Speech and Language in 5-year-olds with Different Neurological Disabilities and the Association between Early and Later Consonant Production

Speech/Language in Neurological Disabilities

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ABSTRACT

The primary aim was to describe speech and language abilities in a clinical group of verbal 5-year-old children diagnosed with neurological disability (ND) in infancy, and the secondary aim was to trace precursors to consonant production at age 5 years (T2) in data from 12 to 22 months (T1). The participants (n = 11, with Down syndrome (DS), cerebral palsy, and chromosomal deletion syndromes) were tested with a battery of speech and language tests. Consonant production at T2 was compared to data on consonant use at T1. At T2, two participants had age-appropriate speech and language and another three had age-appropriate speech, but low results on language tests. The remaining six participants had severe speech and language difficulties. Participants with DS had significantly lower results on consonant production measures. An association between consonant production at T1 and T2 for participants with DS indicates that number of different true consonants might be a predictive measure when evaluating young children with DS.

INTRODUCTION

Speech-language pathologists (SLPs) working in early intervention services often meet young children with neurological disabilities (ND). These disabilities affect the child’s early development in one or several areas, such as gross and fine motor, language, speech, and intellectual and social functioning. After initial medical examination, children may end up with diagnoses such as cerebral palsy (CP), Down syndrome (DS) or other genetic syndromes. In many diagnoses associated with ND, speech and language difficulties are very common.

In CP, the prevalence of communication difficulties lies between 50% and 85%, depending on age at assessment as well as on abilities included in the assessment. Communication difficulties in CP may be due to speech disorders and/or a language disorder, but research so far has mostly focused on speech disorder in CP. Gross motor disability (type and severity) and intellectual functioning have strong associations with communicative functioning in CP.

Individuals with DS face particular challenges when it comes to speech and language ability, in addition to the intellectual disability (ID) associated with the diagnosis. Specific difficulties include expressive grammar and speech intelligibility. Children with DS have language deficits that affect both expressive and receptive language, as well as verbal short-term memory. In addition, individuals with DS have speech disorders, which are complex in nature and cannot be fully explained by intellectual level or anatomical differences. Communication in children with DS may also be affected by autism spectrum disorder (ASD), a diagnosis that is over-represented in DS. The severity of the speech disorders common in DS can be exemplified by a study from Cleland and colleagues, who examined speech production in 15 children with DS at 9–18 years of age. Although large variation was found in articulation proficiency, most participants performed below a 3-year-old level. The nature of the speech disorder in individuals with DS has been a matter of discussion by researchers. Rupela and colleagues examined seven children with DS, aged 3–8 years, for speech characteristics and found childhood apraxia of speech, childhood dysarthria as well as unspecified motor speech disorder among the participants.

Apart from DS, there are many other genetic syndromes that cause ND in children. Providing a comprehensive review of these are beyond the scope of this paper. However, in some genetic syndromes the prevalence of speech and language difficulties is close to 100% (for example, in Fragile X syndrome or Angelman syndrome). Other genetic syndromes present with a slightly lower, but still high risk of speech and language difficulties. For example, in 22q11 deletion syndrome 68% have been reported to have speech-language impairment.

For both CP, DS, and other genetic syndromes, knowledge of the expected developmental trajectories is limited. In addition, many studies have been cross-sectional and longitudinal studies are rare and warranted.
**Association between Early and Later Speech/Language Abilities**

We now know that there is a continuity between children’s early vocalizations and their speech and language development. The early consistent use of different consonants has been associated with better expressive vocabulary in children with typical development.\(^{16,17}\) In children with cleft palate, the number of different consonants produced at 18 months has been associated to articulation proficiency at 3 years of age.\(^{18}\) The fact that babbling variables are associated with later speech and language development in typical development as well as in children with non-neurological disabilities, suggests that they might be fruitful to examine also in children with ND.

