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A Standardized Protocol for Maximum Repetition Rate Assessment in Children

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Keywords

Maximum repetition rate · Assessment · Motor speech · Speech

Abstract

Background/Aims: Maximum repetition rate (MRR) is often used in the assessment of speech motor performance in older children and adults. The present study aimed to evaluate a standardized protocol for MRR assessment in young children in Dutch. **Methods:** The sample included 1,524 children of 2–7 years old with no hearing difficulties and Dutch spoken in their nursery or primary school and was representative for children in the Netherlands. The MRR protocol featured mono-, tri-, and bisyllabic sequences and was computer-implemented to maximize standardization. **Results:** Less than 50% of the 2-year-olds could produce >1 monosyllabic sequence correctly. Children who could not correctly produce ≥2 monosyllabic sequences could not produce any of the multisyllabic sequences. The effect of instruction (“faster” and “as fast as possible”) was small, and multiple attempts yielded a faster MRR in only 20% of the cases. MRRs did not show clinically relevant differences when calculated over different numbers of repeated syllables. **Conclusions:** The MRR protocol is suitable for children of 3 years and older. If children cannot produce at least 2 of the monosyllabic sequenc-

es, the multisyllabic tasks should be omitted. Furthermore, all fast attempts of each sequence should be analyzed to determine the fastest MRR.

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Introduction

Diagnosing a child with motor speech disorders (MSD; e.g., childhood apraxia of speech [CAS], developmental dysarthria) is not always an easy task for a speech-language pathologist (SLP). Children with MSD form a heterogeneous group not only due to etiological factors but also due to individual differences in the pattern of development of the speech disorder leading to individual differences in speech errors [1]. Because of these differences, it is difficult to assess the underlying deficits (issues in speech motor planning, programming, or execution) in children with MSD. In clinical practice, underlying deficits are often examined separately, but multiple factors may be involved [2]. This is why the precise identification of MSD in many children is difficult, especially for those who do not have an evident primary problem [3]. Nevertheless, the precise identification of MSD is important for a correct diagnosis and treatment planning. If the intervention is better adapted to the individual diagnosis, this will ensure better prog-

ress [3], which can lead to great benefits for further speech and language development of the child.

A misdiagnosis or under-diagnosis can occur because SLPs simply rely on a diagnostic checklist for identifying MSD. These lists are often not specific enough to distinguish between the possible different underlying deficits. At the moment, there is no specific test protocol for diagnosing children with MSD [4]. Recently, Shriberg et al. [5], proposed to use a Pause marker to identify children with CAS. This could be helpful, but there is a need for a gold standard for all children with MSD. Furthermore, there are only a few objective instruments for mapping children's motor speech skills and there is no norm referenced assessment based on a large data set. This is a major problem, as a good understanding of normal speech is necessary for the interpretation of MSDs [6]. Finally, the existing assessments may be hard for children to complete and it also demands a lot of the SLP's judgment ability. The outcomes therefore may not represent the children's true abilities [7].

However, an often-used objective assessment for the clinical judgment of the motor speech performance of older children and adults is the maximum repetition rate (MRR) [8–11]. The MRR frequently contains 2 types of stimuli: repetition of monosyllables (papapa) and of multiple syllable sequences (pataka) [9]. MRR is also called diadochokinetic (DDK), and both terms are used in the literature. We choose the term MRR instead of DDK.

There is much debate about using meaningful (e.g., "patticake" or "pat-a-cake") or nonmeaningful stimuli (e.g., "pataka"); however, Williams and Stackhouse [12] concluded that it is desirable to use nonmeaningful stimuli to assure that the children's performance is not influenced by their linguistic abilities. Furthermore, the MRR contains often the consonants /p/, /t/, and /k/ in a sequence [13] such that the 3 major articular organs are examined, namely, the lips, the jaw, and the tongue [14]. Thus, the different consonants represent multiple levels of physiological complexity since each consonant has a different place of articulation and age of acquisition. These consonants cannot be produced in isolation in succession, which is why the consonants are combined with a vowel [13]. Thus, the syllables /pa/, /ta/, and /ka/ were used in several studies [3, 8, 14]. MRR protocols typically consist of multiple components, which increase in complexity. First, the child should repeat the monosyllabic sequences /papa./, /tata./, and /kaka./ . Second, bisyllabic sequences such as /pata./ and /taka./ are administered. The MRR ends with the repetition of the trisyllabic sequences /pataka./ [14]. During the assessment of MRR, children are asked to re-

peat the different sequences in one breath at the highest possible pace. The sequences are meant to be repeated without errors and interruptions [14]. Many children struggle with the unnatural situation of the MRR, which requires a specific approach with regard to instruction and practice opportunities [15]. The MRR appears to be difficult for younger children, who make relatively more articulation errors during MRR tasks as compared to conversational speech [16]. Williams and Stackhouse [12] found in their study of 30 typically developing children aged 3–5 that the MRR was more sensitive when the score was based on accuracy and consistency instead of rate of the productions. However, Yaruss and Logan [16] found no age-related increase in such MRR accuracy and consistency scores. Overall, young children show much more variability in their performance than older children, such that the timing, speed, and fluency of speech movements become less variable when children get older [17]. Several studies showed that children make fewer mistakes during the performance of monosyllabic sequences compared to the multisyllabic sequences [18], and the rate of the sequences decreases as the task becomes more complex [19, 20].

Measuring MRR

The MRR used to be measured perceptually without any support of instrumental methods that can visualize the acoustic waveform. However, Gadesmann and Miller [21] noted that the use of only perceptual evaluation is not acceptable for clinical diagnosis because perceptual measurement is not accurate enough. Nowadays, there are several programs that semi-automatically interpret the various MRR results. Some examples of these types of programs are the DDK Rate Analysis, which is part of the Motor Speech Profile Model [22], TOCS+ MPT Recorder™ [23], and Praat [24]. Although in these programs, the task of the examiner is reduced to simply counting syllables, difficulties still occur when the speaker repeats the syllables quickly and irregularly. In this case, the individual syllables are too close together, which makes it difficult to detect the syllable boundaries which will influence the reliability of the value.

