International Journal of Pediatric Otorhinolaryngology 94 (2017) 23–29

Contents lists available at ScienceDirect

International Journal of Pediatric Otorhinolaryngology

journal homepage: http://www.ijporlonline.com/

Development and evaluation of the LittlEARS® Early Speech Production Questionnaire – LEESPQ

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ARTICLE INFO

Article history:
Received 8 November 2016
Received in revised form
6 January 2017
Accepted 6 January 2017
Available online 9 January 2017

Keywords:
LEESPQ
Early speech and language production
Speech development
Language development
Infant vocalization
Prelexical vocalization

ABSTRACT

Objective: Universal Newborn Hearing Screening programs, now instituted throughout the German-speaking countries, allow hearing loss to be detected and treated much earlier than ever before. With this earlier detection, arises the need for tools fit for assessing the very early speech and language production development of today's younger (0–18 month old) children. We have created the LittlEARS® Early Speech Production Questionnaire, with the aim of meeting this need.
Methods: 600 questionnaires of the pilot version of the LittlEARS® Early Speech Production Questionnaire were distributed to parents via pediatricians' practices, day care centers, and personal contact. The completed questionnaires were statistically analyzed to determine their reliability, predictive accuracy, internal consistency, and to what extent gender or unilingualism influenced a child's score. Further, a norm curve was generated to plot the children's increased expected speech production ability with age.
Results: Analysis of the data from the 352/600 returned questionnaires revealed that scores on LittlEARS® Early Speech Production Questionnaire correlate positively with a child's age, with older children scoring higher than do younger children. Further, the questionnaire has a high measuring reliability, high predictability, high unidemensionality of scale, and is not significantly gender or uni-/multilingually biased. A norm curve for expected development with age was created.
Conclusions: The LittlEARS® Early Speech Production Questionnaire (LEESPQ) is a valid tool for assessing the most important milestones in very early development of speech and language production of German language children with normal hearing aged 0–18 months old. The questionnaire is a potentially useful tool for long-term infant screening and follow-up testing and for children with normal hearing and those who would benefit from or use hearing devices.

1. Introduction

Infant hearing loss in Germany is being discovered earlier than ever before, thanks to an increasing use of Universal Newborn Hearing Screening (UNHS) tests. While such early detection greatly increases access to the benefits of early intervention and subsequent speech and language therapy, it presents a challenge for therapist. In the pre-UNHS days, when hearing-impairment was discovered later, diagnostic and assessment tools for the speech production of hearing impaired children focused on comparatively high word and linguistic levels; age-appropriate in the past but not for today's younger, less physically and audiologically developed hearing-device users.

While many screening tools are available on the German-speaking market, all of them insufficiently assess the preverbal expressive development of children less than 10 months old. Screening questionnaires such as the ELFRA-1 [1] for 12 month olds and the FRAKIS and FRAKIS-K [2] for 18–30 month olds are helpful but, according to scientific standards, their prognostic validities are not significant. The screening questionnaire SBE-2KT [3] is for 21–24 month olds. Diagnostic instruments fulfilling the quality criteria are, unfortunately, only available for older children. The

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http://dx.doi.org/10.1016/j.ijporl.2017.01.007
0165-5876/

ELAN [4] is for children aged 16–26 months. The ELFRA-2 [1] is for children aged 21–24 months. Tests assessing receptive and expressive semantic-lexical and morphologic-syntactical levels are also only available for older children: the SETK-2 [5] is for children aged 2 years–2 years 11 months; and the SETK 3–5 [6] is for children aged 3–5 years. The LittlEARS® Auditory Questionnaire (LEAQ) [7] is for children under 24 months old, but it assesses receptive, not productive, auditory behavior. The Functioning After Pediatric Cochlear Implantation (FAPCI) [8] assesses the development of communication performance on a family-centered scale, but is for children aged 2–5 years. The Infant-Toddler Meaningful Auditory Integration Scale (IT-MAIS) [9] is a parental interview with 10 questions that evaluates the meaningful use of sound in everyday situations (vocal behavior, attachment with hearing instrument, ability to alert to sound, and ability to attach meaning to sound). The Production Infant Scale Evaluation (PRISE) [10] uses a parent questionnaire to access the vocal development of infants with cochlear implants. For the German language, efforts have been made to develop a diagnostic instrument for therapy based on the Stark Assessment of Early Vocal Development – Revised (SAEVD-R) [11,12] which allows the classification of a child's utterances and the assessment of the developmental progress in his/her first 20 months of life and can be used to classify 3 main types of vocalization: vegetative sounds, fixed signals, and protophones. Thus far, results have only been preliminary [13].