**Early Consonant Production in Children with Neurological Disabilities**

Today, there are few studies examining early consonant production in ND, but there are some indications that children with ND have smaller early consonant inventories and that consonant inventory might be predictive of later developmental outcomes. For example, in our earlier study of babbling in children with different kinds of NDs, we observed that the participants used significantly fewer different consonants compared to data from typically developing children.\(^{19}\) For participants with DS, the number of different consonants was the only babbling measure that differed from typically developing controls. Investigating children with high risk for ASD, Paul and colleagues found an association between consonant inventory at 6, 9, and 12 months and diagnosis of ASD at age 2.\(^ {20}\) Other studies have found associations between number of different consonants at an early age and later speech, language, or communication outcomes. For children with CP, Pennington and colleagues found the number of different consonants used at age 2 to be predictive of speech and communication outcomes at 5 years of age (together with other predictors such as type of CP and intelligibility).\(^ {21}\)

Examining a group of children with neonatal risk factors (a group that have an increased risk of ND) and typical controls, Jensen and colleagues\(^ {22}\) found that participants who had language difficulties at 6 years of age used a significantly smaller number of different consonants at 6–14 months. If this suggested association between early consonant production and later speech and language outcomes were confirmed also in children with ND, assessment of the number of consonants used would be a useful addition to early SLP assessments in this group.

When studying speech and language abilities in ND, it is common to focus on one medical diagnosis, for example CP or DS.\(^ {1,7,10}\) The main advantage to this approach is the homogeneity of the group. The caseload of an SLP, however, often contains children with a wide variety of diagnoses, and children can present with similar communication difficulties regardless of etiology. In this study, we wanted to explore speech and language abilities longitudinally in a group of children with different neurological disabilities, who shared as a common feature that they received habilitation services early in life.

**Aim**

This longitudinal, prospective study of Swedish children diagnosed with neurological disability in infancy aimed to describe speech and language abilities at age 5 as well as to compare the participants’ consonant production at age 5 to their consonant production at 12–22 months. The following research questions were posed:

1. What speech and language profiles are represented in a group of speaking children with early-onset neurological disabilities?
2. What differences in consonant production are there between groups based on medical diagnosis?
3. Is there an association between consonant production at age 12–22 months and at 5 years?

Based on previous studies of children with ND, communication disorders are expected to be common in this heterogenic group at age 5, but varying from mild to severe difficulty. Furthermore, participants with DS are expected to have more severe difficulties than participants with other diagnoses. Finally, based on longitudinal studies of acquisition in other groups of children, an association between early consonant production and consonant proficiency at age 5 is expected also in this group of children.

**Materials and Methods**

**Participants**

Eighteen children diagnosed with ND in infancy and who had been assessed before 2 years of age (see Nyman and Lohmander\(^ {19}\) for more details) were approached for a follow-up study at age 5. The participants constituted a convenience sample, recruited among young children receiving habilitation services in the Stockholm region. One participant declined to participate in the 5-year-follow up. Data were collected for the remaining 17 participants. For the purpose of the current paper however, only verbal children able to participate in an articulation test were included, resulting in a sample size of 11 children. The excluded 6 participants all used alternative means of communicating, such as manual sign, vocalizations and facial expressions. They also all had severe language comprehension difficulties.

Data from two time points were included in this study: Time point 1 (T1) (participants’ age range 12–22 months, mean 17.4 months) and Time point 2 (T2) (age range 4:11–5:4 years, mean 5:1 years). The T1 data were originally collected by Nyman and Lohmander.\(^ {19}\) In that paper, data on the participants’ canonical babbling, use of plosives and number of different consonants were presented, together with background information. In the current study, we instead used the number of different true consonants from T1 to answer the research question on associations between early and later consonant production. This T1 data have thus not been previously published. T2 data were collected for the current study and have not been previously reported.

At T2, all 11 participants had diagnoses associated with complex motor and/or cognitive/intellectual disabilities. All participants had
at least one parent with Swedish as their first language. For background information on the participants at age 5, see Table 1. Information on medical diagnoses, hearing level, gestational age and neuroimaging findings were retrieved from medical records. Hearing level data were categorized based on hearing level on the best ear according to the World Health Organization. The categories normal hearing (<26 dB HL), mild hearing loss (26–30 dB HL), moderate hearing loss (31–60 dB HL) and severe hearing loss (>61 dB HL) were used. One participant had not undergone hearing screening.

