

Population-Based Screening of Children for Specific Speech and Language Impairment in Germany: A Systematic Review

Jürgen Kasper^{a,b} Julia Kreis^c Fülöp Scheibler^c Delia Möller^d Guido Skipka^c
Stefan Lange^c Olaf von dem Knesebeck^a

^aDepartment of Medical Sociology, University Medical Center Hamburg-Eppendorf, ^bUnit of Health Sciences and Education, University of Hamburg, Hamburg, ^cDepartment of Non-Drug Interventions, Institute for Quality and Efficiency in Health Care, Köln, and ^dVoice, Speech and Hearing Disorders, University Medical Center Hamburg-Eppendorf, Hamburg, Germany

Key Words

Speech and language delay · Specific language impairment · Primary language disorder · Preschool children · Screening · Systematic review

Abstract

Objective: The study was aimed at evaluating the effectiveness of a systematic population-based screening programme for specific language impairment (SLI) in preschool children in Germany. **Methods:** The study question was divided into a review of (1) evidence from studies evaluating screening programmes, (2) diagnostic instruments in the German language, and (3) studies evaluating speech and language interventions. A systematic database search was conducted between June and October 2007 and was updated in January and again in May 2008. Relevant studies were identified by 2 independent reviewers based on screened titles/abstracts and full texts. **Results:** 4,806 studies were screened. The only existing controlled screening study did not provide data for SLI. No diagnostic study met the inclusion criteria. Sixteen randomized intervention studies were included, 3 studies contributed to the appraisal of earlier

against later initiation of treatment. Most studies were of limited quality. We found indications of short-term positive effects from language therapies in children with SLI. Long-term outcomes were not investigated. No evidence supporting the advantage of earlier treatment initiation was identified. **Conclusions:** The benefit of population-based language screening of preschool children with SLI is not proven. Controlled screening studies are therefore necessary. For Germany, the accuracy of existing diagnostic instruments has not yet been sufficiently examined.

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Introduction

Acquisition of speech and language is a core process in the general development of the child. Therefore, disordered speech or language is likely to indicate a risk of a wide range of serious secondary problems during school age. This applies for instance to school performance, to reading or writing and to social and emotional quality of life. Depending on a child's age and developmental stage, the natural course of speech and language disorders im-

plicates a variety of speech- and language-related symptoms on different linguistic levels, such as an insufficient active or passive lexicon or the inability to perceive or pronounce specific phonemes. One of the earliest symptoms of the disorder is a delayed start of speech. However, late talking may also appear within the range of a normal development. Children who show significant language delay but later catch up with their peers are known in the literature as late bloomers [1, 2]. It is not clear whether those children are more at risk of language disorder than earlier developing children [3]. Diagnostically, there has to be a division between language impairment and specific language impairment (SLI) depending on whether or not the language delay appears isolated or accompanied by other developmental delay. Beyond a differential diagnosis of hearing problems, a diagnosis of SLI therefore requires the exclusion of an involvement of cognitive, motor or emotional development delays, which is difficult to diagnose at an early age.

The prevalence of language development disorder has been controversially reported. Depending on the measures and thresholds used for diagnosis, international estimations of speech and language disorders in general range from 0.65 [4] to 19% [5]. Based on the screening of more than 10,000 three-year-old children, a study in the Netherlands found a proportion of 2.4–5.3%. Data on SLI are rare. The only 2 studies explicitly reporting SLI rates found 7.4 [6] and 0.6% [7]. The latter value, however, refers to the subgroup of expressive SLI only. Likewise, the results from German studies also vary widely [8, 9]. Beyond a reliable basis for estimating the real rate of SLI, these data represent the expression of an obvious difficulty in agreeing on a diagnostic procedure, on a classification or even on a clear disease construct. A systematic review of prevalence studies by Law et al. [10] recommends avoiding the emphasis of any proxy or the averaging of existing findings. Moreover, the diversity of prevalence data indicates the need to move from the prevalence approach to a model of risk. This would involve the identification of key risk factors that are partly amenable. However, there can be no doubt about the fact that developmental speech and language disorders rank among the most frequent disorders of child development.

Few data exist on the present quality and frequency of speech and language screening in Germany. The German health system provides an opportunity to screen for developmental delay during a prevention programme conducted by paediatricians. Approximately 90% of children take part in this programme. However, language screening is not standardized in any way yet. Although there are

initiatives for language screening by the educational institutions in some of the 16 German states (*Länder*), these focus on school readiness rather than on a medical diagnosis. It can be assumed that screening procedures with proven accuracy and validity are not systematically used in any German region.

The international literature [11] suggests that children can benefit from different kinds of treatment. This holds good for children with mainly expressive symptoms more than for those with the receptive type of the disorder, for younger and older preschoolers and referring to several linguistic levels. However, evidence to justify a population-based screening programme furthermore would have to show the preponderance of a potential, additional benefit from earlier treatment initiation over potential side effects of the screening.

Our study was aimed at evaluating the evidence for the effectiveness of a systematic population-based screening programme for SLI in preschool children. This research question was processed by a group of experts on speech and language development and on systematic review methodology at the request of the German Institute for Quality and Efficiency in Health Care (IQWiG), which itself was commissioned by the Federal Joint Committee (G-BA) of the German Statutory Health Care System [12]. Evidence from existing systematic reviews such as those by Nelson et al. [13], Law et al. [10] and the National Health and Medical Research Council [14] was considered insufficient to appraise the potential benefit of screening for its implementation in the German health care system. It would have had to be proven that new studies in the field had contributed considerably to the debate in the meantime. Moreover, as existing evidence is based on international populations and diagnostic methods, it holds for English-speaking children only.

Methods

The research question predominantly implied the review of existing controlled trials, evaluating screening compared to no screening. However, if those were not identified as having sufficient explanatory power, the review had to link 2 other sources of relevant evidence [15, 16]: scientific proof of both the sufficient diagnostic quality of existing German screening procedures and the advantage of earlier compared to later initiation of treatment of SLI. Conclusions drawn from linking these 2 bodies of evidence are limited to the degree of comparability of the populations investigated in the 2 sources with each other and with the population on which the research question is focused [15].

A systematic search was conducted in bibliographic databases (AMED, BIOSIS, CCMed, CINAHL, Cochrane Central Register

of Controlled Trials CENTRAL, EMBASE including EMBASE Alert, ERIC, Heclinnet, Journals@Ovid, MEDLINE, PsycINFO, Psyn dex, SciSearch, Social SciSearch) and publisher databases (Hogrefe, Karger, Kluwer, Krause & Pachernegg, Springer, Thieme). Moreover, the search strategies included the tracking of references published in existing systematic reviews, HTA (health technology assessment) reports and meta-analyses [2, 11, 13, 14, 17–22], the consultation of German experts and searches by hand in journals. The first comprehensive search was conducted between June and October 2007 and was updated in January and again in May 2008.