There is no uniform method of measuring the MRR, which makes it difficult to compare the results of different children worldwide. There are 3 methods being used: (1) counting syllables repeated in a certain amount of time (count by time), (2) measuring the time needed to repeat a given number of syllables (time by count), or (3) assess how many syllables can be produced in one breath [21]. As a consequence, there is large variability with respect to the collected norm data, which in its turn leads to difficulties with the interpretation of the MRR results [7, 12].

Clinical Use of the MRR

The MRR performance of children with MSD differs compared to typically developing children. Authors [14, 15, 25, 26] of 4 separate studies concluded that children with MSD (spastic dysarthria and CAS) differ in their performance on the MRR. More recently, Murray et al. [27] advised to use an oral motor assessment to diagnose CAS, which includes the trisyllabic sequence, /pataka/, and polysyllabic word accuracy to diagnose CAS. The mentioned authors concluded that the MRR is a valuable tool in the differential diagnosis of underlying speech motor deficits, which is supported by the differences in MRR performances between children diagnosed with dysarthria [15] and apraxia of speech [14] as compared to controls. Others dispute this because they did not find such differences between children with a typical development and children with MSD [28, 29]. Our opinion is that, although MRR does not necessarily reflect the primary speech disorder in all cases with SSD, MRR can play a role in the differential diagnosis to assess disorders in underlying articulomotor planning and programming [3, 30]. Interpreting only performance on the MRR task is insufficient to assess the underlying speech problem; this requires multiple tasks and the assessment of a comprehensive speech profile [30]. Thus, in a large validation study, we assessed performances on the MRR as well as other speech tasks (picture naming [PN], nonword imitation [NWI], word and nonword repetition [WR, NWR]) with the recently developed diagnostic instrument, Computer Articulation Instrument (CAI) [31] – in a group of 1,524 typically developing children. Factor analyses on the task performances showed separate factors for each of the 4 tasks [32]. The diagnostic value of these norm data resides in now being able to compare MRR performance of children with MSD to typical development.

As discussed above, SLPs use the MRR as an assessment tool for children with MSD. To date, available norm data are based on small samples of children, especially in the younger age groups (2–5 years old), and there is still much debate on the manner to conduct the MRR and the method to calculate the MRR. The aim of this study was to optimize a standardized protocol – which was based on previous studies in the Dutch language [14, 15, 25]. Other research questions of this study were: are children aged 2–7 years able to perform the MRR task, and what kind of instruction and how many attempts do children need to produce their fastest sequence; during clinical work, we noticed some children were slower after the instruction to go as fast as possible.

Materials and Method

Participants

This study was part of a large normative study of a new Dutch instrument, the CAI, to assess children's speech problems (for more details see Maassen et al. [31]). A total of 1524 Dutch-speaking children in the Netherlands were recruited between January 2008 and April 2015. The children were recruited via nurseries (47) and mainstream primary schools (71) in the Netherlands. The sample was representative for gender, urbanization, and geographic region. Inclusion criteria were as follows: (1) no hearing difficulties and (2) the Dutch language was spoken in the nursery or primary school. Table 1 shows the number of subjects per MRR sequence per age group (14 age groups were selected) and gender of the children. Not all children executed all MRR tasks. Furthermore, in some cases, the audio files were damaged due to technical problems or the individual syllables were not recognizable because of background noise and the recordings were excluded from the sample.

The children were randomly selected from a list based on age group and gender. All parents/caregivers of the randomly selected children were asked for permission via an informed consent letter to include their child anonymously in this large study. If parents gave permission, they filled out a short questionnaire containing questions about the speech and language development, multilingualism, and health condition (e.g., loss of hearing) of the child. The protocol has been assessed by an ethics committee (Radboud University Nijmegen Medical Centre) and the study was carried out.

Data Collection

The children were seen individually by 2 SLP students or 1 SLP. In total, 14 SLPs assessed the younger children (2–4 years of age) and 110 SLP students administered the CAI for the older children (4–7 years of age). All these research assistants were trained to assess children with the CAI by the first 2 authors, and a precise instruction in the form of a guideline was given. To assure a flawless administration of the CAI, students worked in pairs.

An assessment session with a child contained the 4 tasks of the CAI: PN, NWI, WR, NWR, and MRR [31, 32].

The assessment was administered at the child's nursery or primary school in a quiet room (or the room with the least possible background noise). The CAI was administered using a laptop, and the acoustic signal was automatically stored on the computer's hard disk in one recording for each of the different tasks. The child and research assistant were seated in front of the computer next to each other with a microphone and both wore a headset, or speakers were present, to provide a good sound level of the automated instruction of the CAI. The whole CAI would take approximately 30 min with the MRR being the last section of the CAI. The administration of the MRR took about 5–10 min per child.

MRR Administration

A protocol (Table 2) for the assessment of the MRR task was developed based on previous studies in Dutch and other languages [14, 15, 33]. The instruction was given by the CAI computer program to maximize standardization, and the children were asked to imitate the following sequences: first 3 monosyllabic sequences (/papa./, /tata./, and /kaka./), followed by 1 trisyllabic sequence (/pataka.../) and finally 2 bisyllabic sequences (/pata./ and /taka./). First, the children were asked to repeat a short sequence of 3 syllables (e.g., /papapa/) in a normal speaking rate,