The participants had a mean gestational age of 35 weeks and 3 days (range 22 weeks, 6 days to 40 weeks, 4 days). For one participant, data on gestational age were missing. Thus, a big proportion of the participants were born prematurely; five were born at 36 weeks of gestation or earlier. Neuroimaging findings were available for five of the participants and showed that different forms of brain damage were represented, such as white matter injuries (n = 3) and basal ganglia injuries (n = 1). One participant had normal findings. For participants with early-diagnosed genetic syndromes (n = 6), neuroimaging had not been performed, as per clinical guidelines. None of the participants had been evaluated for ASD or ID at the time of the T2 data collection. Thus, there was no information in the medical records on non-verbal cognitive functioning.

Procedures

Data Collection at Time Point 1

Assessment at T1 was based on audio-video recordings of 35–45 minutes of parent–child interaction, using a standardized procedure and set of toys. A trained research assistant did all assessments based on the audio-video recordings. A babbling observation was performed, where occurrence of different babbling variables was noted using an observation form. The observation form contained a list of all 18 Swedish consonant sounds. The rater marked all consonant sounds that were heard at least twice. Other consonant sounds could be added on the form if heard at least twice. More details on the data collection at T1 can be found in the paper by Nyman and Lohmander.

Data Collection at Time Point 2

At T2, the participants were assessed using a test battery, covering a broad range of speech and language abilities. In addition, spontaneous speech samples were elicited by using pictures or play material with toys of the child’s choice. Prior to testing, parents were interviewed about their child’s abilities and based on this, the appropriate tests to use for each participant were chosen. All testing was performed by the first author at a habilitation clinic and was audio- and video recorded using a Sony HDR-CX250E video camera and a Sony ECM-MS957 or ECM-MS907 external microphone. For one participant with significant motor disability (participant CP1), the testing was adapted so that the motor demands were minimized. For example, a response by pointing to pictures was replaced by saying the number of the chosen picture.

Receptive Language

Receptive language was assessed using the Swedish version of Test for Reception of Grammar-2 (TROG-2) or the Swedish translation of Reynell Developmental Language Scales-III (RDLS) depending on the child’s ability to participate in formal testing. TROG-2 measures receptive grammar. Sentences of increased grammatical complexity are read to the participant while four contrasting pictures are shown. The task for the child is to indicate which picture matches the sentence. Results are presented in number of correct blocks, where each block represents a grammatical structure. The results are presented as raw scores (number of correctly solved blocks out of a maximum of 20).

RDLS contains both an expressive and receptive subtest, but only the receptive subtest was used. The receptive subtest includes tasks such as pointing to objects, acting out the test leader’s instructions using the toys and pointing to the picture that matches the sentence uttered by the test leader. The results are presented as raw scores.

Expressive Language

Expressive language was assessed with different methods described below. The five longest utterances for each child were identified based on all spontaneous communication during the one-to-two-hour long assessment. First, sections of the video recordings containing long utterances were identified for each child. These sections were then analyzed by two experienced SLPs and the five longest utterances were transcribed orthographically. All spoken words and words communicated by augmentative and alternative communication (manual sign)
were included. One participant (CP1), who communicated with spoken words and a communication book with symbols was excluded from this analysis. Mean maximum utterance length was calculated by taking the five longest utterances, adding up the number of words and dividing it by five. Both transcribers analyzed all video recordings separately, to enable reliability analysis. When possible, the participants were also tested with the subtests Expressive Vocabulary and Sentence Recall from the Clinical Evaluation of Language Fundamentals-4 (CELF-4). Expressive Vocabulary is a picture-naming test, where the answers are scored as correct or incorrect. In Sentence Recall, the task is to repeat the sentence produced by the test leader. The subtest consists of 24 sentences of increasing length. The child’s production is scored from 0 to 3, where 0 = correct production, 1 = one error, 2 = two to three errors and 3 = four or more errors. Results on both CELF-4 subtests are presented as scaled scores around a normative M = 10 and SD = 3, based on Swedish norms. For participants who could not be tested with the Expressive Vocabulary subtest, parental checklists were used. Based on the interview with the parent, either Swedish Communicative Development Inventory III (SCDI-III) or the Swedish Communicative Development – words and gestures (SECDI w&g) were used. In SCDI-III, the parent marks the words the child produces from a checklist of 100 words. Results were compared to Swedish age norms for four-year-olds. In SECDI w&g, the parent marks the words that the child understands or understands and produces from a checklist of 370 words. Only words that were both understood and produced were used. Results are presented as raw scores.