We have been developing LEESPQ (LittlEARS® Early Speech Production Questionnaire) to meet the needs of children aged 0–18 months. It is our intention to use the LEESPQ within the LittlEARS® battery, alongside the LEAQ [14] and the My LittlEARS® Diary [15].

Based on a literature review and the clinical study data gathered in a longitudinal study on preverbal speech and language development [17,18] the first questionnaire has been developed by an experienced group of professionals. The version with 24 age-dependent questions addressed reflexive behavior, crying, prelingual utterances, canonical babbling, jargon, and first words of children aged 0–18 months [16]. A small, independent sample of parents, therapists, and physicians then assessed this questionnaire for 1) understandability of the questions and examples, 2) usability, 3) quality, and 4) completeness. We incorporated their suggestions into a revised version of the questionnaire. The LEESPQ is primarily intended for parents to complete after observing their child's natural behavior while at home or in a familiar setting. It also aims to provide professionals with crucial information about children's earliest developmental milestones. In addition to being convenient for the parent and non-invasive for the child, parent-completed questionnaires are useful diagnostic tools to assess infants' and toddlers' communication abilities [19]; although when speech therapists and professionals evaluate the completed questionnaires, they should remember that parents are prone to overestimating their children's abilities [20].

Our present purpose is to develop the LEESPQ and validate its ability to assess very early speech and language production development. Furthermore, we would like to determine the suitability of the LEESPQ (1) for documenting the speech and language development of normal-hearing children (2) as a general screening tool to support UNHS testing, (3) for use by pediatricians and ENT practices, and (4) as a test instrument for scientific studies.

2. Methods

2.1. Subject recruitment and inclusion criteria

600 pilot questionnaires of the LEESPQ (LittlEARS® Early Speech Production Questionnaire) were distributed to pediatricians and daycare centers in Mainz, Germany, and to daycare centers and parents in Innsbruck, Austria. The pediatricians and daycare centers functioned merely as distribution points wherefrom parents could pick up the questionnaire.

Parents were asked to complete the questionnaire based on their child's behavior. If they had seen the child exhibit the questioned behavior at least once, they were instructed to answer “yes”. If they hadn't or if they weren't sure, they were instructed to answer “no”. Every “yes” answer is worth 1 point; every “no” is 0 points. A space was provided at the end of the questionnaire for comments.

In order to be included, all children had to be 0–18 months old, seemingly without disability, and, according to the parents, of normal hearing. As the questionnaires were only available in the German language, we assumed that the children, if multilingual, were proficient in German.

2.1.1. Ethics

This validation study was approved by the Rhineland-Palatinate ethics committee, Germany (837.082.09 (6587)). Participants returned the signed declaration of consent when they returned their completed questionnaire.

2.2. A note on how the scores are determined

The maximum possible score on the pilot version of the LEESPQ was 47 points (including the production of consonants). Some of the 24 numbered questions have sub-questions whose answers contribute to the final score. Only closed questions (answerable with a yes/no) are worth points. All sub-questions which are open questions (i.e. cannot be answered yes/no) are not worth any points; rather, they have been kept in the questionnaire only to give a more detailed picture of a child's abilities, or to give additional information relevant to a child's cultural or language setting. To determine a child's final score, one must add up only the number of “yes” answers. See Appendix A for a sample of questions.

The LEESPQ contains a consonant table where children were awarded 1 point for each of the 16 consonant sounds in questions 13 and 13.1 (“Which consonant sounds does your child produce?”) that they could produce.

2.3. Validation procedure

As the LEESPQ is intended to complement the LittlEARS® test battery and be used, alongside the LEAQ, for screening and follow-up purposes, the research and validation for the LEESPQ were based on and managed in the same way as the LEAQ was [7].

A list of 24 questions referring to the speech production abilities of very young children was compiled and administered to the parents. The parents' answers were statistically analyzed within the model of the classical test theory in order to evaluate the psychometric characteristics of the remaining ones [21,22]. We used SPSS statistics 19 for statistical analyses [28].