The studies identified by the abovementioned strategies passed through 2 rounds of assessment by 2 independent working investigators (J. Kasper, D.M.). Using titles and abstracts, the studies were screened for potential relevance regarding 1 of the 3 questions. Studies that could not be excluded with certainty were then examined in detail in full text. In cases of doubt a third investigator was consulted (F.S.). Studies meeting the inclusion criteria were assessed concerning their quality and potential biases, and data were extracted. These steps were conducted by the investigator (J. Kasper or D.M.) and reviewed twice (J. Kreis and G.S.).

To be included in the pool of studies for question 1, studies had to evaluate a screening procedure in a control group design using an unselected population. Studies contributing to question 2 had to outline accuracy data on the detection of SLI within a sample from the general population. This does not assume a language test or screening instrument could distinguish SLI from other language impairments. By use of subsequent diagnostic steps, it is nevertheless possible to determine a detection rate for SLI referring to the first test. There had to be sufficient data to generate 4-field tables, including screening positives and screening negatives and true positives as well as true negatives. For inclusion of those studies, the suitability of the reference standard was an important criterion. A reference standard was considered suitable when it was validated and not less comprehensive than the screening instrument it was compared to. Therapy studies (question 3) were included either if they evaluated a treatment method for SLI by employing a randomized controlled design with a non-treated SLI group as control, or if they allowed for comparison of effects achieved by treatment in more than 1 age group. Data were extracted from all studies meeting the inclusion criteria and summarized descriptively. All studies were rated for risk of bias, based on methods of randomization and allocation concealment, blinding, sample size calculation, comparability of groups, patient flow and intention-to-treat strategy. The study protocol was put up for public discussion. Concerns and recommendations by German experts were thoroughly scrutinized and incorporated.

Results

Screening

Out of 2,198 references, 2,031 were excluded on the basis of their title and abstract. To the remaining 167 studies another 50 studies were added, following recommendations by German experts commenting on the protocol as mentioned above. Two publications out of these

217 searched by full text were included in the review [23, 24], although they did not explicitly report results for children with SLI (fig. 1). Both referred to the same study (Rotterdam study). However, we considered this evaluation study of population screening important as an example of the kind of study that would principally allow for an appraisal of the study question.

The study was aimed at evaluating a standardized screening procedure in a particular region of the Netherlands by a controlled study. According to experience, 85% of all children participate in regular paediatric screening in Dutch child health centres. By cluster randomization, 55 of a total of 91 child health centres in the particular region were allocated to either screening by use of a standardized instrument (VTO, vroegtijdige onderkenning van ontwikkelingsstoornissen) [25] or to screening practice as usual. Paediatricians in the intervention clusters were trained in conducting the language test, whereas physicians in the control group received neither training nor test manuals. A cohort of a total of 10,942 children between 15 and 24 months old was identified that would receive regular screening twice within a specific period of a little more than 2 years. As primary endpoints, type of school, speech and language performance as well as a prognostic judgement were assessed at 8 years of age. Although it was assumed that 85% of the children would attend the regular child screening, in the study only 55% of the children randomized into the screening group had speech and language screening. As the overall response rate was unfortunately low (19–47%, depending on the outcome measure), a bias cannot be excluded. The authors report a significant effect of a 1% difference (between 2.7 and 3.7%; $p = 0.032$, one-sided testing) in school attendance between screened and unscreened children. This effect was not significant by two-sided testing. No statistically significant differences were shown between the screening and control groups based on two-sided testing at the age of 8 years. The publication does not provide an intention-to-treat analysis to prohibit potential biases due to selective dropouts. However, although there was a subgroup of 15 children with SLI within the group of children with positive screening result, no results are outlined for this group regarding the endpoints of the study [24]. Therefore, it is not clear to what extent the study results apply to the children in the focus of this review.

Diagnostics

A pool of 1,302 references, including 1,269 from database searches and another 33 which were alluded to by