Consonant Production
Consonant production was assessed using the Swedish articulation and nasality test (SVANTE). The minimal standard set was used, including 30 words representing 10 of the 18 different Swedish consonants. Six plosives (/p/,/b/,/t/,/d/,/k/,/g/), three fricatives (/f/,/v/,/s/) and one nasal consonant (/n/) were included. All consonants were assessed in initial position and in three different words (except for /v/ and /s/ in two words. /s/ was also assessed in final position in two words). Phonic transcription of the elicited words of the SVANTE was made using semi-narrow transcription. Two experienced SLPs transcribed the whole material independently for determination of inter-transcriber reliability. They also re-transcribed the material of three randomly selected participants (corresponding to 27% of the total number of participants) for calculation of intra-transcriber reliability.

Presence of Motor Speech Disorder
Presence of motor speech disorder was assessed based on the audio- and video recorded articulation test. One experienced SLP (the first author) listened to all recordings and assessed whether the participants’ speech showed signs of motor speech disorder or not. Signs of motor speech disorder included imprecise articulation, frequent vowel substitutions or distortions, short breath groups and harsh or breathy voice quality. Age appropriate distortions were not considered signs of motor speech disorders, and neither were substitutions in which the target sound was replaced with another, distinctly articulated, sound.

Functional Intelligibility
Functional Intelligibility in everyday contexts was rated by parents using the Swedish translation of the Intelligibility in Context Scale (ICS). The ICS consists of seven items where the parent rates how well the child is understood by the parent, immediate family, extended family, the child’s friends, acquaintances, the child’s teachers and strangers. Ratings are made on a 5-point scale (5 = always, 4 = usually, 3 = sometimes, 2 = rarely, 1 = never). Based on all seven items, a mean can be calculated. The mean can also be expressed in words, for example a mean of 3.5 can be interpreted as the child being understood by others sometimes to usually.

Ethical Considerations
The study was approved by the Regional ethics board in Stockholm (dnr 2017/206-31 and 2013/1989-32). Parents to all participants gave written consent to participating in the study and were informed that they at any point could withdraw this consent without any consequences for them or their child.

Analysis
Data from the babbling forms at T1 were scrutinized and analyzed with regards to the number of different true consonant sounds used by the participants. Thus, all glottals and glides were excluded. Furthermore, as the distinction voiced-voiceless cannot be expected to be established in young children, consonants were counted as the same if they had the same articulatory place and manner. For example, /p/ and /b/ were counted as the same consonant sound.

All tests from T2 were scored according to the manuals, using Swedish age norms. Age appropriate language was defined as results above -1.5 SD on TROG-2 and the subtests from CELF-4.

Two measures were drawn from the SVANTE transcriptions: the percentage of consonants correct (PCC) and the number of established consonants. PCC was calculated across the 30 occurrences of the 10 target consonants in the minimal standard set of the SVANTE. Each target consonant occurred 2–5 times. All instances of an observed consonant symbol not matching the target consonant symbol were scored as incorrect. Errors expressed in diacritics such as weak articulation or de-voicing were however scored as correct. All words attempted were included in the PCC calculations. Age appropriate consonant production was defined as a PCC at 88% or higher, corresponding to a result at or above −1.5 SD for the age group (as by the SVANTE norm data).

As the material in SVANTE mini allowed for analyzing also the presence of /l/ and more occurrences of /t/ and /v/, these were added when calculating the number of different consonants. Thus, the analysis of the number of established consonants was based on 39 occurrences of 11 target consonants in the 30 elicited words in SVANTE mini. As defined in the SVANTE manual, a consonant was considered established if it was produced in at least two of the elicited words. To be
counted, the consonant had to be transcribed using the correct IPA symbol, however diacritics were ignored. For /s/ a slightly different approaches was taken. As /s/-distortions are common in typically developing children at this age, interdental, lateral, and palatal realizations were accepted.\textsuperscript{17} The total number of consonants included was \(11:/p/, /b/, /t/, /d/, /k/, /g/, /n/, /f/, /v/, /s/, \) and /l/.