Table 1. Age and gender for the 14 age groups of the normative sample

Age group (years)	Mean age (years)	Total number of subjects	Gender		Subjects per sequence, <i>n</i>					
			boys	girls	/pa/	/ta/	/ka/	/pataka/	/pata/	/taka/
2.0–2.3	2.1	72	30	42	59	59	58	55	57	52
2.4–2.7	2.5	102	55	47	79	81	80	70	77	66
2.8–2.11	2.9	101	46	55	83	82	81	71	81	69
3.0–3.3	3.1	104	52	52	90	90	88	83	89	78
3.4–3.7	3.5	110	61	49	90	92	94	89	94	83
3.8–3.11	3.9	102	57	45	95	95	94	86	94	90
4.0–4.3	4.1	100	55	45	85	84	81	80	83	83
4.4–4.7	4.5	115	60	55	93	94	94	89	95	91
4.8–4.11	4.9	116	56	60	94	94	94	91	92	90
5.0–5.3	5.1	121	66	55	104	106	106	103	104	103
5.4–5.7	5.5	128	71	57	113	111	114	109	112	114
5.8–5.11	5.9	117	64	53	103	105	104	101	102	104
6.0–6.5	6.2	117	69	48	107	108	107	104	108	106
6.6–6.11	6.8	119	57	62	108	108	109	108	109	109
Total		1,524	799	725	1,303	1,309	1,304	1,239	1,297	1,238
Sample, %		100	52.4	47.6	84.5	84.9	84.6	80.4	84.1	80.3

Table 2. Assessment protocol for the MRR

	Sequence	Trial	Instruction
1	pa	papapa pa...6× pa...12× pa...≥9×	Sequence of 3 syllables, normal speech rate Sequence of 6 syllables, normal speech rate After an audio example, a faster speech rate (5 syllables per second), sequence of 12 syllables As fast as possible (without an example) a sequence of minimal 9 syllables
2	ta	Tatata ta...6× ta...12× ta...≥9×	Sequence of 3 syllables, normal speech rate Sequence of 6 syllables, normal speech rate After an audio example, a faster speech rate (5 syllables per second), sequence of 12 syllables As fast as possible (without an example) a sequence of minimal 9 syllables
3	ka	kakaka ka...6× ka...12× ka...≥9×	Sequence of 3 syllables, normal speech rate Sequence of 6 syllables, normal speech rate After an audio example, a faster speech rate (5 syllables per second), sequence of 12 syllables As fast as possible (without an example) a sequence of minimal 9 syllables
4	pataka	pataka pataka...4× pataka...4× pataka...≥3×	Sequence of 3 syllables, normal speech rate Sequence of 12 syllables, normal speech rate After an audio example, a faster speech rate (5 syllables per second), sequence of 12 syllables As fast as possible (without an example) a sequence of minimal 9 syllables
5	pata	pata pata...3× pata...6× pata...≥4×	Sequence of 2 syllables, normal speech rate Sequence of 6 syllables, normal speech rate After an audio example, a faster speech rate (5 syllables per second), sequence of 12 syllables As fast as possible (without an example) a sequence of minimal 8 syllables
6	taka	taka taka...3× taka...6× taka...≥4×	Sequence of 2 syllables, normal speech rate Sequence of 6 syllables, normal speech rate After an audio example, a faster speech rate (5 syllables per second), sequence of 12 syllables As fast as possible (without an example) a sequence of minimal 8 syllables

MRR, maximum repetition rate.

Table 3. Analysis protocol for calculating the MRR

The sequence is considered correct if:

- The syllables are pronounced fluently in succession; dialect variances are accepted
-

The sequence is considered partially correct if:

- The sequence contains a single error (e.g., /papatapapapa/); then the sequences before and after the error are considered, and the longest and best sequence (at least 3 syllables) is selected
 - The sequence contains noise or other interfering elements, but a good sequence can be analyzed before or after the noise or interfering element; then the longest and best sequence is selected (at least 3 syllables)
 - The sequence contains pauses or interruptions; then the series are evaluated before and after the pause, and the longest and best sequence (at least 3 syllables) is selected; pauses can arise from:
 - Inhalation: The child inhales during the execution of the sequence and then continues with the sequence; this also applies to syllables that are pronounced on an inhalation
 - Rhythm: The child deviates from the rhythm of the sequence and a pause occurs; this is seen in waveform representation with a striking distance between 2 syllables
-

The sequence is considered incorrect if:

- The sequence in total consists of 4 syllables or less
 - The sequence is influenced by phonological processes (e.g., substitution, reduction, assimilation, metathesis, addition); these sequences were marked in an excel file for error analysis
 - The sequence is influenced by one of the following issues:
 - Noise or other interfering elements
 - Noise due to an interruption on the part of the examiner or other audible sounds, that makes the individual syllables unrecognizable
 - Sound volume
 - Low volume that makes the individual syllables unrecognizable
 - Syllables cannot be distinguished
 - Syllables in the waveform cannot clearly be distinguished from each other
-

MRR, maximum repetition rate.

followed by a longer sequence of 6 syllables in a normal rate (e.g., /papapapapapa/). The next instruction included imitation of a series of several syllables at a faster rate (the audio example contained 12 syllables at a faster rate). Finally, the children were asked to produce the syllable sequences as fast as possible. If necessary, the child got 3 attempts for every sequence (the CAI was programmed a maximum of 3 attempts per sequence) to collect an accurate or faster repetition of the sequence; the third attempt was given if the first 2 were both incorrect or the research assistant had the impression that the child could produce a faster rate.

If a child refused to utter a sequence, the research assistant tried to motivate the child and the sequence would be repeated or the research assistant presented the next sequence. If the child kept on refusing during the next sequences, the session was ended.

MRR Analysis

After all the data with the basic protocol were collected, the process of analyzing the samples started with the goal to maybe alter the protocol procedure of assessing and analyzing the MRR task. Since the program stored one whole recording of all trials per child, the recordings were cut in smaller sequences by hand with Praat software, version 6.0.21 [24].

Six students of HAN University of Applied Sciences and 3 SLPs analyzed the mono-, tri-, and bisyllable sequences according to a protocol, which is shown in Table 3. They were all trained by the first author and started with analyzing one practice sample of one audio-recording, which contained all the 6 sequences. The stu-

dents received instructions on how to use and interpret the protocol (e.g., which syllable sequence is suitable for further analyses if the child took a breath or pause?). Only the last 2 items of the MRR task were analyzed (those elicited by the instructions “faster” and “as fast as possible”). Any occurring speech errors were registered per sequence in an excel file (e.g., /papadada/).