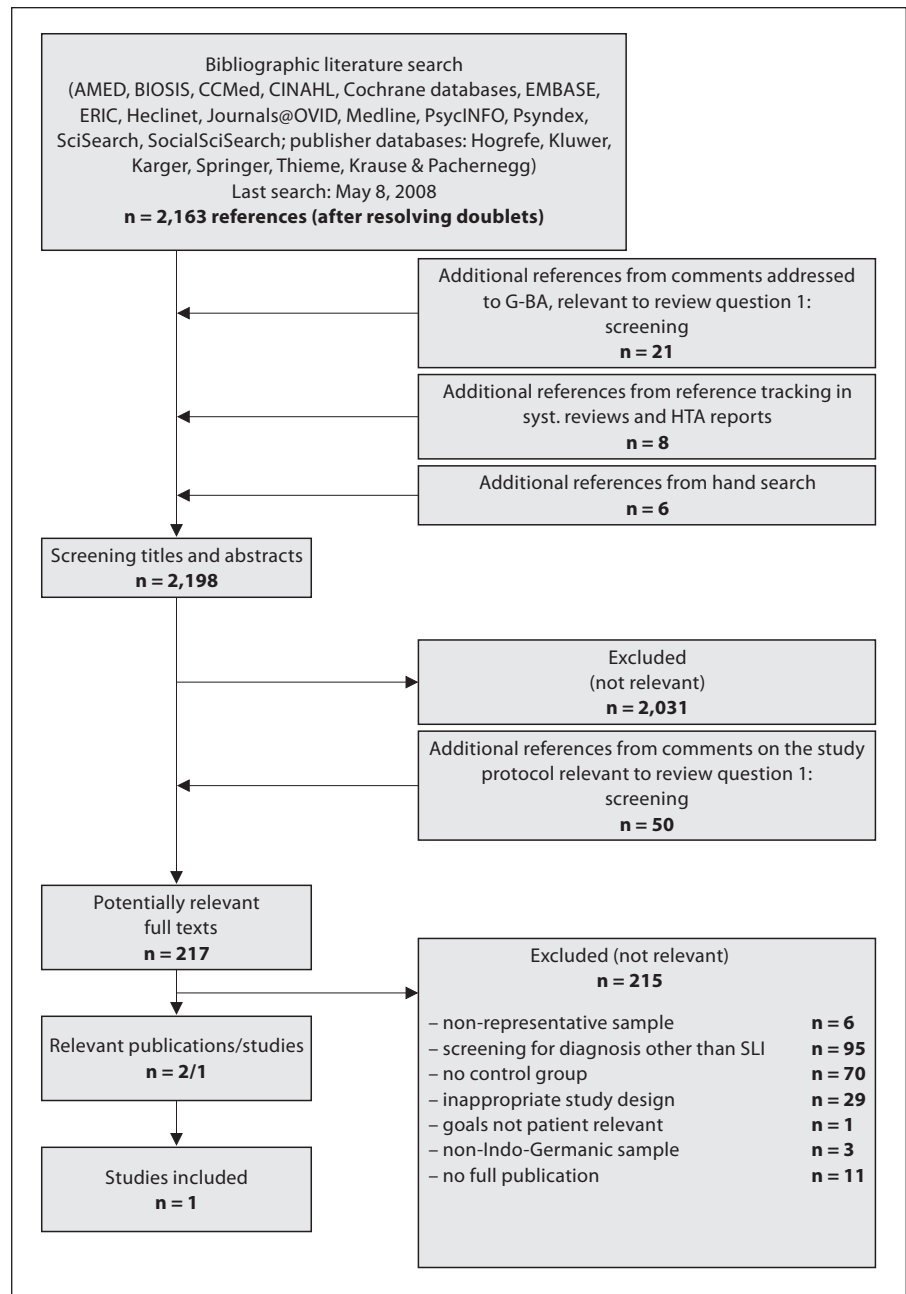


Fig. 1. Review question 1. Studies evaluating speech and language screening.

German experts or were found by reference tracking in systematic reviews, were screened based on titles and abstracts. As a result, 1,159 studies were excluded. The remaining 143 studies potentially relevant to the diagnostic review question were complemented by 79 full publications contributed by the experts (fig. 2).

None of these 222 studies screened in full text met the inclusion criteria. This means that the diagnostic quality

of the tests currently used in German-speaking countries to screen for specific speech and language development is not known. Nevertheless, since a variety of tests is in daily use by German speech and language experts, this result might need to be explained in more detail. Twenty-five publications (and 1 additional unpublished manuscript: Neumann, 2008) of diagnostic studies on 17 speech and language tests met the criterion of using any reference test

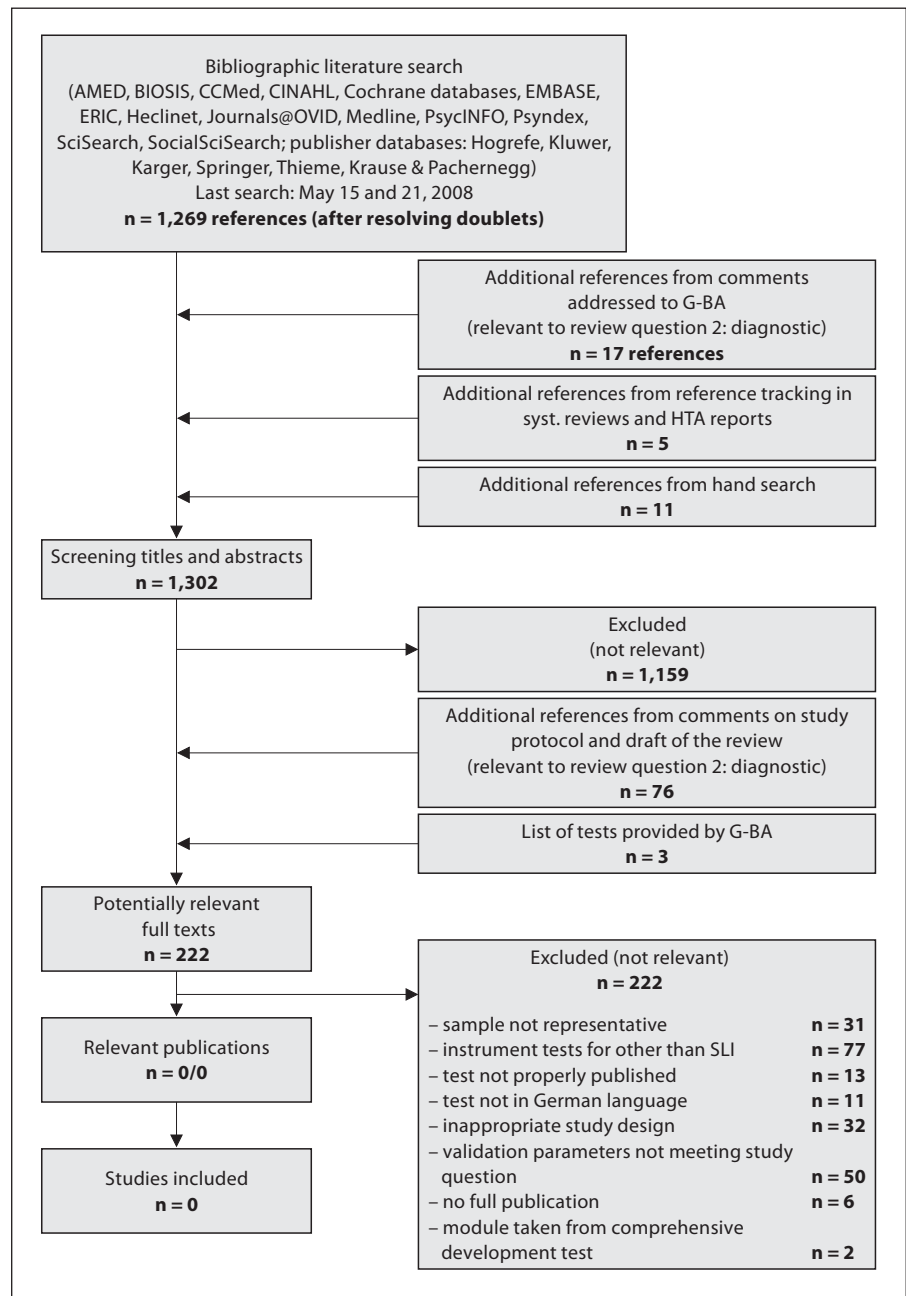


Fig. 2. Review question 2. Studies validating screening test in German language.

for proof of accuracy [1, 26–49]. Some of the reference tests used could be challenged regarding their suitability for this purpose, e.g. judgements of kindergarten teachers or results of tests that themselves are screening tests rather than comprehensive speech and language tests. Nevertheless, the difficult decision on the suitability of the reference test was superfluous since, in case of doubt, other criteria were always breached (table 1). Most of the studies

reported correlations to describe the extent of convergence with the sum score or with subscales of the reference test. This kind of data lacks information about the natural frequency of agreement between 2 tests and does not suffice to appraise test quality. Complete 4-field tables were reported for 5 tests [*Elternfragebögen für die Früherkennung von Risikokindern* (ELFRA-1, ELFRA-2); *Kindersprachscreening* (KiSS); *Sprachbeurteilung durch Eltern* –