Reliability

Number of Different True Consonants at 12–22 Months

Nyman and Lohmander\textsuperscript{19} reported a fair agreement between two observers for the measure number of different consonants at T1. To further strengthen the reliability of the similar measure number of different true consonants, five randomly selected participants were re-assessed by the first author. The consonants identified by the main observer were compared to the consonants identified in the re-assessment by the first author. Across the five participants, the main observer identified 25 consonants, 16 of which were also identified in the re-assessment. When considering also similar, but not identical consonant sounds (/β/-/v/, /g/-/γ/) the agreement was 19/25 consonant sounds, or 76%.

Consonant Production Measures

Intra-class correlation coefficients (ICC) were calculated to assess intra- and inter-transcriber reliability of the consonant production measures. For PCC, inter- and intra-transcriber reliability were based on the PCC values calculated for each transcriber. For the number of established consonants, only inter-transcriber reliability was calculated, based on the number of established consonants obtained from the transcriptions of each transcriber. For PCC, inter-transcriber reliability was excellent: ICC (2,1; single measures, absolute agreement) = 0.904, \(p < .001\), as was intra-transcriber reliability: ICC (3,1; single measures, absolute agreement) = 0.987, \(p = .001\) and ICC (3,1; single measures, absolute agreement) = 0.991, \(p = .004\), respectively. For the number of established consonants the inter-transcriber reliability was excellent: ICC (3,1; single measures absolute agreement) = 0.977, \(p < .001\).

Mean Maximum Utterance Length

To assess inter-observer reliability for maximum utterance length, ICC was used. The calculations were based on the mean maximum utterance length for each participant, comparing the two transcribers. Inter-transcriber reliability was excellent: ICC (2,1; single measures, absolute agreement) = 0.972, \(p < .001\).

Statistical Analysis

Statistical analyses were performed using IBM SPSS Statistics, version 25 and 27. Data were analyzed using descriptive statistics (mean/median, standard deviation, and range). Differences between groups based on medical diagnoses were analyzed using the Mann–Whitney U-test. Correlations between consonant production at T1 and consonant production at T2 were analyzed using Spearman rank correlation.

Results

Descriptive statistics for the speech and language variables can be found in Table 2 and in the first column of Table 4. In Table 3, individual results for the participants are summarized. In summary, two participants (CP4 and CD2) had age appropriate results for all speech and language measures. One participant (CP3) had age appropriate results on all measures except on sentence recall. Two participants (CD2 and CP2) had age appropriate speech, but language difficulties. The remaining participants (DS1, DS2, DS3, DS4, DS5, CP1) had severe difficulties with both speech and language. The parent-reported mean score results for intelligibility varied from 2.9 to 5. Three participants showed signs of motor speech disorder; all three had difficulties with both speech and language.

In Table 4, descriptive statistics are presented for the speech and language measures in groups based on medical diagnosis (DS, CP, and chromosomal deletion). The DS group had significantly lower results than the CP group, both regarding PCC (\(U = 0, p = .016\)) and number of established consonants (\(U = 1.5, p = .032\)). Due to the small group size, the group with chromosomal deletions was not included in this comparison.

In the figures, the number of different true consonants at 12–22 months over PCC at 5 years is plotted. Figure 1A shows the results for the whole group of 11 participants. Figure 1B shows the results for the subgroup of participants with DS. A Spearman rank correlation showed a moderate correlation between number of different true consonants at T1 and PCC at T2, however, the association did not reach significance (\(r_s = 0.553, p = .077\)). When studying the subgroups based on medical diagnosis separately, there was, however, a significant correlation for the DS group between number of different true consonants at T1 and PCC at T2 (\(r_s = 0.894, p = .041\)).