The audio-recordings, containing just one sequence and attempt, were analyzed by the first author and one of the SLPs with the help of a customized Praat-script (developed by one of the authors; HT). The script detected and marked syllable onsets by localizing the noise burst of the voiceless plosives. The first and the last syllables were excluded because speakers often produce the first syllable with a longer duration and higher intensity [14] and the last syllable is also often lengthened [34]. Before extracting the syllable durations and MRR, the marked syllable onsets were depicted in the waveform and inspected visually, and any errors in the number of syllables indicated by the script were corrected manually. If corrected manually, the script could not give the separate durations of all the individual syllables and only the MRR value (total number of syllables divided by total duration of the sequence) was given. Figure 1 gives an example of one of the sequences with the markers. Only sequences with a remaining minimum of 3 syllables were included in the analysis. In some cases, the script could not detect syllable onsets correctly. These samples were analyzed by hand to determine the number of syllables and the duration of the sequence. Eventually, all data of the MRR were merged in SPSS version 24 for Windows (SPSS Inc., Chicago, IL, USA).

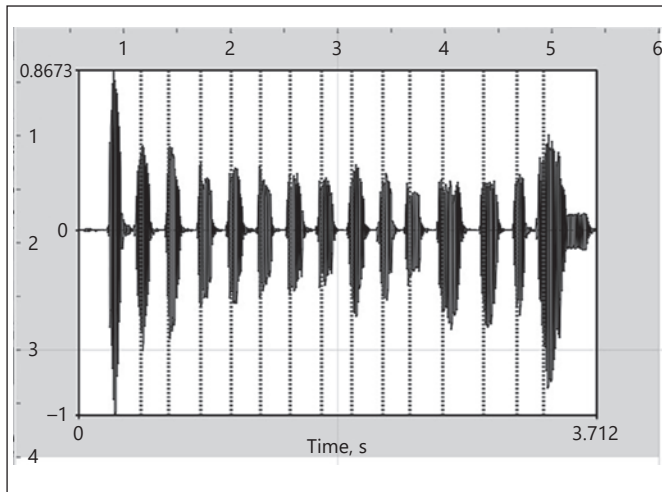


Fig. 1. Example of the analysis with the Praat-script of one sequence /tatata/, fastest first attempt.

Reliability

Interrater and test – retest reliability were examined and described. We will only describe the result of this study; all details can be found in the publication by van Haaften et al. [32, 35]. Interrater reliability was good for the monosyllabic sequences /pa/ (intra-class correlation coefficient [ICC] 0.81) and /ka/ (ICC 0.83) and sufficient for /ta/ (ICC 0.77). For /pataka/, /pata/, and /taka/, we found insufficient interrater reliability with ICCs ranging from 0.41 to 0.62. Especially, the younger children had difficulty performing the /pataka/, /pata/, and /taka/; had we included whether the attempts were successful or not, the ICC might have been much higher. Test – retest reliability showed a sufficient reliability measure (ICC 0.70) for /pa/, and for the other sequences, the test – retest reliability was insufficient with ICCs ranging from 0.18 to 0.60. Reasons for these low scores could be the rapid development of the younger children during the interval between test and retest or a test- retest training effect because children were significantly more competent on /pataka/ on the second test than on the first test ($t[53] = -3.02, p = 0.004$).

Statistical Analysis

First, frequency tables were constructed to determine how many children produced the different sequences of the MRR. Then, a comparison was made between the completion of the different sequences, for example, monosyllabic sequences versus multisyllabic sequences. Frequency tables were also constructed for the MRR value of the different sequences and attempts. Means and SDs of all parameters were calculated per age group, and repeated-measures analysis of variances was conducted to compare the best performance on the fast (with example) and the fastest (without example) attempt per sequence. Furthermore, to determine if the gold standard of 10 syllables should be maintained, means and SDs over all the first 3–10 syllables each were calculated per sequence and differences were investigated. Furthermore, ICCs with 2-way mixed-effects models featuring no fixed effects were calculated between the MRRs over each of the numbers of syllables compared with the gold standard of 10 syllables.

Results

First, the results of all children are described to answer the questions if children of all ages can perform the MRR tasks and if all children can perform all the different MRR tasks (mono-, bi-, and trisyllabic sequences). Subsequently, we investigate whether one of the instructions (fast or faster) elicits faster MRRs and whether it matters to ask multiple attempts per sequence. The last part of the results addresses the question if there is a difference in MRR when calculated over <10 syllables per sequence.

Ability to Perform the MRR Task

Tables 4 and 5 show the number of children executing and failing the different sequences of the MRR. For the 2–4-year-olds, not all audio-recordings included all MRR sequences because sometimes the child refused to utter one or multiple sequences and sometimes the SLP foresaw that the child would not execute the bi- and trisyllabic sequences after finishing the monosyllabic sequences. These cases are marked No sequence in Tables 4 and 5. A sequence is marked *Fail* if the child refused to complete the sequence, if not enough syllables were detected (minimum of 3), if an irregular rhythm (distinct pause) was executed, or if the child made errors (e.g., /pada/ instead of /pata/). For /pa/, 62 children refused to utter any syllables, for /ta/ 91 children, /ka/ 77 children, /pataka/ 156 children, /pata/ 100 children, and /taka/ 129 children. For each of the monosyllabic sequences, the results show that about 80% of children could produce the sequence correctly. For the multisyllabic sequences, the percentage of children that could produce the sequence correctly is lower, that is, 65.1% for /pataka/ and slightly higher percentages for /pata/ (75.9) and /taka/ (77.7%).

Next, we investigated the number of correctly produced sequences per individual. Table 6 provides an overview of the number of monosyllabic sequences that children in the different age groups have performed, showing that only 21% of the children under the age of 3 can perform all 3 monosyllabic sequences.