Table 1. Methodological basic characteristics of diagnostic studies

Index test	Reference test (specific index test)	Method of validation	4-Field table provided or prepared	Accuracy of SLI detection indicated	Reference No.
AWST	kindergarten teachers' judgement	correlation	none	no	26
	complete sentences and complete words subtests from PET (Psycholinguistic Development Test) (Angermaier 1977)	correlation	none	no	
	test battery not further explained	multiple correlation	none	no	
BUEVA	WET (Vienna Development Test) (Kastner-Koller and Deimann 1998)	correlation	none	no	29
	Potsdam education questionnaire (Esser and Wyschkon 2000)	correlation	none	no	
	school years (at 8 years of age)	correlation	none	no	
ELAN	MFED 2-3 (Munich Functional Development Scales) (Köhler and Egelkraut 1994)	correlation	none	no	30
	expressive lexicon subscale of ELAN/16-26 months; AWST (Active Vocabulary Test 3-6) (Kiese-Himmel 2005)	correlation	none	no	
ELFRA-1	SETK-2 (language development test) (Grimm 2000)	correlation	none	no	31
	SETK-2 (Grimm 2000)	correlation	none	no	32
	ELFRA-2 (parent questionnaire for the early recognition of children at risk) (Grimm 2006)	sensitivity + specificity ^a	available	no	33, 49
ELFRA-2	SETK-2 and 3-5 (Grimm 2000)	correlation	none	no	31
	subscales of the BSID II (Bayley Scales of Infant Development) (Bayley 1993)	correlation	none	no	
	SETK-2 and 3-5 (Grimm 2000)	sensitivity + specificity ^a	incomplete	yes ^a	27
	subscales of the SETK 3-5 (Grimm 2001)	correlation	none	no	32
	ELFRA-3, adaptation of ELFRA-2 for 3-year-olds	sensitivity + specificity	available	no	34
	ELFRA-3	sensitivity + specificity ^a	available	no	49
	SETK-2 (Grimm 2000)	correlation	none	no	
	SETK-2 (Grimm 2000)	sensitivity + specificity ^a	incomplete	yes	1
	SETK-2 (Grimm 2000)	sensitivity + specificity ^a	incomplete	yes	35
	RDLS-III (Reynell Developmental Language Scale) (Gibbard 1994)	sensitivity + specificity ^a	incomplete	yes	
	SETK-2 (Grimm 2000)	sensitivity + specificity ^a	available	no	37
	ELFRA-3	sensitivity + specificity ^a	available	no	36
Questionnaires on language development	(draft of questionnaire vs.) lexicon and MLU from language sample	correlation	no	no	38
FRAKIS/ FRAKIS-K	type and frequency from language sample (FRAKIS: lexicon; questionnaire on early childhood development)	correlation	no	no	39
	MLU from language sample (FRAKIS: morphology of flexions, sentence complexity)	correlation	none	no	
	FRAKIS lexicon (FRAKIS-K)	correlation	none	no	
Screening of developmental disorders in 3-year-olds, language	development test for toddlers (Bühler and Hetzer 1966)	correlation	none	no	40
	self-developed questionnaire for parents and kindergarten teachers addressing stage of language development (no reference)	correlation	none	no	
	school marks in year 5	regression	none	no	

Table 1 (continued)

Index test	Reference test (specific index test)	Method of validation	4-Field table provided or prepared	Accuracy of SLI detection indicated	Reference No.
HASE	first to third level: school marks in German, reading, spelling, reading speed, reading comprehension	correlation	incomplete	no	27
KiSS	<i>Majority judgement within an expert panel; base rate free</i>	<i>sensitivity + specificity^b</i>	<i>yes</i>	<i>deducible^c</i>	28
	<i>Majority judgement within an expert panel; base rate fixed</i>	<i>sensitivity + specificity^b</i>	<i>yes</i>	<i>deducible^c</i>	
KISTE	LSV (Landau Language Development Test) (Götte 1976)	correlation	none	no	41
	judgement by kindergarten teachers/investigator/education questionnaire (no reference)	correlation	none	no	42
	HSET (Heidelberg Language Development Test) (Grimm and Schöler 1991)	correlation	none	no	
	LSV for preschoolers (Götte 1976)	correlation	none	no	
LSV	FTF-W (Frankfurt Test for Five-Year-Olds): lexicon (Raatz 1971)	correlation	none	no	43
	form creation subtest of the LSV; grammar subtest of the PET (Angermaier 1977)	correlation	none	no	
	ratings of communication by students (no reference)	descriptive	none	no	
MSVK	WST (lexicon test) for school beginners (Kamratowski and Kamratowski 1969)	correlation with sum score	none	no	44
	AWST (Active Vocabulary Test) (Kiese-Himmel 2005)	correlation with sum score	none	no	
	HSET (Grimm and Schöler 1991)	correlation with sum score	none	no	
	teacher judgement of language comprehension	correlation with sum score	none	no	
SBE-2-KT	ELFRA-2 (Grimm 2006)	sensitivity + specificity	available	no	45
SEV	judgement of speech therapy students	specificity ^d	none	no	46
SSV	SETK 3–5 (Grimm 2000); SSV for 3-year-old children	sensitivity + specificity	available	no	47
Teddy Test	kindergarten teacher questionnaire (no reference)	correlation	none	no	49
	Binet-Simon-Kramer Test (no reference)	correlation	none	no	
	learning test for preschoolers (no reference)	correlation	none	no	
	HAWI-VA (Hamburg-Wechsler Intelligence Test) (no reference)	correlation	none	no	

References to reference tests are in parentheses (round brackets) and are to be found in the reference studies listed in the right column (by reference No.). The table lists all 17 index tests validated by use of any reference test (form or scale of index test); text in italics refers to the unpublished status of the manuscript sent by the author of the KiSS. Expansions of study abbreviations are either provided in English equivalents or can be found in the reference studies. MLU = Mean length of utterances.

^a Values provided by the publication did not refer to representative proportions of screening positives and screening negatives. Thus, the data neither allow for a comparison with other tests nor for generalization. ^b Due to a rate of 23.5% of SLI, this reference test is debatable. ^c Due to cases of comorbidity in the screening positives, values are not reliable. ^d Reference test only for screening positives.