Discussion

The main finding from this follow-up study of children with early diagnosed ND whose early consonant production has been described in detail,\textsuperscript{19} was that only two of the 11 participants from the first assessment (before 2 years of age) reached age appropriate result on both speech and language measures at age 5 years. Furthermore, participants with DS had more severe difficulties with consonant production compared to participants with other diagnoses and there was a

| Table 2. Proportion of participants with age appropriate results on speech and language outcomes for 11 children with ND at age 5. |
|-----------------|-----------------|-----------------|
| Speech/language measure | n               | % age appropriate* |
| Language comprehension | 11              | 27% (n = 3)        |
| Expressive vocabulary   | 11              | 27% (n = 3)        |
| Sentence recall          | 4               | 50% (n = 2)        |
| PCC                      | 11              | 45% (n = 5)        |

*Age appropriate = ≥ 1.5 SD on each test according to the norms.
significant association between early and later consonant production in children with DS. Children with DS who used a high number of different true consonants at T1 also had higher PCC score at T2.

**Speech and Language Profiles**

The proportion of participants with age appropriate speech and language was 2/11, but given that none of the six excluded participants had age appropriate speech or language abilities, the true proportion of age appropriate abilities of the whole sample is actually even smaller, 2/17. This confirms what is already known in children with neurological disabilities: that speech and language disorder is common and should not be ignored. Speech disorders (as measured using number of established consonants and PCC based on single words) were present in six of the speaking participants and PCC values were very low compared to the SVANTE age norms. The speech disorder also seems to affect intelligibility. Although parent-reported functional intelligibility varied, many of the parents to participants with speech disorder reported that their child “sometimes” or “rarely” was understood by others (range for the participants with speech disorder 2.9–3.6 as compared to 3.6–5.0 for participants with age appropriate speech). Although the impact on intelligibility by speech disorder cannot be distinguished from the impact by language difficulties in this sample, it is likely that speech disorders do affect everyday functioning and social participation in these children.

When it comes to language measures, all participants with a speech disorder also had language difficulties, both receptive and expressive. There was quite small variability in the language test results; with the exception of one participant (CP3), participants either had non-age appropriate results on all language measures, or age appropriate results on all language measures for the participants on speech and language measures used at age 5. Bold figures represent non-age appropriate results (for measures were age-related norms are available).

| Language comprehension: test used (score) | Expressive vocabulary: test used (score) | Sentence recall, scaled score | Mean maximum utterance length | PCC | Number of established consonants | Established consonants | Motor speech problems | Functional intelligibility |
|------------------------------------------|------------------------------------------|-------------------------------|-----------------------------|-----|---------------------------------|------------------------|-------------------------|--------------------------|
| ID                                       | RDLS (6)                                  | SECDI w&g (79)               | 2.6                         | 25% | 1                               | /b/                    | Yes, motor speech involvement | 3.1                     |
| DS2                                      | TROG (0)                                  | SCDI-III (<5th percentile)   | 2.0                         | 33% | 2                               | /b, d/                 | No                      | 3.0                     |
| DS3                                      | TROG (0)                                  | SCDI-III (<5th percentile)   | 2.8                         | 29% | 4                               | /t, n, v, l/           | No                      | 2.6                     |
| DS4                                      | RDL (15)                                 | SECDI w&g (235/370)          | 2.6                         | 42% | 6                               | /p, b, t, d, n, s/     | No                      | 3.6                     |
| DS5                                      | TROG (0)                                  | -                             | 2.0                         | 56% | 5                               | /p, b, n, l, s/        | Yes, motor speech involvement | 2.9                     |
| CP1                                      | TROG (0)                                  | SECDI w&g (193/370)          | -                           | 57% | 5                               | /p, d, g, n, s/        | Yes, severe dysarthria   | 3.1                     |
| CP2                                      | TROG (0)                                  | CELF (1)                      | 1                           | 96% | 11                              | /p, b, t, d, k, g, n, f, v, s, l/ | No                      | 5.0                     |
| CP3                                      | TROG (11)                                 | CELF (10)                     | 4                           | 10.8 | 93% | 11                              | /p, b, t, d, k, g, n, f, v, s, l/ | No                      | 3.6                     |
| CP4                                      | TROG (13)                                 | CELF (9)                      | 10                          | 13.4 | 93% | 11                              | /p, b, t, d, k, g, n, f, v, s, l/ | No                      | 5.0                     |
| CD1                                      | TROG (0)                                  | CELF (2)                      | 4.8                         | 100% | 11                              | /p, b, t, d, k, g, n, f, v, s, l/ | No                      | 3.4                     |
| CD2                                      | TROG (5)                                  | CELF (9)                      | 7                           | 10.4 | 100% | 11                              | /p, b, t, d, k, g, n, f, v, s, l/ | No                      | 4.3                     |

*Results for the RDLS in raw scores. All participants tested with RDLS performed under the mean for Swedish two-year-olds.