In order to determine the capability of carrying out the bi- and trisyllabic sequences in relation to the children's abilities to produce the monosyllabic sequences, we cross-tabulated the number of correctly produced monosyllabic sequences with the correct production of the bi- and trisyllabic sequences (Table 7). The results indicate that children who can produce at least 2 monosyllabic sequences are more likely to subsequently also correctly produce a bi- or trisyllabic sequence. The children who can only produce <2 monosyllabic sequence have a much

Table 4. Fail and pass of all monosyllabic sequences

Age group (years)	/pa/					/ta/					/ka/				
	n	pass, %	fail, % errors	<3 syllable	no sequence, %	n	pass, %	fail, % errors	<3 syllable	no sequence, %	n	pass, %	fail, % errors	<3 syllable	no sequence, %
2.0–2.4	59	30.5	1.7	39.0	28.8	59	32.2	1.7	32.2	33.9	58	34.5	3.4	37.9	24.1
2.4–2.8	79	35.4	2.5	44.3	17.7	81	34.6	3.7	37.0	24.7	80	35.0	6.3	32.5	26.3
2.8–3.0	83	66.3	3.6	21.7	8.4	82	64.6	1.2	20.7	13.4	81	63.0	3.7	23.5	9.9
3.0–3.4	90	71.1	1.1	18.9	8.9	90	75.6	0.0	13.3	11.1	88	75.0	2.3	14.8	8.0
3.4–3.8	90	66.7	5.6	20.0	7.8	92	71.7	1.1	14.1	13.0	94	69.1	5.3	18.1	7.4
3.8–4.0	95	90.5	1.1	5.3	3.2	95	87.4	2.1	5.3	5.3	94	88.3	3.2	3.2	5.3
4.0–4.4	85	85.9	7.1	3.5	3.5	84	90.5	2.4	3.6	3.6	81	91.4	1.2	2.5	4.9
4.4–4.8	93	96.8	0.0	1.1	2.2	94	93.6	2.1	2.1	2.1	94	92.6	3.2	1.1	3.2
4.8–5.0	94	97.9	0.0	1.1	1.1	94	97.9	1.1	0.0	1.1	94	96.8	0.0	0.0	3.2
5.0–5.4	104	99.0	1.0	0.0	0.0	106	94.3	0.9	1.9	2.8	106	95.3	1.9	0.0	2.8
5.4–5.8	113	99.1	0.0	0.9	0.0	111	94.6	2.7	0.0	2.7	114	95.6	2.6	0.0	1.8
5.8–6.0	103	99.0	1.0	0.0	0.0	105	100.0	0.0	0.0	0.0	104	99.0	1.0	0.0	0.0
6.0–6.6	107	98.1	1.9	0.0	0.0	108	99.1	0.9	0.0	0.0	107	98.1	1.9	0.0	0.0
6.6–7.0	108	99.1	0.9	0.0	0.0	108	97.2	1.9	0.0	0.9	109	99.1	0.9	0.0	0.0
Total	1,303	1,095	24	122	62	1,309	1,095	20	103	91	1,304	1,091	33	103	77
Sample, %	100.0	84.0	1.84	9.4	4.8	100.0	83.7	1.5	7.9	6.9	100.0	83.7	2.5	7.9	5.9

Table 5. Fail and pass of all tri- and bisyllabic sequences

Age group (years)	/pataka/					/pata/					/taka/				
	n	pass, %	fail, % errors	<3 syllable	no sequence, %	n	pass, %	fail, % errors	<3 syllable	no sequence, %	n	pass, %	fail, % errors	<3 syllable	no sequence, %
2.0–2.4	55	16.4	23.6	18.2	41.8	57	22.8	15.8	35.1	26.3	52	23.1	13.5	11.5	51.9
2.4–2.8	70	17.1	24.3	20.0	38.6	77	29.9	11.7	32.5	26.0	66	34.8	15.2	13.6	36.4
2.8–3.0	71	23.9	32.4	11.3	32.4	81	45.7	11.1	24.7	18.5	69	52.2	10.1	10.1	27.5
3.0–3.4	83	47.0	21.7	13.3	18.1	89	57.3	14.6	13.5	14.6	78	66.7	10.3	7.7	15.4
3.4–3.8	89	42.7	24.7	9.0	23.6	94	60.6	10.6	18.1	10.6	83	59.0	14.5	6.0	20.5
3.8–4.0	86	62.8	17.4	7.0	12.8	94	75.5	9.6	9.6	5.3	90	86.7	4.4	1.1	7.8
4.0–4.4	80	68.8	17.5	0.0	13.8	83	80.7	9.6	2.4	7.2	83	80.7	9.6	1.2	8.4
4.4–4.8	89	75.3	18.0	1.1	5.6	95	89.5	4.2	1.1	5.3	91	89.0	6.6	0.0	4.4
4.8–5.0	91	68.1	24.2	0.0	7.7	92	84.8	10.9	0.0	4.3	90	81.1	13.3	0.0	5.6
5.0–5.4	103	79.6	14.6	0.0	5.8	104	91.3	4.8	1.0	2.9	103	84.5	12.6	1.0	1.9
5.4–5.8	109	83.5	12.8	0.0	3.7	112	89.3	7.1	0.0	3.6	114	91.2	5.3	0.0	3.5
5.8–6.0	101	91.1	8.9	0.0	0.0	102	94.1	5.9	0.0	0.0	104	91.3	7.7	0.0	1.0
6.0–6.6	104	86.5	11.5	0.0	1.9	108	95.4	4.6	0.0	0.0	106	94.3	5.7	0.0	0.0
6.6–7.0	108	91.7	7.4	0.0	0.9	109	100.0	0.0	0.0	0.0	109	96.3	3.7	0.0	0.0
Total	1,239	807	218	58	156	1,297	985	105	107	100	1,238	962	111	36	129
Sample, %	100.0	65.1	17.6	4.7	12.6	100.0	75.9	8.1	8.3	7.7	100.0	77.7	9.0	2.9	10.4

lower chance of performing a tri- and bisyllabic sequences, showing a weak positive relation ($r_s = 0.278$, $n = 1,524$, $p < 0.001$).

Choosing the Best Sequence and the Number of Syllables

Children under 35 months of age have more difficulty with executing the different sequences for that reason

these children were excluded in further analyses which resulted in the inclusion of 1,041 children.

