]	24	3.07	-3.75	6.23	3.51	54.36	<.001	.11
EC _[1] vs FM _[2]	2.05	3.58	-2.43	4.36	4.49	96.06	<.001	.24
EC _[1] vs GM _[2]	2.05	3.58	-3.75	6.23	5.81	122.04	<.001	.24
FM _[1] vs GM _[2]	-2.43	4.36	-3.75	6.23	1.32	6.00	.016	.01

Table 4 - Profile of DS: comparisons between pairs of the five BSID-III subscales using new weighted scores

Legenda. CO = Cognitive scale; RC = Receptive communication scale; EC = Expressive communication scale; FM = Fine-motor scale; GM = Gross-motor scale.

subscales with the former tends to be lower; Receptive communication and Fine-motor subscales with the former tends to be higher; Receptive communication and Gross-motor subscales with the former tends to be higher; Expressive communication and Fine-motor subscales with the former subscales tends to be higher; and Expressive communication and Gross-motor subscales with the former tends to be higher. In summary, in the DS sample using the new weighted scores, Expressive and Receptive communication subscales tend to be higher than Cognitive, Fine-motor, and Gross-motor subscales. Table C reported in Supplementary material and Figure 4 show the comparisons of five BSID-III measures between two age groups with DS using new weighted scores. There was a significant difference between the two age groups of children with DS in the Expressive communication subscale where scores of children with DS aged 12-24 months were higher than those of older children. In contrast, the statistically significant difference by age in the Fine-motor subscale highlights lower performance at 12-24 months than in the 25-36 months range There is also a small effect of age on the Cognitive subscale, with the score of toddlers aged 12–24 months slightly higher than toddlers aged 25–36 months. No age effects for the Gross-motor subscale were observed.

DISCUSSION

Down syndrome is characterized by an atypical pattern of development of processes involving cognitive, language, motor, self-care, and personal-social domains (Ferreira-Vasquez & Lamonica 2015; de Lima Freire et al., 2014; Karmiloff–Smith et al., 2016). The aim of this study is to investigate the cognitive, linguistic, and motor development of toddlers with DS assessed by BSID–III in order to give a contribution to the study of the developmental features of these toddlers in their early years of life.

Although the use of BSID–III scales presents some limitations with samples with developmental delays (Anderson & Burnett, 2017; Ferri et al., 2005), they are well suited to outlining a profile characterized by strengths and

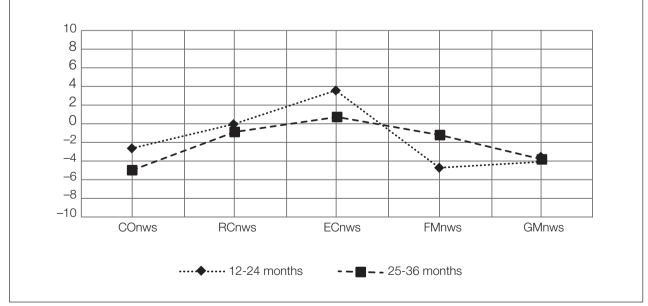


Figure 4 – BSID–III profiles of two age groups using new weighted scores

Legenda. CO = Cognitive scale; RC = Receptive communication; EC = Expressive communication; FM = Fine-motor; GM = Gross-motor.

weaknesses. This is necessary for the customization of early clinical intervention, through direct monitoring of the toddlers' behavior, and the involvement of parents who are important sources of additional information. The present study suggests the use of an alternative psychometric method of converting raw scores into weighted ones, in order to overcome the limitation of the floor effect that occurs when we use the Italian version of the BSID–III scales with samples with intellectual disabilities.

Comparing toddlers with DS to the TD group, using the traditional BSID-III conversion method, the results showed that the toddlers with DS achieve a significantly lower profile in each developmental domain than the TD group. Observation of the profile of the group with DS highlights a higher expressive communication score compared to the scores in the BSID-III other subscales. This result could be explained if we consider the nature of the items present in the Expressive communication subscale (EC). These items assess expressive general communication, which include not only verbal skills but also pre-verbal and gestural skills, defined as a strength in toddlers with DS (Abbeduto et al., 2007; te Kaatvan den Os et al., 2017). Although, as the administration of BSID-III items proceeds, those items assess increasing complex skills, following the acquisition time of typical development toddlers. So, the first EC items assess gestural, non-verbal, and pre-verbal communication skills. Then, they gradually tend to focus on increasingly complex verbal language skills. This could explain the lower score obtained by DS toddlers aged 24-36 months than by DS toddlers aged 12–24 months, in agreement with the literature that suggests that the difficulties in language development of children with DS are more evident when the verbal language acquisition phase begins (Abbeduto et al., 2007; Levy & Eilam 2013).