2.4. Item and scale analyses

To assess the items' suitability for inclusion in the questionnaire, the index of difficulty, the discrimination index, and the selectivity index were calculated for each item. The present questionnaire's difficulty gives an indication for the age-dependence of the items. A high value means that an item is “easy”, while a low value indicates that an item is “difficult”.

The discrimination index indicates the correlation of an item with the total score. A high correlation means that the item has a considerable impact on the total score and clearly differentiates between children with highly developed speech production ability

and those without.

As a further criterion for evaluating the suitability of an item, the selectivity index was calculated. Items with a selectivity parameter (s) ≥ 0.3 and a correlation with age (r) ≥ 0.1 are rated as suitable for inclusion in the questionnaire.

For the scale analysis, the correlation between the total score and the age of the children (Pearson's correlation coefficient), the full test reliability of the questionnaire based on split-half reliability measures (Spearman–Brown split-half coefficient), the predictive accuracy of the scale (Guttman's lambda) and the internal consistency of the scale (Cronbach's alpha) were determined. Further, the influence of the child's gender and the influence of unilingualism versus multilingualism on the total score were assessed.

2.5. Establishing a norm curve

Standardized expected values were calculated from the data to generate a norm curve to see the development of speech production ability of children between 0 and 18 months of age. The standardized expected values were derived from a regression model or curve fitting with "age" as independent and "total score" as the dependent variable. A second order polynomial model according to the least squares method was the best model to generate a norm curve. From this structural equation, standardized expected values of age-dependent total scores were calculated for each age group. These values are considered to be the "normative scores" for the age-specific speech production ability of a child.

The results of the regression analysis were also used to determine the confidence intervals in which the age-specific values are found with 95% probability. The lower band of the confidence interval was defined as the critical lower limit (called minimum value) for children with normal hearing. Scores above this limit are considered consistent with normal, age-appropriate speech production development (with a probability of 95%), while scores below this critical lower limit could raise concern.

3. Results

3.1. Subjects

60.8% (365/600) of the questionnaires were returned, however 13 did not meet the inclusion criteria and were thus were excluded. 352 questionnaires were analyzed, yielding a study population of 161 male and 186 female children. 5 children were of unstated gender. The age ranged from 8 days (0 months) to 18 months old (see Table 1). Every child had, according to his/her parents, normal hearing and had been subject to a newborn hearing screening test. 72.2% (254/352) children were unilingual, 21.9% (77/325) were multilingual, and 6% (21/352) were of unstated linguistic background. As the questionnaire only asked if they were multilingual or bilingual, we don't know in which language the multilingual children were dominant.

Table 1
Number of children per age group.

Age (months)	Number of children	Age (months)	Number of children
0–1	10	>9–10	11
>1–2	11	>10–11	16
>2–3	18	>11–12	40
>3–4	39	>12–13	22
>4–5	16	>13–14	14
>5–6	25	>14–15	10
>6–7	47	>15–16	17
>7–8	16	>16–17	12
>8–9	8	>17–18	20

3.2. The validation results

3.2.1. Item and scale analyses

The results of the item analyses were not statistically satisfactory and therefore are not shown. This finding was expected because constructing a questionnaire for such young children necessitates including a wide range of items to assess their development. These items (e.g. questions: 1. Does your child cry to attract attention?; 2. Does your child make movements with her/his lips?; 3. Does your child make movements with her/his lips?) were nonetheless, included because they contain clinically important information.

The correlation between age and total score was calculated to obtain information about the ability of the questionnaire to measure age-dependent speech production ability. The determined value of $r = 0.775$ indicates a high and very significant correlation ($p < 0.001$) of the "yes" answers from the total score with the child's age as follows: The older the child, the more likely a higher total score will be reached.

The Spearman-Brown split-half coefficient was calculated to estimate the test reliability of the questionnaire. The resulting split-half reliability of $r = 0.546$ indicates the high measuring reliability. The predictive accuracy of the scale was calculated using Guttman's lamba (λ). The value found was $\lambda = 0.835$, which indicates high predictability.

Homogeneity or internal consistency is the unidimensionality of a scale, i.e. its suitability to assess exactly one construct (here: speech production ability). A measure for internal consistency is Cronbach's alpha (α), which generally increases when the correlations between the items increase. A value of $\alpha = 0.863$ was reached, which indicates that the questionnaire almost exclusively assesses speech production ability.