During data collection, the question raised which attempt or sequence (the one after the instruction “faster” or “as fast as possible”) would actually be the fastest MRR, as we observed that children do not always go faster if they have been instructed to go as fast as possible. In addition, some children got up to 3 attempts to produce a sequence

Table 6. Numbers of children producing 0–3 of the 3 monosyllabic sequences correctly per age group

Age group (years)	Monosyllabic sequence			
	0	1	2	3
2.0–2.4	42	12	9	9
2.4–2.8	58	18	12	14
2.8–3.0	32	14	20	35
3.0–3.4	23	13	19	49
3.4–3.8	31	14	18	47
3.8–4.0	8	8	14	72
4.0–4.4	18	5	13	64
4.4–4.8	23	2	8	82
4.8–5.0	23	0	4	89
5.0–5.4	16	2	7	96
5.4–5.8	14	3	10	101
5.8–6.0	12	1	3	101
6.0–6.6	9	0	7	101
6.6–7.0	10	0	7	102
Total	319	92	151	962
Sample, %	20.9	6.0	9.9	63.1

fast or as fast as possible because the administrator estimated that the child could go faster. As there is no evidence in literature, to our knowledge, regarding which attempt is the fastest overall, we compared the performance of the children after the instructions “fast” and “as fast as possible,” as well as the performances on the extra attempts. Of the total group, 742 children got >1 attempt for at least one of the sequences. To determine whether instruction has an effect on the realized rate, we compared the best attempt of the children on the “fast” instruction with the best attempts on the “as fast as possible” instruction (Table 8). A repeated-measures analysis of variance yielded significant effects of instruction for all sequences except for /ka/ (Table 8), indicating that for all sequences except /ka/, children achieved a higher rate on average upon the instruction “as fast as possible” as compared to the preceding “faster” instruction. However, the data must be interpreted with caution, since the effect sizes are rather small, in particular for the monosyllabic sequences (Table 8).

Number of Syllables

In recent MRR protocols [9, 14–16], the number of syllables that are required/prescribed for analysis is 10 or 12 syllables per sequence. Our clinical experience, however, is that not many children can produce 10- or 12-syllable sequences, especially children with MSD. Because the aim

of our research project is to develop an assessment for children with MSD, it is important to evaluate if the protocol can also be administered with <10 syllables. Therefore, Kruskal-Wallis test (none of the sequences met the test for equality of variance) per sequence was executed to see if there are differences between the MRR values for each of the sequence lengths with a minimum of 3 syllables and combining the sequences longer than 10 syllables. Sequence /pa/ showed a significant result, and no significant difference in syllable rate between the different sequence lengths was observed for the other sequences (Table 9). In Table 10, the descriptive values of the mono-, tri-, and bisyllabic sequences are presented.

To compare MRR when calculated over different numbers of syllables per child and not between the children as described in Table 10, the MRRs of each successive number of syllables were calculated for children who produced 10 or more syllables in a sequence. Differences between the mean syllable rate for each of the successive sequence lengths from 3 to 9 were studied by estimating ICCs. Table 11 shows good to excellent ICCs for every sequence length (except for the mean syllable rate of sequence length 3 in comparison with the mean rate sequence length 10 of /pata/, which has a moderate ICC).

Discussion

In this study, we adapted an existing MRR protocol and evaluated this protocol in a sample of 1,524 typically developing Dutch children from 2 to 7 years old; the largest group of children of which MRR assessment is described thus far. The results showed first that children under 30 months of age have severe difficulty with executing the tasks properly and even for children up to 3 years of age, it is still difficult. Most of the previous studies [16, 36] described groups of children from 3 years and older, simply because this is the youngest age at which children tend to be referred to an SLP [37]. Although there still is much debate about administering the MRR at this young age, these studies concluded that children from 3 years of age can perform the MRR task. The present results corroborate and extend these findings in a large sample, showing that administering MRR tasks in younger children is indeed problematic. For that reason, we conclude that MRR should not be assessed in children under the age of 3 and we adjusted the MRR protocol for future use accordingly (which is part of the CAI test battery).

Second, the results showed that children who have difficulty performing the monosyllabic series cannot per-

Table 7. Cross-tabulation of numbers of children correctly producing 0–3 bi- or trisyllabic sequences in relation to their number of correctly produced monosyllabic sequences

Monosyllabic sequences	Tri- and bisyllabic sequences									
	none		one		two		three		total	
	<i>n</i>	%	<i>n</i>	%	<i>n</i>	%	<i>n</i>	%	<i>n</i>	%
None	300	19.7	15	1.0	3	0.2	1	0.1	319	20.9
One	50	5.3	17	1.1	17	1.1	8	0.7	92	6.0
Two	29	1.9	24	1.6	56	3.7	42	3.8	151	9.9
Three	33	2.2	84	5.5	226	14.8	619	56.6	962	63.1
Total	412	27.0	140	9.2	302	19.8	670	44.0	1,524	100.0

Table 8. Comparison of the best performance on the 2 instructions “faster” (with example) and “as fast as possible” (without example)

Sequence	Instruction	<i>n</i>	MRR mean	SD	df	F	<i>p</i> value	ω^2
/pa/	Faster	790	4.4	0.6	1,550	26.601	<0.001	0.009
	Fastest	752	4.5	0.7				
/ta/	Faster	821	4.2	0.5	1,615	56.115	<0.001	0.0019
	Fastest	719	4.4	0.7				
/ka/	Faster	829	4.2	0.5	1,613	0.314	0.575	0.000
	Fastest	775	4.2	0.6				
/pataka/	Faster	687	3.7	0.7	1,411	84.558	<0.001	0.049
	Fastest	627	4.1	1.0				
/pata/	Faster	735	4.1	0.6	1,477	88.687	<0.001	0.049
	Fastest	707	4.5	0.9				
/taka/	Faster	743	4.1	0.6	1,476	75.459	<0.001	0.041
	Fastest	677	4.4	0.9				

n gives the number of children who produced the sequence; the statistical test and the calculation of mean and SD were conducted with repeated-measures ANOVA on less (*df* + 1) pairwise comparisons.
MRR, maximum repetition rate.

form the bi- and trisyllabic sequences. In itself this seems obvious since the bi- and trisyllabic sequences are articulatorily much more difficult to pronounce than the monosyllabic sequences [6, 38]. The importance, however, is that this establishes that MRR for the monosyllabic sequences and MRR for the bi- and trisyllabic sequences should be separate outcome measures that should both be included in the MRR task report. Furthermore, we included in the protocol that the bi- and trisyllabic sequences should not be administered if children could not produce the monosyllabic sequences to reduce the burden of the test battery.