*Results for the TROG as number of correct blocks. Age appropriate results on the TROG-2 was defined as a result at or below –1.5SD, according to Swedish norms.

*Results for the SECDI w&g in raw scores. All participants tested with the SECDI w&g performed below the median for 28-months-old children (270 out of a maximum 370).

*Results from SCDI-III in percentiles, compared to norm values for Swedish 4-year-olds.

*Results for the CELF in raw scores (mean = 10, SD = 3). Age adequate results are defined as results above –1.5 SD.

*PCC based on single words, as measured by the SVANTE.

*As represented by mean score on the ICS.

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Table 4. Descriptives of speech and language measures for all participants and for groups based on medical diagnosis (Down syndrome, Cerebral palsy, Chromosomal deletion). Means (standard deviations) are provided.

| Variable                                      | All participants (n = 11) | Down syndrome (n = 5) | Cerebral palsy (n = 4) | Chromosomal deletion (n = 2) |
|------------------------------------------------|---------------------------|-----------------------|------------------------|-----------------------------|
| Mean maximum utterance length                 | 5.80 (4.27)               | 2.4 (0.37)            | 10.27 (3.43)           | 7.60 (3.96)                 |
| PCC                                           | 65.82 (30.93)             | 37.0 (12.35)          | 84.75 (18.55)          | 100 (0)                     |
| Number of established consonants              | 7.1 (4.0)                 | 3.6 (2.1)             | 9.5 (3.0)              | 11 (0)                      |
| Functional intelligibility                     | 3.6 (0.8)                 | 3.0 (0.4)             | 4.2 (1.0)              | 3.9 (0.6)                   |

*Based on the SVANTE.

*Based on the ICS.
measures. Notably, language difficulties were observed also in participants who had no or only mild articulation difficulties (participants CD1 and CP2). Furthermore, one participant (CP3) had difficulties with repetition of sentences only. Sentence repetition has been suggested as a measure of underlying language ability and difficulties with demanding repetition tests are common in developmental language disorder (DLD). The presence of a neurological disability per definition excludes children from the diagnosis of DLD and instead the term “language disorder associated with ...” is

Figure 1. A) The association between Number of different true consonants at T1 and percentage of consonants correct at T2 for the whole group of 11 participants. B) The association of Number of different true consonants at T1 and Percentage consonants correct at T2 for the participants with Down syndrome.
recommended. The rationale for this is that certain neurobiological conditions are likely to affect which intervention is appropriate. It is important to remember, however, that children with ND may have language disorder with characteristics very similar to those of DLD. From a clinical perspective, this highlights the importance of assessing language skills, including language comprehension, in children with ND, even if they do not have a speech disorder. If this is not performed, difficulties with language comprehension and/or difficulties with verbal short-term memory might go unnoticed, causing difficulties in everyday life and with later school performance.

**Speech and Language Profiles by Medical Diagnoses**

The participants with DS scored low on articulation measures in the current study. In fact, all participants with DS scored lower than all other participants when it comes to PCC. Given the ID associated with DS, this is not surprising. The results for the subgroup of participants with DS is in line with previous research, which has shown that many children with DS present with severe speech and language disorder. A closer inspection of the words produced by the DS participants showed that they often deleted whole syllables, as well as final consonants. Consonant deletion is also described by Cleland and colleagues in their group of older children and adolescents with DS. Signs of motor speech disorders were present in two participants with DS. They both exhibited imprecise consonants and one of them also used frequent vowel substitutions. The tests used in the present study did not permit a further analysis of speech motor control or a differential diagnosis of, for example, phonological difficulties, childhood apraxia of speech, or childhood dysarthria.