To determine if the LEESPQ is gender biased, the total scores of male children and female children were compared. Statistical analysis (Independent sample T-test: $t = 0.549$; $p = 0.583$) did not reveal significant differences. Age stratified results did not show significant differences between genders either. These results indicate that the questionnaire is suitable for both genders (Table 2).

Additionally, the total scores of unilingual ($n = 254$) and multilingual children ($n = 77$) and were compared. The unilingual group had a higher mean total score (19.8, $\pm SD = 5.29$) than the multilingual group (18.8, $\pm SD = 5.65$), but the difference was not significant (Independent sample T-test: $t = 1.387$; $p = 0.166$) (see Fig. 1).

3.2.2. Establishing a norm curve

Fig. 2 shows a scatter plot of the raw data, the norm curve generated from them and the lower band of the 95% confidence interval. The best model, explaining 61.1% of the entire variance (adjusted $R^2 = 0.611$) was a second order polynomial model according to the least squares method with the regression equation $y = 10.196 + 1.364 * age - 0.027 * age^2$.

The production of consonants shows a high variety amongst all tested children. Our study shows that throughout the age groups unvoiced fricatives (i.e.: /f/, /s/, /ʃ/), unvoiced plosives (i.e. /k/, /t/), and approximants (i.e.: /l/, /r/) are most difficult to produce. The voiced nasal consonant "m" seems to be easiest, followed by the voiced plosives/b/, and/d/ (See Table 3 for complete results).

4. Discussion

The results of this study confirmed that the LEESPQ (LittlEARS® early speech production questionnaire) is a statistically validated, accurate, gender independent, and age dependent tool for the quick and easy assessment of milestones in early speech production in

Table 2
Standardized expected mean value (with speech production development according to age) and standardized minimum values (lower 95% confidence interval) of age-dependent speech-production ability. The completed month defines the previous age category.

Age (months)	Expected value	Minimum value	Age (months)	Expected value	Minimum value
0–1	8	5	>9–10	20	14
>1–2	11	6	>10–11	21	15
>2–3	15	7	>11–12	22	16
>3–4	15	8	>12–13	23	16
>4–5	16	9	>13–14	23	17
>5–6	17	10	>14–15	23	18
>6–7	18	11	>15–16	25	18
>7–8	20	12	>16–17	25	19
>8–9	21	13	>17–18	27	19

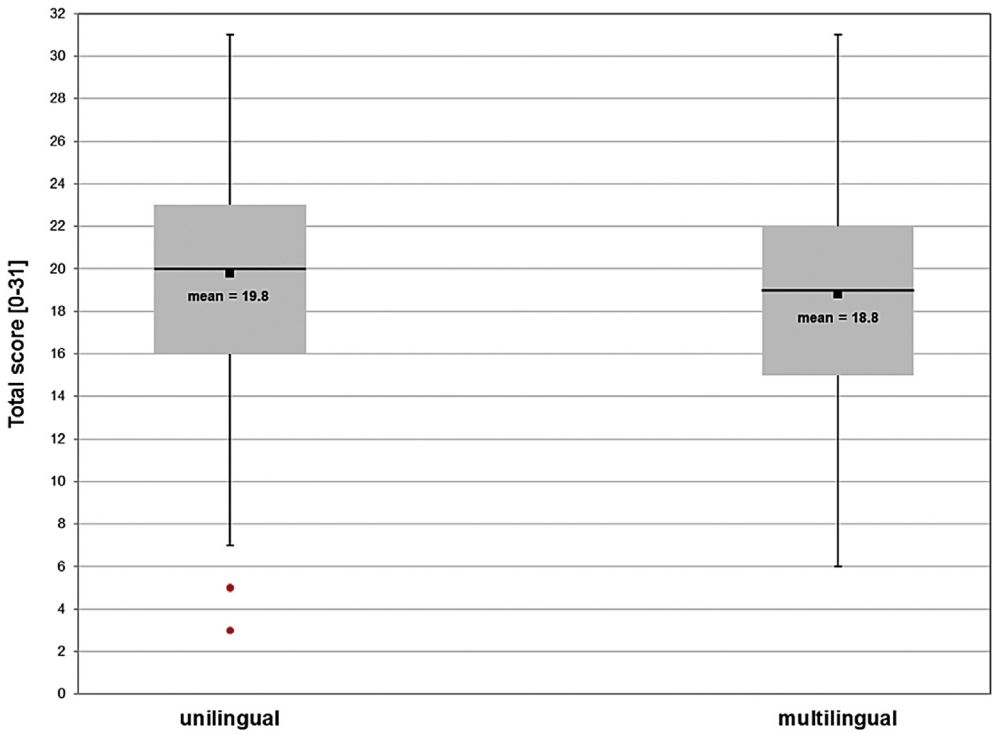

Fig. 1. Comparison of total scores between unilingual (n = 254) and multilingual children (n = 77). Note: Mean values are displayed as black quadrats, median values as horizontal lines. The red circles represent outliers. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