According to the assessment protocol, the test administrator is instructed to ask children to redo the sequence

Table 9. Kruskal-Wallis statistics for the comparison of MRR values for the different numbers of syllables in the produced sequences

Sequence	<i>n</i>	df	H	<i>p</i> value
/pa/	983	8	20.29	0.009
/ta/	987	8	12.99	0.112
/ka/	981	8	14.21	0.076
/pataka/	893	8	7.472	0.487
/pata/	953	8	13.82	0.087
/taka/	934	8	15.51	0.050

MRR, maximum repetition rate.

Table 10. Mean (and SD) syllable rate for each of the different sequence lengths per number of syllables

Syllables, <i>n</i>	/pa/ (<i>n</i> = 992)			/ta/ (<i>n</i> = 996)			/ka/ (<i>n</i> = 990)			/pataka/ (<i>n</i> = 902)			/pata/ (<i>n</i> = 962)			/taka/ (<i>n</i> = 943)		
	<i>n</i>	M	SD	<i>n</i>	M	SD	<i>n</i>	M	SD	<i>n</i>	M	SD	<i>n</i>	M	SD	<i>n</i>	M	SD
3	183	4.5	0.8	165	4.4	0.8	140	4.1	0.7	120	4.2	1.2	115	4.2	0.8	111	4.2	0.9
4	158	4.6	0.7	161	4.4	0.7	143	4.2	0.7	142	4.1	0.9	156	4.5	1.0	119	4.5	0.9
5	155	4.6	0.6	129	4.6	0.7	124	4.2	0.6	113	4.1	1.0	128	4.4	0.9	100	4.4	0.9
6	112	4.5	0.6	104	4.4	0.7	135	4.2	0.6	101	4.1	0.9	127	4.6	2.0	128	4.6	0.9
7	107	4.7	0.6	107	4.4	0.7	131	4.3	0.5	113	4.0	0.9	96	4.5	0.7	119	4.4	0.8
8	75	4.6	0.5	76	4.4	0.6	90	4.2	0.6	63	3.9	0.8	92	4.5	0.9	83	4.3	0.7
9	68	4.7	0.7	82	4.5	0.5	74	4.3	0.5	68	4.1	0.8	60	4.6	0.7	70	4.4	0.7
10	43	4.6	0.6	61	4.5	0.5	45	4.2	0.5	62	3.9	0.8	53	4.6	0.7	65	4.5	0.7
>10	91	4.7	0.5	111	4.6	0.6	108	4.3	0.6	120	3.9	0.8	135	4.5	0.7	148	4.5	0.7

up to 2 times again if he/she suspects it was not performed at the child's maximum capacity. In 14% of the cases, the child was asked to repeat a sequence, and in 2% of the cases, the child got a third attempt of one or more of the sequences. Our results showed that most children were actually the fastest at the first attempt compared to the other attempts; only about 20% of the children were faster on the second or third attempt. However, it seems important to give children a second or even a third attempt if the administrator expect children to be even faster, because for about 12% of the attempts, the child was faster at the second or third attempt. In most protocols, there is a gradual build-up of number of syllables and pace of the sequences to be produced. After several trials, the children can be asked to produce the sequence as quickly as possible without an example. The expectation was that children show the fastest rate with the instruction to go as fast as possible, but this has not been explored in any published data. On the surface, a substantial number of children performed the fastest MRR with the instruction to go faster. The results showed a distinct pattern underneath. For the 2 monosyllabic sequences /pa/ and /ta/ and for the bi- and trisyllabic sequences, the instruction to perform the sequence "as fast as possible" yielded the fastest MRR, while for /ka/, the performance was the same between the 2 instructions. However, the effect sizes are very small, and therefore, it is debatable if the difference between the performance for the 2 instructions is clinically relevant. The difference could be an effect of learning how to conduct the task. Within the protocol, the child first practices the sequence (build up), and when the child is familiar with the sequence, the child is asked to produce it as fast as possible, thereby requiring maximal performance. However, we noticed children going louder and not that

much faster, and the effect size of the difference between the 2 instructions is very small. The advice is to choose the fastest attempt that can be performed with either the last or the second last instruction and/or attempt.

Recent studies report the use of 10–12 syllables [9, 14–16]. However, this study showed that a large number of especially the youngest children do not reach the criterion of sequence length 10. Instead, they produce sequence lengths in the range from 3 to about 10 syllables after exclusion of the first and last syllables. Gadesmann and Miller [21] compared the following methods of the same sequence children pronounced: number of syllables for the first 5 s, the time of pronouncing a number of repetitions (5 times) and the total duration of the maximum sequence length uttered in one breath, and thereby showed that the results are identical irrespective of the method of assessment. Based on this study and our own data, we conclude that a sequence of at least 5 syllables, such that the mean rate is based on measuring the duration of at least 3 syllables, is sufficient.

MRR is the most common measure, but in the literature, there are also indications that other measures of the MRR task can provide valuable information on the development of speech motor skills and therefore a better understanding of the underlying problems in children with MSD. In children with MSD, measuring speech variability can yield important information about the speech motor control system and to support the identification, assessment, and treatment of the underlying speech process [12, 15, 25, 38–41]. The coefficient of variation of the syllable durations could be added to investigate the variability of the sequences, as well as the normalized pairwise variability index, which in previous studies has been used to investigate stress-timing and syllable-timing [42, 43].