There was considerable variation in speech and language performance among the children with CP in the present study, but three out of the four participants had speech or language difficulties. Interestingly, this is similar to the prevalence reported by Hustad and colleagues for 4-year-olds with CP (26 out of 34 participants, 76%). As in the present study, both speech and language disorders were reported. Previous studies investigating only prevalence of speech disorder, have reported lower prevalence figures (see for example Nordberg and colleagues). In the present study, one participant had severe dysarthria whereas the other three showed no clear signs of motor speech disorder. Two of the other participants, however, had language difficulties. The fact that speech and language disorders do not always co-exist in the current sample, underlines the importance of including both speech and language abilities in SLP assessments of children with CP.

The two participants with chromosomal deletion both had age appropriate speech, but one of them had language difficulties. Due to the small number of participants in this subgroup, no conclusions can be drawn from this result.

**Associations between Early and Later Consonant Use**

When studying early vocalizations in children at risk of speech and language difficulties, the presence of canonical babbling (i.e., babbling that consists of a consonant and a vowel, with a rapid transition between them) is a commonly used variable. Indeed, canonical babbling is a more commonly used measure than consonant inventory in research in ND and does seem to be affected in diagnoses such as ASD, Fragile X Syndrome, Rett’s syndrome, and CD. The presence of canonical babbling was assessed at T1 in the participants included in the current study, but the wide age span at T1 did not allow for valid comparisons to the data from 5 years of age. Another reason for using the number of different true consonants as a predictor is that although presence of canonical babbling seems to be necessary for the development of speech in children with ND, it is not enough. In other words, canonical babbling might lack in sensitivity as a predictor of later speech and language skills for children with ND. Possibly, more detailed measures of the quality of vocalizations, such as the number of consonants used, might prove a more specific measure.

There was a significant association between the number of different true consonants at T1 and consonant production at T2 for participants with DS in this study. Children with DS who used a high number of different true consonants at T1 also had higher PCC score at T2. Although the association between early consonant use and later articulation proficiency has not previously been found in DS, the idea that children with DS differ in their speech and language development compared to children with other NDs is not new. Canonical babbling, for example, seems to be surprisingly unaffected in DS, at least considering the speech and language difficulties that children present with later in life. Compared to children with other NDs however, the subsequent development of vocalizations and language in toddlerhood and early preschool years does seem to continue at a slower rate in DS. The mechanisms behind the particular speech and language difficulty in DS are not fully understood, but difficulties in both oral motor control and auditory short-term memory are present in DS. The results regarding the association between early and later consonant use in DS in the current study are preliminary and need to be confirmed in larger cohorts. If confirmed, consonant measures might be useful when assessing toddlers with DS.

**Limitations**

The small number of participants is one factor that limits the generalizability of the findings of this explorative study. A larger group of participants would have been desirable, and the results therefore need to be interpreted with caution. Nevertheless, as there are only few published studies in the field, we believe that the results contribute to the knowledge on speech and language development in children with ND, especially since the design is longitudinal and the participants were recruited before age 2 years. Hearing level and intellectual level are two important factors that affect children’s speech and language development, especially in ND. In this study, it was not possible to assess hearing level or intellectual level in parallel to the speech and language assessment, and instead, we had to rely on previously performed clinical evaluations, retrieved from medical records. Thus, hearing levels were not systematically assessed for all participants, and for some participants, assessments had not been done since infancy. It is therefore hard to estimate the influence of hearing loss on the speech and language results in the participating children,
especially in the participants with DS, whose speech and language has been found to be affected by degree of hearing loss. As for intellectual level, none of the participants had undergone a psychological assessment, according to the medical records. This is accordance with clinical guidelines in Sweden, recommending that such an evaluation should be performed before 6–7 years of age, the age when Swedish children start school. Consequently, we did not have access to information regarding level of intellectual functioning apart from the fact that the participants with DS all had some degree of intellectual disability – as a part of their syndrome.

Conclusions

In a clinical representative sample of 11 speaking children early identified with ND, only a minority had age appropriate speech and language abilities and severe speech and language difficulties were common across ND diagnoses. However, participants with DS performed lower than participants with other types of ND on two consonant production measures: PCC and the number of established consonants. The results point to the need to properly evaluate speech and language in children with ND. As the number of different true consonants at 12–22 months was associated to articulation at 5 years of age for participants with DS, the number of different consonants used might be a promising measure when evaluating speech and language in children with DS during the second year in life.

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Declaration of interests

The authors report no conflicts of interest.

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