Therefore, with the exception of the scores obtained by the youngest toddlers with DS on the Expressive communication subscale, the results showed a general flattening of the scores that returns a flat profile of development. Thus, the method of converting raw scores to weighted scores proposed by Hessl et al. (2009) and extended by Orsini et al. (2015) was adopted. Results with new weighted scores showed that this method retrieves the individual variability of the scores obtained on the BSID–III scales and outlines a developmental profile characterized not only by deficits but also by potential strengths of toddlers with DS. Therefore, in agreement with literature, which emphasize that receptive communication skills, along with gestural and pre–verbal ones are less impaired than other skills in children with DS (Abbeduto et al., 2007; Bello et al., 2014; Caselli et al., 1998; Eggers & Van Eerdenbrugh, 2018; Ferreira–Vasques & Lamônica, 2015; Galeote et al., 2011; Næss et al., 2011; Patterson et al., 2013; te Kaat-van den Os et al., 2015; te Kaat-van den Os et al., 2017; Witecy & Penke, 2017; Zampini & D'Odorico 2011), the results showed that toddlers with DS obtain higher scores on both the Expressive and Receptive communication subscales than on Cognitive and Motor subscales. Thus, these findings suggest that in the earliest years of the life of toddlers with DS, preverbal and receptive communication skills may be strengths in their development. Since an important positive correlation between use of pre-verbal, gestural communication, and later vocabulary development (Caselli et al., 1998; Roberts, Price & Malkin, 2007; Yoder & Warren, 2004; Yoder, Woynaroski, Fey & Warren, 2014), has been highlighted, these results support the importance of maintaining and reinforcing preverbal communication skills at this developmental stage in toddlers with Down syndrome, which is also in line with what Yoder et al. (2014) revealed on the effectiveness of early communication interventions.

These findings could have important clinical implications because preschool children with DS often access intervention programs, provided by the Italian Health System, only after their second/third year of life, starting with psychomotor therapies. While access to language and communication therapies (typically speech therapy) occurs only after their third/fourth year of age. As already stated by Caselli et al. (1998) and Yoder et al. (2014), the development of intervention programs aimed at reinforcing communication skills before the age of 3 years is crucial, as it is possible to reduce delay in the acquisition of expressive language.

Another interesting result with the use of new weighted scores is that it hasn't shown a significant difference between Fine-motor and Gross-motor scores of toddlers with DS, suggesting a possible general impairment in the motor domain in toddlers with DS in the first three years of life. This seems in contrast with the literature that suggests a more impairment in fine-motor skills than gross-motor ones, however this difference was observed in studies with DS samples older than 36 months (i.e., Ferreira–Vasquez & Lamônica, 2015). A recent review by Needham et al. (2021), highlighted that there are few studies, in the literature, that analyze fine-motor skills in preschool children with DS and there are no studies aimed at analyzing the difference between fine-motor and gross-motor skills in toddlers with DS aged between 12 and 36 months. Fine-motor skills are very important to self-care, academic achievement, and everyday autonomy, so there is a need to deepen understanding of the impairment of fine-motor skills at this early stage of development in children with DS in order to improve early intervention programs (Alesi & Battaglia, 2019; Needham et al., 2021).

While there are no significant differences between the two age groups with DS on the Receptive communication and Gross-motor subscales, a small difference can be observed in the Cognitive subscale, with the score of toddlers with DS aged 25–36 months lower than the score of toddlers with DS aged 12–24 months. These results seem to be in line with the literature that considers mental delay to increase with age (Godfrey & Lee, 2018; Grieco et al., 2015; Karmiloff–Smith et al., 2016; Patterson et al., 2013; Robert & Richmond, 2015; Wester Oxelgren et al., 2019).

A statistically significant difference was observed between the scores of the two age groups with DS on the Expressive communication and Fine-motor subscales. In particular, the Expressive communication scores of the group with DS aged 25-36 months, were significantly lower than the score of toddlers of DS aged 12-24 months, confirming the results obtained with the traditional raw score conversion method. Instead, the group with DS aged 24-36 months had significantly higher scores than toddlers with DS aged 12-24 months on the Fine-motor scores, suggesting a possible decrease in the delay of fine-motor development, between the second and third year of life. This result is consistent with the study by Hauck, Felzer-Kim and Gwizdala (2020) that points out that the delay in fine-motor development would tend to decrease with age, highlighting that the gap between fine-motor development of children with DS and children with TD from 0 to 18 months would tend to narrow with age even though the speed of development is lower in children with DS. Our results, together with Hauck et al. (2020), could suggest that in the first 3 years of life fine-motor skills would increase in relation to age. Further research should therefore

investigate the trajectories of fine-motor development in the first 3 years of life in order to plan rehabilitation interventions that support positive trends in the development of these skills.