Kurztest (SBE-2-KT); *Sprachscreening für das Vorschulalter* (SSV)], 3 of them with 4-field tables based on representative proportions of screening positives and screening negatives. According to the detection of SLI, 4-field tables were presented for 2 of these tests (ELFRA-2, KiSS). Detection rates of tests for the wider diagnosis of language impairment cannot be assumed for SLI as a matter of course. Some others reported values for sensitivity and for specificity of the ELFRA-2 compared with the SETK-2 (*Sprachentwicklungstest für zweijährige Kinder*), ELFRA-3 and RDLS-III (Reynell Developmental Language Scale) [1, 34–37, 49]. These values resulted from applying the reference standard to all screening-positive children and to a selective sample of children who were screened negative. Since biases due to this procedure are likely and cannot be controlled, the reported values cannot be seen as representative either for the original sample or for the general population [50, 51]. An unpublished manuscript (Neumann, 2008) presented validation results for the KiSS test. Since the combination of 4 language tests (RDLS-III, *Psycholinguistische Analyse kindlicher Sprechstörungen*, *Patholinguistische Diagnostik bei Sprachentwicklungsstörungen* and AWST, *Aktiver Wortschatztest*, for 3- to 5-year-old children) was found by the authors of this study to be unsuitable as a reference criterion, the KiSS was compared to an expert rating. However, as the authors admit, this reference was still in a premature developmental stage. For instance, the raters were given a baseline rate for positive screening before judgement. Moreover, the targeted diagnosis differed from SLI. Auditory perception and processing disorders were excluded, comorbidity was included. Therefore, data provided by the 4-field tables cannot be assumed to be valid for SLI. This means that no test could be included in the review.

Therapy

From a pool of 1,232 references including 1,172 from database searches and another 60 recommended by German experts found by reference tracking, 1,025 studies were definitely excluded on the basis of titles and abstracts. The remaining 207 potentially relevant studies were complemented by 55 publications contributed by German experts and were screened in full text. Of these, 203 had to be excluded (65 had non-representative samples, 72 pursued research questions different to ours, 53 studies were of unsuitable design, 4 used outcomes rated as not patient relevant, 2 studies did not contain any additional information, and for 7 studies no full publications were available). In all, 59 publications referring to 57 studies were identified as being relevant to answer re-

view question 3 [52–110] as they investigated the efficacy of any speech and language interventions (S+L interventions) in children with SLI; 33 of them were designed as randomized controlled trials [52–66, 69–71, 73, 75–77, 79, 81, 82, 87, 88, 90, 91, 94, 99, 101–103, 107], 24 built study groups using other strategies. Most of the studies (41) tested an intervention group against an active control group. Trials using a waiting group or sham intervention design were reported in 16 studies [52–66, 69–71]. Although less rigorous criteria were applied to the search of studies investigating the moderating influence of age on the efficacy of speech or language intervention, only 3 such studies could be identified (fig. 3) [52, 67, 68].

Trials comparing 2 treatment methods without a no-treatment study arm cannot be used for appraisal of the general efficacy of interventions. The comparison of 2 active groups provides relative efficacy information only and is therefore not suitable to make conclusions regarding efficacy in an absolute sense. Therefore, out of the pool of 57 studies, we selected the 16 using a sham intervention or waiting control group design to appraise the efficacy of S+L interventions for SLI.

The potential bias was rated as low in 2 of the 16 studies [56, 60]. Fourteen studies revealed important deviations from guidelines on the execution and reporting of randomized controlled trials [111]. Only 2 studies described appropriate randomization and concealment strategies [52, 62]. A sample size calculation was reported in 3 studies [52, 53, 61]. Out of 7 studies with a high number of dropouts, or no reported dropouts, only 1 conducted an intention-to-treat analysis [52]. In most of the studies the primary outcome was not clear.

With the exception of 2 studies [54, 63], all interventions evaluated in the 16 publications were conducted on English-speaking samples. One study reported results of S+L interventions provided to German children [54], another one to Chilean children [63]. The interventions addressed children between mean ages of 25 months [65, 53] and 13 years [56]. The study sample size ranged from 14 to 216 children. However, most of the studies included 20–30 children in total. All children had, as a minimum, problems concerning expressive language. Some studies employed more rigorous inclusion criteria such as a cutoff for the lexicon. Interventions were group therapies or single therapies at varying frequency and length over a period of 3 weeks to 12 months. With regard to the therapeutic approach and the intervention goal, the studies also differed widely. Some conducted broadly designed intervention programmes, for instance the Hanen Program for Parents [60, 61], while