Table 11. Intraclass correlation coefficients for the comparison of syllable rate for each of the sequence lengths 3–9 with 10 or more

Sequence	Comparison syllables	<i>n</i>	ICC	CI lb	CI ub
/pa/	3 with 10	92	0.904	0.858	0.936
	4 with 10	92	0.945	0.918	0.963
	5 with 10	92	0.966	0.949	0.977
	6 with 10	92	0.975	0.962	0.983
	7 with 10	92	0.985	0.978	0.990
	8 with 10	92	0.994	0.991	0.996
	9 with 10	92	0.997	0.996	0.998
/ta/	3 with 10	105	0.882	0.831	0.918
	4 with 10	105	0.910	0.871	0.938
	5 with 10	105	0.936	0.907	0.956
	6 with 10	105	0.965	0.949	0.976
	7 with 10	105	0.978	0.968	0.985
	8 with 10	105	0.991	0.987	0.994
	9 with 10	105	0.995	0.993	0.997
/ka/	3 with 10	102	0.843	0.776	0.891
	4 with 10	102	0.904	0.886	0.934
	5 with 10	102	0.929	0.897	0.952
	6 with 10	102	0.957	0.936	0.970
	7 with 10	102	0.977	0.966	0.984
	8 with 10	102	0.986	0.980	0.991
	9 with 10	102	0.995	0.993	0.997
/pataka/	3 with 10	58	0.881	0.807	0.928
	4 with 10	58	0.899	0.835	0.939
	5 with 10	58	0.913	0.857	0.947
	6 with 10	58	0.962	0.936	0.977
	7 with 10	58	0.982	0.969	0.989
	8 with 10	58	0.983	0.972	0.990
	9 with 10	58	0.989	0.981	0.993
/pata/	3 with 10	86	0.735	0.620	0.819
	4 with 10	86	0.806	0.718	0.869
	5 with 10	86	0.869	0.806	0.913
	6 with 10	86	0.918	0.877	0.946
	7 with 10	86	0.953	0.929	0.969
	8 with 10	86	0.978	0.966	0.985
	9 with 10	86	0.992	0.988	0.995
/taka/	3 with 10	105	0.824	0.752	0.877
	4 with 10	105	0.863	0.805	0.905
	5 with 10	105	0.905	0.864	0.935
	6 with 10	105	0.945	0.921	0.963
	7 with 10	105	0.974	0.962	0.982
	8 with 10	105	0.987	0.980	0.991
	9 with 10	105	0.995	0.992	0.996

ICC, intraclass correlation coefficient; CI lb, confidence interval lower bound; CI ub, confidence interval upper bound.

However, some reservation is required in this respect as the current speech-to-result set-up for most variability measures is not yet sufficiently automated to serve as an easily applicable analysis tool in the daily practice of speech therapists [42]. The goal for us is to see if and how the assessment of variability as an outcome measure of the MRR task could be implemented in the CAI.

In this study, we asked parents or caregivers whether their child had a history of hearing problems and if they had any doubts about his/her hearing. It is possible that the child could have a mild hearing problem because parents and caregivers can overlook a mild hearing problem [44]. In the Netherlands, the hearing of all children is recorded during the regular governmental hearing screening after 2 weeks after birth (neonatal screening) and at the age of 4 [45]. Furthermore, the research assistants were asked to pay particular attention to signs of hearing problems. This is why we did not include a whole hearing screening, but it is possible that a few children had a mild hearing problem.

In the field of adult MSD, there has been debate about the potential utility of nonspeech oral motor tasks [46–49], and recently, Staiger et al. [50] suggested that MRR is not a speech-like skill, and therefore, MRR is unusable in clinical assessment of MSD in adults. We would like to stress here, however, that results that hold for adults with acquired disorders do not necessarily hold for children with developmental disorders. As pointed out elsewhere in this special issue following Bishop [51] and Karmiloff-Smith [52, 53], developmental disorders are characterized by association rather than dissociation of functions [54, 55]. Whereas the adult speech production system is highly redundant, and the different processes and representations are highly overlearned, children have an incomplete system that is still in development. At the age of 4–6 years, children still make speech errors in conversational speech or in naming pictures that can be based on an incomplete phonological system or an immature motoric speech system [30]. The dissociation between MRR and other speech tasks found for adults thus cannot be extended to children. In fact, correlations between performance on speech tasks and different nonspeech motor tasks have been found in several groups of children with speech disorder, among which children with CAS [56] and children with Fetal Alcohol Spectrum Disorders [57].

In addition, and even more importantly, the MRR serves an important function in differential diagnosis of developmental speech disorders, as, for example, also expressed in the 2011 Speech-language pathology med-

ical review guidelines from the American Speech-Language-Hearing Association [58]. Several studies have reported differences between children with and without MSD on the MRR [14, 15], and the MRR has been shown to be discriminative between CAS and developmental dysarthria [14, 25]. We therefore propose that for a comprehensive speech assessment the following tasks should be administered: PN, NWI, WR, NWR, and MRR [32]. This study yields directions for administering the MRR tasks and norm values to interpret the performances relative to typically developing children. Research with diverse groups of children with SSD with the comprehensive test battery is required to validate the MRR and evaluate its contribution to the speech diagnosis. Such studies are currently conducted by our research group.

In summary, the new MRR protocol describes how to assess children from 3 years of age; if a child cannot perform >2 monosyllabic sequences, the session can be ended. In the clinical report of the MRR, the score for the monosyllabic and for the bi- and trisyllabic sequences must be given separately. Children do not have to be encouraged to perform a sequence of at least 10 syllables. For each MRR sequence, the test administrator should analyze the attempts the child has produced upon the last 2 instructions and then determine which attempt was the fastest.

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Statement of Ethics

Parents and guardians have given their written informed consent, and the schools/daycares have given their consent to recruit the children at their facility.

The research institute’s committee on human research (Radboudumc) declared the study can be carried out (in the Netherlands) without an approval by an accredited research ethics committee.

Disclosure Statement

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Author Contributions

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