In conclusion, this study makes an important contribution to the knowledge of what happens across developmental domains in toddlers with Down syndrome (DS) and highlights that the developmental profile of children with DS is not only characterized by delays or deficits, but also by resources and strengths that are essential for clinicians and researchers to identify appropriate early intervention. We know that, often, standardized scores return a flat profile when assessing the development of individuals with developmental delays. However, the method of Hessl et al. (2009) and Orsini et al. (2015) allows for more accurate research designs and greater understanding of the development of populations with developmental delay or intellectual disabilities, because it allows to capture intra– and inter–individual variability of scores, overcoming the floor effect.

Despite these appreciable results and implications, there are some limits to the present study. First, it is based on cross– sectional research so it necessitates caution in interpreting age related changes, which could be better captured by studies with follow up. Second, the study is limited to assessing the cognitive, linguistic and motor development of DS toddlers, but does not consider emotional and socio–adaptive behavior, so future research could also investigate this development domain to better define the global functioning profile. Third, our assessment of cognitive, language, and motor development is limited to the use of the BSID–III, so future research could compare the results to other development assessment instruments, to further investigate the validity of Hessl et al.'s (2009) and Orsini et al.'s (2015) method to overcome the issues of the floor effect.

Finally, future research could also use this alternative method of converting raw scores to weighted scores to better investigate the effectiveness of treatments, as the scores obtained may be more sensitive to change.

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SUPPLEMENTARY MATERIAL

BSID-III subscaless	Group v (n =	with DS 144)	-	Group TD <i>t</i> -test and group mean comparison (<i>n</i> = 144)					arisons
	М	SD	М	SD	Diff.	t ₍₂₈₆₎	р	Cohen d	95% CI
Cognitive (CO)	1.68	1.21	9.71	2.78	-8.03	31.79	<.001	3.74	[-4.15, -3.38]
Receptive (RC)	1.63	1.18	10.20	2.83	-8.57	33.51	<.001	3.94	[-4.37, -3.57]
Expressive (EC)	3.03	1.59	10.16	2.87	-7.13	26.09	<.001	3.07	[-3.43, -2.75]
Fine-motor (FM)	1.31	.81	9.61	2.92	-8.30	32.86	<.001	3.86	[-4.28, -3.50]
Gross-motor (GM)	1.81	1.52	9.90	3.21	-8.09	27.32	<.001	3.21	[-3.59, -2.88]

Table A - Comparisons on five BSID-III subscales between DS and TD groups using traditional weighted scores

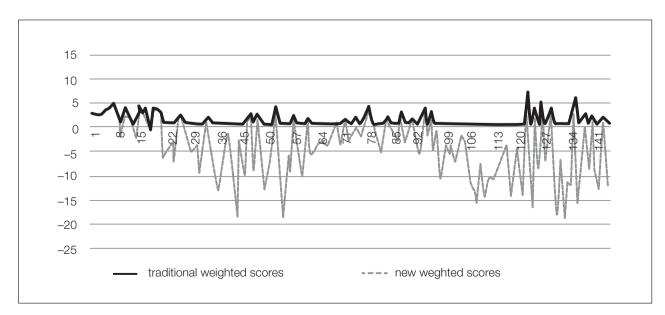
 Table B – Comparison of Five BSID-III measures between two age TD and DS groups using traditional weighted scores