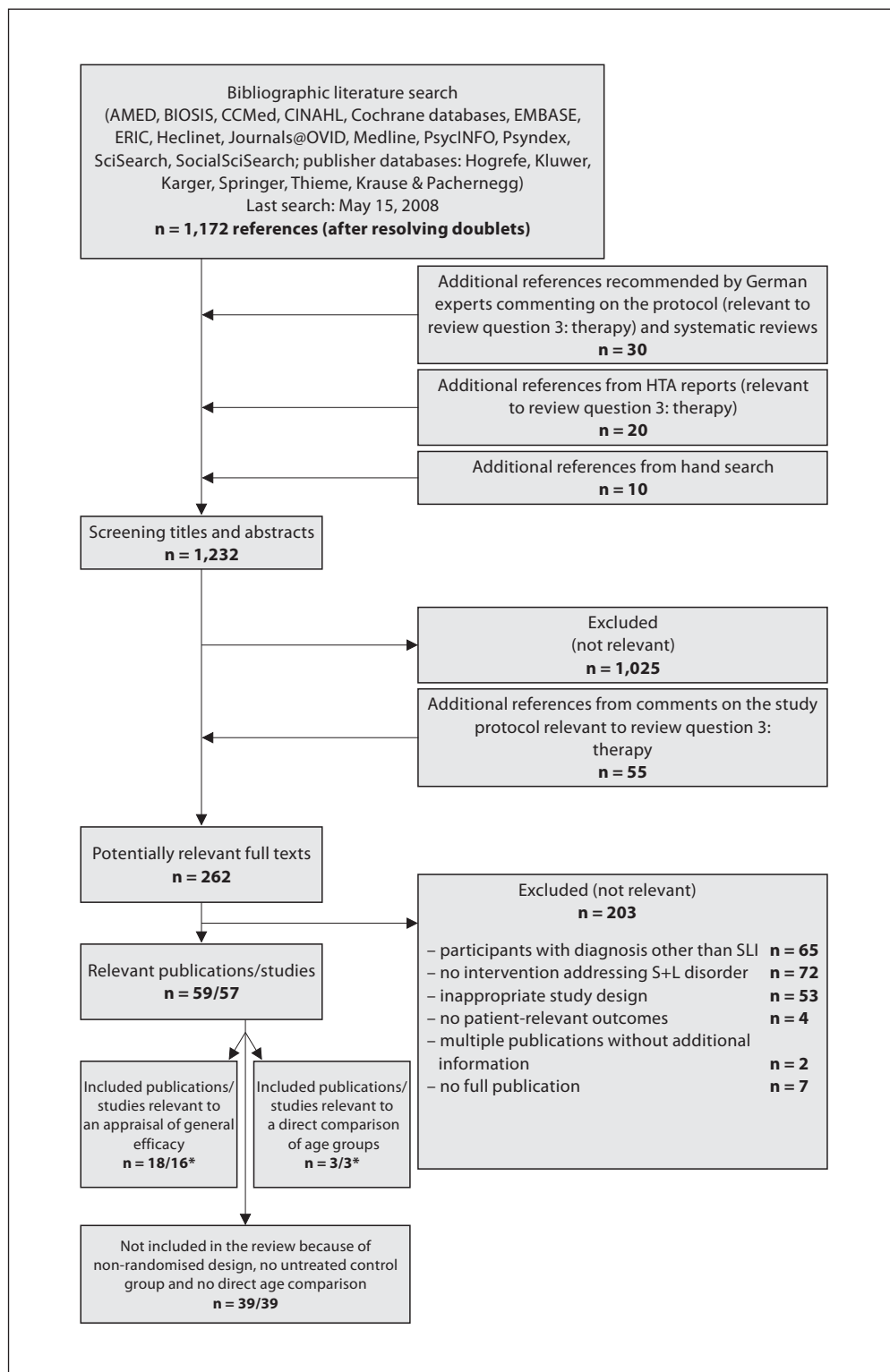


Fig. 3. Review question 3. Studies evaluating S+L interventions for children with SLI. * One study was relevant for both the question for general efficacy and the question for direct comparison of age.

Table 2. Effects of intervention studies

Linguistic level	Construct addressed	Operationalization	Reference No.	Effect	p (2-sided)	
Morphology and syntax	language complexity	spontaneous language sample: mean length of utterances	52	–	0.337	
			57	/	<0.001	
			65	/	0.003	
		mothers' report: mean length of utterances	71	–	>0.38	
			57	/	<0.001	
			57	/	<0.001	
		spontaneous language sample: language complexity	61	–	[<0.05 ^a]	
			57	/	<0.001	
		Derbyshire Language Scheme Picture Test	61	/	[<0.01 ^b]	
			64	/	<0.001	
		number of linguistic markers	53	/	0.001 ^c	
				/	0.013 ^c	
		grammar	SETK-2: sentence production		/ /	0.017/0.021 ^{c, d}
			SETK 3–5: encoding of semantic relations		–	0.376 ^c
		morphology	ELFRA-2: morphology		/ /	<0.001/0.062 ^{c, d}
SETK 3–5: learning of morphological rules			–			
syntax	ELFRA-2: syntax		/ /	<0.001/0.062 ^{c, d}		
questions	number of correct questions	70	/	<0.001		
verb argument structure	video-based test	56	/	0.007 ^e		
Semantics and lexicon	lexicon	language sample: different words	61	/	[<0.01] ^b	
			65	/	<0.001	
			64	/	<0.001	
		number of different words in a game instruction	65	/	<0.001	
			60	–	n.s.	
			61	/	[<0.02] ^b	
		language sample: total number of words	65	/	<0.001	
			53	/ /	0.016 ^c /0.018 ^{c, d}	
		ELFRA-2: expressive lexicon	SETK-2: word production		/	0.016 ^c
			SETK-2: comprehension I: words		–	[0.057]
		Derbyshire Language Scheme Picture Test	57	/	<0.001	
			57	/	<0.001	
		parents' report: lexicon	60	/	[≤0.02] ^b	
			61	/	[<0.01] ^b	
		specific aspects of lexicon	language sample: different target words	60	–	n.s.
61	/			[<0.01] ^b		
language sample: different control words	60		–	n.s.		
	61		/	[<0.01] ^b		
language sample: frequency of target words	61		/	[<0.02] ^b		
language sample: frequency of control words	61	–	[<0.05] ^a			
Phonetics and phonology	consonant production	language sample: percentage of consonants correct	52	/	0.001	
			55	–	0.09	
			61	–	n.g.	
		language sample: inventory/position of consonants (2 operationalizations)	61	early: /	[<0.05] ^b	
				medium: /	[<0.05] ^b	
		late: –	[<0.05] ^a			
		initial: /	[<0.05] ^b			
		final: –	[<0.05] ^a			
	articulation	Goldman-Fristoe Test of Articulation	52	/	0.001	
		rating of phonetic inventory	69	/	<0.01	
	phonological awareness	Phonological Abilities Test	55	/	<0.01	
		Comprehensive Test of Phonological Processing (blending words)	58	–	n.g.	
	phoneme repertoire	language sample: number of vocalizations	61	–	n.g.	
	syllable structure	language sample: 3 complexity levels	61	level 1: –	n.s.	
				level 2: –	n.s.	
			level 3: /	[<0.01] ^b		

Table 2 (continued)

Linguistic level	Construct addressed	Operationalization	Reference No.	Effect	p (2-sided)
	phoneme production	phonological test: production level, pronunciation sample, centroid	66	/	0.05
	phoneme identification	identification test	66	/	0.02
	phonological development	Assessment of Phonological Processes – Revised	52	/	0.003
		phonological error rate	62	-	0.26 ^f
Pragmatic-communicative	understandability	language sample: number of understandable utterances	57	/	0.008
		language sample: number of utterances	61	-	n.g.
		language sample: words per minute	61	-	n.g.
		total number of words	64	/	<0.001
	relevance of utterances comprehension	frequency of references to actions typical of playing	64	/	<0.001
		language sample: proportion of understandable utterances	71	-	>0.38
		65	/	<0.001	
More complex constructs	narrative competence	discourse analysis	63	form: / content: /	<0.001 ^g 0.005 ^g
	general language development	RDLS	57	P: / V: /	<0.001 <0.001
			57	I: / G: /	<0.001 <0.001
		Comprehensive Assessment of Spoken Language Bristol Language Development Scales	58	-	n.s.
			62	-	0.73 ^d
		Preschool Language Scale	62	P: - V: /	0.44 ^f 0.025 ^f
			62	-	0.44 ^f
	impairment disability	62	-	0.56 ^f	
		62	-	0.56 ^f	
	sentence comprehension	Token Test for Children	58	-	n.g.
		SETK-2: comprehension II: sentences	53	-	[0.323]
		SETK 3-5: understanding sentences	-	-	[0.113]
	expressive speech competence	SETK 3-5: both subtests ≥40	-	/	0.028 ^g
		SETK 3-5: min. 1 subtest ≤35	-	-	0.105 ^g
	literary language	Wechsler – objective reading dimension	55	R: - S: -	n.s. n.s.
		Non-Word Decoding Test	55	/	<0.05
individual measure for improvement	compensation/non-compensation of individual study inclusion criterion	62	-	0.46 ^d	
	compensation/non-compensation of any study inclusion criterion	62	/	0.036 ^d	

I = Content; G = grammar; R = reading; n.g. = not mentioned; n.s. = not statistically significant; P = language production; S = writing; V = language comprehension; CDI = Communicative Development Inventory.