normal hearing German-language children, aged 0–18 months. This validation is an important preparatory step towards developing a validated assessment questionnaire for use with children aged 0–18 months with hearing loss, as their audio-lingual development requires more vigilant and proactive concern.

Toddlers begin learning their first words around the age of 1 year old. They learn to assign words to objects and to categorize words into concepts. While this 12-month mark is an indicator, previous studies have found a high variance amongst children [23]. This corresponds well with our findings. Based on literature review, we had assumed that children would produce their first words around the age of 12 months and two words utterances by the age of 18 months, however, in some children this wasn't the case. This indicates that the questionnaire detects age appropriate behaviors. Results below the critical value can be a sign of a possible hearing loss or other disability.

Few children in our sample scored the full 31 points (excluding the consonants points). The mean value at age 18 months was 27.1 points (range 21–31, SD: 3.04). 25% of all children scored below 25.75 points, 50% of the children scored below 28 points. 75% of the children scored below 29 points. Only 25% of all participating children at age 18 months scored above 29 points. The statistical analysis shows that some questions (especially the later questions) were more difficult, unsurprisingly since these asked such higher level abilities as producing or imitating identifiable words and sounds. This finding could be an indication that the age-limit of 18 months could be increased to 20–24 months. More data is needed to substantiate this.

The expected scores for each age group can be found in Table 2. If a child scores above the expected value, parents can feel confident with their child's speech production development. If, however, a child scores less than would be expected for his/her age, it may, but does not necessarily, indicate that the child's speech production is lower than age-appropriate. Such a score should be regarded a cause for investigation, not alarm, and the child should be retested in the presence of a professional (speech and language therapist). A complete anamneses was not done with the children included in the study, therefore this is only theoretical.

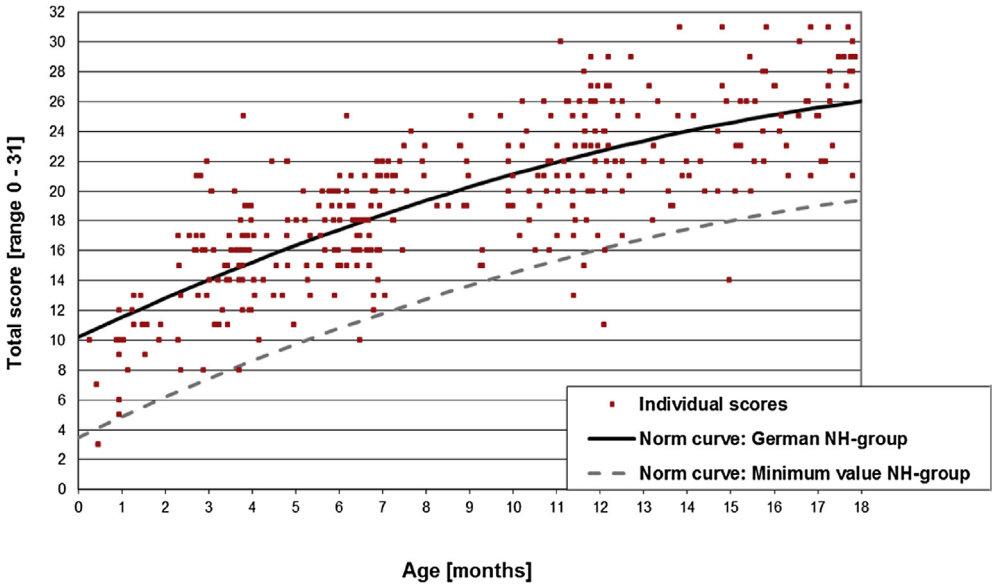

Fig. 2. Regression curve (quadratic) with age as independent and total score as the dependent variable. Individual scores, standardized expected values (norm curve) and standardized minimum values (lower 95% confidence interval) of age-specific speech production abilities of the German normal hearing sample.