		nonths : 72)	25-36 months (<i>n</i> = 72)		<i>t</i> -test and group mean comparisons				
TD group	М	SD	М	SD	Diff.	<i>t</i> ₍₁₄₂₎	р	Cohen d	95% CI
Cognitive (CO)	9.64	3.19	9.78	2.31	14	.30	.765	.05	[28, .38]
Receptive (RC)	10.64	3.15	9.76	2.43	.88	1.87	.064	.31	[65, .02]
Expressive (EC)	10.25	3.29	10.07	2.40	.18	.38	.707	.06	[39, .27]
Fine-motor (FM)	9.35	2.97	9.88	2.87	53	1.08	.280	.18	[15, .51]
Gross-motor (GM)	10.74	3.28	9.06	2.94	1.68	3.24	<.001	.54	[88,21]
DS group									
Cognitive (CO)	1.79	1.16	1.57	1.25	.22	1.10	.272	.18	[52, .14]
Receptive (RC)	1.58	1.23	1.68	1.12	10	.50	.621	.08	[25, .41]
Expressive (EC)	3.60	1.54	2.46	1.44	1.14	4.59	<.001	.76	[-1.11,43]
Fine-motor (FM)	1.31	.74	1.32	.87	01	.10	.918	.02	[-0.31, .35]
Gross-motor (GM)	1.72	1.24	1.89	1.76	-1.17	.66	.513	.11	[22, .44]

BSID-III subscales	12-24 1	with DS nonths 72)	Group v 25-36 r (<i>n</i> =	nonths	<i>t</i> -test and group mea			iean compa	an comparisons		
	М	SD	М	SD	Diff.	t ₍₁₄₂₎	р	Cohen d	95% CI		
Cognitive (CO)	-2.71	5.79	-4.89	6.44	2.18	2.13	.035	.35	[69, .03]		
Receptive (RC)	.13	2.76	60	3.34	.73	1.43	.155	.24	[57, .09]		
Expressive (EC)	3.55	1.67	.56	4.29	2.99	5.51	<.001	.91	[-1.27,58]		
Fine-motor (FM)	-3.78	5.24	-1.09	2.69	2.69	3.88	<.001	.64	[.31, .99]		
Gross-motor (GM)	-3.77	5.82	-3.74	6.65	.02	.02	.983	.00	[33, .33]		

Table C – Comparison of five BSID-III measures between two age groups with DS using new weighted scores

Figure A – BSID-III Cognitive subscale (CO)





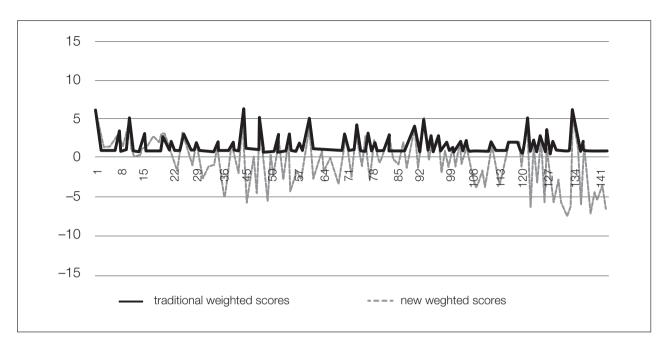
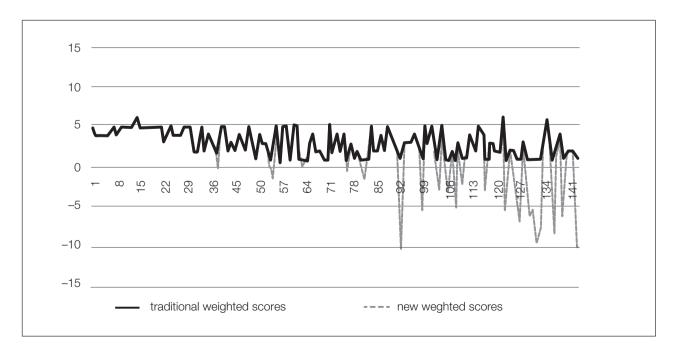


Figure C – BSID-III Expressive communication subscale (EC)



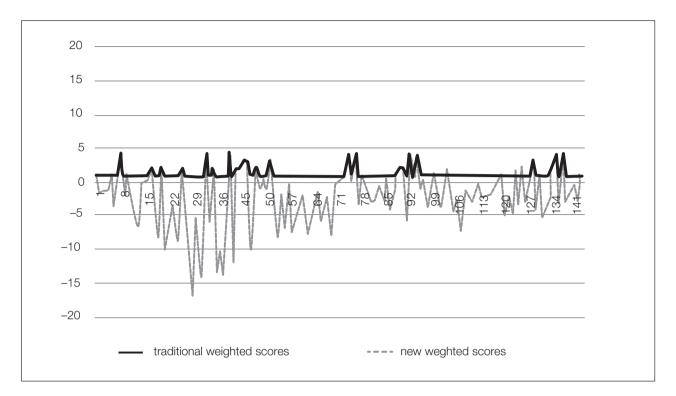


Figure D – BSID-III Fine-motor subscale (FM)

Figure E – BSID-III Gross-motor subscale (GM)