^a One-sided p; not significant by two-sided testing (own calculation). ^b One-sided p; also significant by two-sided testing (own calculation). ^c Declared as p from one-sided testing in the publication [53], following own calculation referring to two-sided testing.

^d Group difference at 6/12 months of follow-up. ^e Difference between all 3 groups (2 intervention groups, 1 control group) at 6/12 months of follow-up. ^f Aggregated data analysis for 6 and 12 months of follow-up. ^g Own calculation.

others focused on more specific language competences such as particular aspects of sound production or speech perception [64].

The 16 studies addressed outcomes referring to all linguistic levels: morphosyntactic, semantic and lexicon, phonetic-phonological, and pragmatic-communicative (table 2).

Effects of S+L Interventions Provided to Children with SLI on Health-Related Quality of Life

One study [62] addressed the effect of speech and language therapy on health-related quality of life using the 'handicap' scale of the Therapy Outcome Measures tool. No effects were shown at any measurement point between intervention and control groups.

Speech- and Language-Related Effects of S+L Interventions Provided to Children with SLI

Goal achievement concerning the field of speech and language outcomes was assessed by use of spontaneous speech samples (to evaluate the mean length of utterances, lexicon or percentage of consonants correct), by parent reports and by validated test instruments. Most of the studies employed multiple measurements, some using various operationalizations for the same parameter, such as assessing lexicon directly by a language sample and by asking the parents.

Results for the Morphosyntactic Level

Six studies addressed complexity of language, some of them by using more than one operationalization [52, 57, 61, 64, 65, 71]; 3 out of 6 showed positive effects of the intervention. Three studies addressed aspects of grammatical, syntactic and morphological ability [53, 56, 70], all of them showing positive results.

Results for the Semantic Level and Lexicon

Lexicon was assessed by 6 studies [53, 57, 60, 61, 64, 65], most of them using multiple measurements. Four studies showed clearly positive results, 2 studies showed mixed results. Girolametto et al. [59–61] investigated the use of specific words, yielding mixed results.

Results for the Phonetic-Phonological Level

Seven studies addressed parameters referring to the phonetic and phonological level, such as the percentage of correctly pronounced consonants, articulation, and either receptive or expressive phonological abilities or comprehensive phonological assessment [51, 55, 58, 61, 62, 66, 69]. Three studies showed positive, 2 studies showed mixed results. Two studies showed negative results.

Results for the Pragmatic-Communicative Level

Pragmatic measures of communication were addressed by 5 studies [57, 61, 64, 65, 71], such as the number or frequency of words within utterances [57] and the relevance or comprehensibility of utterances [71]. The results were ambiguous: 3 were positive, 2 negative.

Results for Comprehensive Language Assessments

Results for constructs referring to more than one of the abovementioned levels, such as comprehensive speech and language tests or discourse analysis [63], were reported by 6 authors. Two of them yielded positive results, 2 showed ambiguous results and 2 studies showed negative results [53, 55, 57, 58, 61, 64].

Psychosocial and Emotional Effects of S+L Interventions on Children with SLI

Three studies reported psychosocial effects of language interventions [59, 62, 64]. Using the Child Behaviour Checklist, Girolametto et al. [59] found a significant difference on the 'externalization' scale, but not on the 'internalization' scale. Control group children were described as more aggressive than children in the intervention group. However, this already applied to baseline measurement. On the Vineland Adaptive Behaviour Scales, an effect of increased social competency was found in one study [64] and not in another one [62].

Cognitive Effects of S+L Interventions on Children with SLI

Two studies [59, 62] assessed play behaviour, which was different from the control group in the study by Girolametto et al. [59] 2.5 months after the intervention, and did not differ between groups in the study by Glogowska et al. [62] 6 and 12 months after the intervention. However, no mechanism could be recognized by which the authors assumed the potential effect to be mediated. The attention level did not differ between groups in the study by Glogowska et al. [62].

Results for Earlier Compared to Later Initiation of S+L Treatment for Children with SLI

Beyond the general proof of efficacy of S+L interventions, the advantage of earlier over later initiation of such interventions is crucial for an appraisal of the benefit of screening. The related data source is quite limited [52, 67, 68]. Only one of these studies used a randomized control design [52]. In this trial an additional measurement point for both study groups was arranged after the waiting control group had received the intervention. However, the 4-month delay between the 2 groups seems too short, and a comparison of the results would have required another delay after treatment had been finished in both groups. Sommers et al. [67] investigated schoolchildren who had already passed the critical age period. Tschirner et al. [68] only provide a graphical subgroup analysis of age and therapy setting without any statistical analysis. Theoretically, indirect comparisons between age-heterogeneous study groups from different trials could be considered for appraisal as well. As a prerequisite, these pairs would have to be comparable regarding time and amount of therapy, severity of the disorder, and outcomes. This cannot be assumed for any pairs built within our pool of intervention studies. In summary, since none of the studies intended to investigate age effects, they were not designed accord-

ingly. Casual occurrence of study cohort differences regarding age turned out to be insufficient to appraise potential effects of earlier compared to later initiation of therapy.

Results for Side Effects

Side effects or undesired effects of S+L interventions such as labelling or damage of self-image were not addressed in any of the studies included in the review.

Discussion

The present study was aimed at appraising potential population-based screenings in Germany for SLI at pre-school age. Population screening would be recommendable if benefits of early detection were to outbalance any harm that inevitably arises. The position of the break-even point depends on the degree of advantage affected children can expect from earlier initiation of therapy on the one hand, and on the detection rates of true positives and true negatives on the other hand.