Table 3
Distribution of consonants production by age group of infants with normal hearing. m = age in months; (%) = percental distribution within the age group; n = number of children within the age group;/M,N,B, … S/ = produced consonants; for example 5 (28) means: 5 out of 18 three months old infants produced/m/, these are 28% of this age group.

		M	N	B	P	D	T	L	F	J	W	R	SCH	G	K	NG	S
2m	n = 11 (%)	1 (9)	–	–	–	–	–	–	–	–	–	–	–	–	–	–	–
3m	n = 18 (%)	5 (28)	2 (11)	1 (6)	–	–	–	1 (6)	–	1 (6)	–	2 (11)	–	2 (11)		2 (11)	1 (6)
4m	n = 39 (%)	8 (20)	3 (8)	4 (10)	1 (3)	1 (3)	–	1 (3)	2 (5)	2 (5)	1 (3)	9 (23)	–	5 (13)	1 (3)	7 (18)	1 (3)
5m	n = 16 (%)	5 (31)	3 (19)	–	1 (6)	1 (6)	–	1 (6)	–	–	–	2 (13)	–	4 (25)	–	4 (25)	–
6m	n = 25 (%)	16 (64)	6 (24)	4 (16)	2 (8)	2 (8)	1 (4)	4 (16)	2 (8)	5 (20)	4 (16)	5 (20)	–	6 (24)	1 (4)	5 (20)	–
7m	n = 47 (%)	26 (55)	4 (8)	15 (32)	6 (13)	11 (23)	3 (6)	2 (4)	2 (4)	1 (2)	4 (9)	10 (21)	–	3 (6)	2 (4)	6 (13)	–
8m	n = 18 (%)	13 (81)	6 (38)	10 (63)	3 (19)	1 (6)	2 (13)	4 (25)	4 (25)	3 (19)	5 (31)	4 (25)	5 (31)	2 (13)	1 (6)	1 (6)	2 (13)
9m	n = 8 (%)	8 (100)	4 (50)	8 (100)	4 (50)	4 (50)	2 (25)	–	1 (13)	2 (25)	2 (25)	2 (25)	–	3 (38)	3 (38)	1 (13)	1 (13)
10m	n = 11 (%)	10 (91)	7 (64)	10 (91)	6 (55)	10 (91)	4 (37)	1 (9)	3 (27)	3 (27)	4 (36)	2 (18)	–	4 (36)	1 (9)	3 (27)	2 (18)
11m	n = 16 (%)	14 (88)	4 (25)	8 (50)	8 (50)	6 (38)	4 (25)	2 (13)	1 (6)	2 (13)	2 (13)	8 (50)	4 (25)	4 (25)	1 (6)	2 (13)	1 (6)
12m	n = 40 (%)	34 (85)	17 (43)	28 (70)	17 (43)	22 (55)	12 (30)	11 (28)	6 (15)	12 (30)	14 (35)	12 (30)	8 (20)	16 (40)	7 (18)	12 (30)	6 (15)
13m	n = 22 (%)	21 (91)	9 (39)	14 (61)	10 (44)	16 (70)	5 (22)	7 (30)	3 (13)	6 (26)	3 (13)	6 (26)	2 (9)	9 (39)	2 (9)	7 (30)	5 (22)
14m	n = 14 (%)	11 (79)	7 (50)	10 (71)	8 (57)	7 (50)	4 (29)	3 (21)	–	2 (14)	1 (7)	2 (14)	2 (14)	3 (21)	–	2 (14)	1 (7)
15m	n = 10 (%)	9 (90)	6 (60)	7 (70)	5 (50)	7 (70)	6 (60)	4 (40)	1 (10)	2 (20)	4 (40)	1 (10)	–	3 (30)	2 (20)	–	1 (10)
16m	n = 17 (%)	12 (71)	6 (35)	11 (65)	8 (47)	10 (59)	7 (41)	4 (24)	4 (24)	4 (24)	3 (18)	5 (29)	3 (18)	5 (29)	1 (6)	–	3 (18)
17m	n = 12 (%)	12 (100)	8 (67)	9 (75)	7 (58)	9 (75)	7 (58)	8 (67)	2 (17)	6 (50)	6 (50)	5 (42)	5 (42)	8 (67)	4 (33)	6 (50)	6 (50)
18m	n = 19 (%)	18 (95)	10 (53)	16 (84)	14 (74)	15 (79)	11 (58)	11 (58)	9 (47)	8 (42)	11 (58)	11 (58)	7 (37)	13 (68)	8 (42)	6 (32)	9 (47)

The LEESPQ is not sensitive to gender-specific differences in speech production development. Although evidence exists that male children have a higher within-family incidence of language disorders than do female children [24–26], it may be that these differences are more evident in late toddlerhood [27] than earlier. Our results on gender independency tend to support this, as our subjects were younger than 18 months, however, it is also possible that our sample size may not have been large enough.