Our results show that neither by searching screening studies nor by linking the evidence from diagnostic and therapeutic studies a solid database can be identified to answer this question satisfactorily. The informative value of the only existing screening study for our particular research question is strongly limited by methodological weaknesses of the study [112] and by its focus on speech and language disorder in general. Furthermore, none of the available German tests reported comprehensive detection rates or could be recommended for screening. Regarding research question 3, the studies selected to appraise evidence of the efficacy of S+L interventions reported more positive than negative outcomes. However, most of the studies were only of weak quality. The justification of design-specific characteristics such as sample size, selection of particular diagnoses, and intervention strategy is not explicit in most of the studies. Since the interventions were evaluated in terms of small steps referring to particular speech and language phenomena, the relevance and sustainability of potential effects can be challenged. Long-term effects were not reported. This means that the review of our study pool hardly permits any generalizability of intervention effects. It is not clear which factors regarding characteristics of the disorder, the children or the intervention these effects refer to. The fact that the two largest and least biased studies [58, 62] showed no language-related effects implies even more cautious appraisal of the results.

Moreover, the review showed that hitherto no research has been published to answer the question on whether children with SLI would benefit from earlier initiation of a therapy. Potential side effects of screening, such as consequences of false-positive test results, were not addressed by any study.

Our search identified a couple of other systematic reviews studying similar research questions. A review of screening for speech and language disorder was intended to inform guideline development by the US Preventive Service Task Force [13, 22]. Another review focusing on the same question was conducted at the request of the National Health Service [2, 11]. According to our review, both studies concluded that, due to insufficient evidence regarding basic questions, population screening should not be recommended. This conclusion was reaffirmed by the short report of the National Screening Committee 2005 [113]. Although answering the same review question, the studies differ from ours in some detail. Due to the later appearance of the studies by de Koning et al. [23] and van Agt et al. [24] which were found by our search, neither Nelson et al. [13] nor Law et al. [10] could identify any controlled screening trials. No appraisal was made by the two reviews leading to statements about the German diagnostic instruments. However, both reviews investigated the efficacy of interventions addressing primary language disorder. Moreover, a Cochrane review focusing on this question was published by Law et al. [11] as well as a meta-analysis based on the same data [20]. Referring to the same time frame, the two other reviews did not identify an identical study pool. Due to a more rigorous application of inclusion criteria, some of the studies selected by Law et al. [10, 11, 20] and Nelson et al. [13] were excluded from our review. In particular, some studies in the Law and in the Nelson study pools were excluded from our review because of an unclear diagnosis of the children, or because a sham or waiting condition was missing. In contrast to our study, Law et al. [10, 11] included only the primary outcomes in their reviews. Notwithstanding our study, Nelson et al. [13] limited the study pool to those investigating children not older than 5 years and published in the English language. Both Law and Nelson conclude that S+L interventions are efficacious concerning language outcomes. Law et al. [10] differentiated between expressive phonological disorders and expressive syntactic and receptive types, of which only the first showed a clear benefit. With regard to the heterogeneity of the study pool, Nelson et al. [13] valued her findings of proof of efficacy as not generalizable. More-

over, they claimed missing evidence for a long-term efficacy of such therapies.

Regarding our search strategy, we consider it unlikely that relevant studies have been neglected. However, some unpublished studies were difficult to acquire. Finally, we were unable to obtain one unpublished thesis (Lancaster G: The effectiveness of parent administered input training for children with phonological disorders; MSc thesis, City University, London, 1991) identified by other systematic reviews. However, we do not assume this study would have changed our results.

As indicated by strongly diverging reports of prevalence [6, 7], the proceedings used for the diagnosis of speech and language disorders and SLI diverge. Since neither a gold standard nor a reference standard exists, study samples are selected by use of different instruments, cutoffs or expert judgements. Moreover, the distinction between primary and secondary language delay, as it is used in the international literature, implies a much less rigorous use of the diagnostic criteria compared to the definition of SLI by the ICD-10. Beyond the exclusion of evident causes of language delay such as hearing or psychiatric disorders, the ICD-10 requires the application of the discrepancy criterion, which is used to relate language delay to a child's overall development status. In a strict sense, none of the studies in the review study pool met the ICD-10 criteria for SLI. We included studies reporting a procedure for excluding hearing problems or cognitive deficits as possible reasons for language delay, although no attempts were made to apply the discrepancy criterion. This means that, as a concession to international research practice, we had to tolerate some diagnostic heterogeneity in our study pool. In a stricter sense, this limits the explanatory power of our results when applied to our target group, which was SLI.

Our results regarding the effectiveness of S+L interventions are presented on a descriptive level of analysis. They do not allow a clear estimation of the degree to which children can benefit from different kinds of S+L interventions. However, the outcomes represented in our study pool were quite heterogeneous. Moreover, outcomes appeared in varying forms of operationalization in the studies extracted, which would have made a quantitative comparison difficult. Furthermore, the studies within the pool were also diverse regarding their clinical background. A meta-analysis would therefore have been meaningless, observed differences within groups of analysis would have been difficult to attribute and genuine differences in effects would have been obscured [114, 115].

Since, on the whole, a sample size calculation was not outlined and primary endpoints were not explicit, a selec-

tive publication seems likely. Moreover, no study protocol of any of the studies in our pool was published in advance.

The study results imply a pronounced need for further research. This applies to each of the fields addressed by our study questions. Proof of efficacy of S+L interventions that can provide support for population screening has to consider the potential benefit of an earlier initiation of therapy. This requires a change to more sophisticated study designs to evaluate and compare effects of treatment of comparable disorders at earlier and later stages in the process of language development. Since a potential advantage of earlier treatment is not likely to appear and cannot be reasonably expected to be expressed in short-term outcomes as they are commonly used in speech and language evaluation trials, more sustainable and patient-relevant outcomes have to be addressed, such as, for instance, school performance. Moreover, the bigger the difference in age between two study groups, the more the law of equal treatment becomes a serious problem. On the other hand, an age-adjusted treatment limits the comparability of effects within groups. Another fundamental difficulty of studies addressing the benefit of earlier treatment consists in the comparability of study samples. Due to higher false-positive rates in samples screened at an earlier age, on average these children would be assumed to show more improvement just because of a higher percentage of spontaneous remission of speech delay. As becomes obvious, designing a reasonable intervention study for this particular purpose is not an easy task, and well-designed, controlled trials of screening programmes are necessary. However, our study shows that at present neither a screening instrument with known detection rates nor a reference standard for the validation of the latter exists in Germany. With regard to explanatory power and the extreme efforts to conduct a screening study, it is important to resolve the issue of a lack of diagnostic measurement before its initiation.

Conclusions

At present, the benefit of population-based language screening for preschool children with SLI is not proven. This does not exclude a potential benefit. However, the review shows that even prerequisites for a controlled study evaluating language screening are lacking. Research is needed in Germany to validate a diagnostic reference standard and, moreover, to validate a screening test.

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