Our results indicated that the unilingual group scored higher, although not significantly higher, than the multilingual group. This should be interpreted with caution as the multilingual group was much smaller than the unilingual group.

This study could have been improved if we had had a more equal number of children an each age group. While all the age groups had more than 10 children, as it was a prerequisite, the individual age groups weren't very cohesive, ranging from 10 to 47 children (mean 19.5). Future studies should consider this.

Future versions of the questionnaire could be more specific on determining if the testing language (here German) is, or is expected to become, the child's dominant language. Despite not finding a significant difference between unilingual and multilingual children's results, we believe it possible that if data gathered to validate the LEESPQ in a particular language is harvested from children who don't primarily hear/use that language, the results may be skewed. Thus, in future versions, unilingual/multilingual questions should be modified to make it possible to exclude children whose dominant language is not the language being tested.

This pilot German-language version of the LEESPQ will be adapted to an international version following results of this pilot study and like the LEAQ, will be adapted to many other languages; indeed research to this end is currently being conducted. Unlike the LEAQ, however, the LEESPQ is not language independent and as such will have to be validated separately for each language. In doing so, future studies must consider phonic differences between the

target language and the parent German questionnaire. Language independency or the independency across language families might be difficult to prove due to the different amount of consonants within each language. Further, children may start producing different consonants at different times depending on the language into which they are growing and how consonant-rich its lexis is. While the consonants inventory is still seen as an important part of the questionnaire, especially in the clinical evaluation, it should have a simple "Yes/No" answer possibility. Individual consonants are listed, and a child's production of them may be noted as a point of additional detail, but do they not count towards the final score. This should help to make results more comparable across languages.

Future research should also focus on validating the LEESPQ for use with children who would benefit from or are using hearing devices (e.g. hearing aids and/or cochlear implants). Future research could also explore if the LEESPQ can be productively used with children as old as 24 months.

5. Conclusion

The LittlEARS® Early Speech Production Questionnaire (LEESPQ) is a valid tool for assessing the most important milestones in very early development of speech and language production of German language normal hearing children aged 0–18 months old. It has good age-dependency, internal consistency, reliability, predictive accuracy, and is not influenced by gender. As it has an excellent correlation between age and total score, the LEESPQ is a potentially useful tool for long term follow-up testing (18 months or even 2 years) and for children who would benefit from or use hearing devices.

Conflicts of interest

Authors Joanna Brachmaier, Edda Amann and Vanessa Hoffmann are employed at MED-EL, as are Petra Kuss, Maria Wechselberger, and Michael Todd. Bianka Wachtlin and Annerose Keilmann received research funding from MED-EL.

Acknowledgments

We thank Petra Kuss and Maria Wechselberger for assisting in data collection and Michael Todd, who provided medical writing services, all on behalf of MED-EL, GmbH. Furthermore, many parents and teachers in various day nurseries in Innsbruck, Austria and Mainz, Germany actively supported this project in cooperation with MED-EL.

Appendix A

Examples of items used in the LittlEARS® Early Speech Production Questionnaire:

Question	Answer	Example
1. Does your child cry to attract attention?	□ yes □ no	n/a
5. Does your child cheer or laugh out loud?	□ yes □ no	While playing or taking a bath
9. Does your child produce vowels?	□ yes □ no	a, e, i, o ,u
12. Do the utterances of your child sound melodious?	□ yes □ no	n/a
13. Does your child produce consonants?	□ yes □ no	
13.1. If so, please circle the consonants your child produce.	m n b p d t l f j w r sch g k ng s	
14. Does your child produce individual syllables?	□ yes □ no	"bah", "mam", "dah", "dee"
24. Does your child produce two-word utterances?	□ yes □ no	"Mummy come.", "Ball here."

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