

Intervention for mixed receptive–expressive language impairment: a review

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ABBREVIATIONS

RCT	Randomized controlled trial
RELI	Receptive–expressive language impairment
SES	Standardized effect size
SLI	Specific language impairment
SLT	Speech and language therapist

Studies indicate that language impairment that cannot be accounted for by factors such as below-average non-verbal ability, hearing impairment, behaviour or emotional problems, or neurological impairments affects some 6% of school-age children. Language impairment with a receptive language component is more resistant to intervention than specific expressive or phonological delays, and carries a greater risk of comorbid behavioural difficulties as well as adverse outcomes for language development and academic progress. This paper considers underlying explanations that may account for receptive–expressive language impairment. It also reviews evidence for the effectiveness of intervention from theory and recent systematic reviews, trials, and speech and language therapy practice.

Children with receptive–expressive language impairment (RELI), also referred to as ‘receptive language disorder’¹ or ‘mixed receptive–expressive disorder’,² form a subset of those with speech, language, and communication needs who commonly have problems understanding both spoken and written language. They have particular difficulties in comprehending vocabulary and grammar and inferring meaning and will also have problems with expressive language. Some may have difficulties in pragmatics, i.e. the use of language in social contexts.³

Population studies indicate that some 6% of 5-year-old children have significant difficulties in language functioning.^{4,5} However, variability is observed across studies in the stringency of criteria and the nature of the measures used to define language impairment.^{6,7} For example, composite scores are often used which do not distinguish between the child’s language production (expressive language skills) and understanding of language (receptive language skills). However, studies that do make this distinction indicate that 2 to 4% of 5-year-olds have RELI.^{5,8} A detailed study of referral rates in one primary care trust in the UK (providing local community health services based on 1100 referrals to speech and language therapists [SLTs] over a 15-month period⁹) would suggest a UK prevalence of 4.5%, which is at the higher end of these estimates. Caution is required, however, as local factors such as staffing and resources for intervention would have influenced referral patterns.

RELI is likely to have a marked long-term impact on the outcomes for language development,^{10–12} literacy,^{10,12,13} behaviour, and social development.^{12,14,15} There are links also

to mental health problems.^{16,17} Studies consistently reveal that RELI is a higher risk factor for adverse long-term outcomes than specific expressive language impairment (ELI), which highlights the importance of effective early intervention.

Intervention for RELI may be informed by an understanding of levels of explanation based upon relevant theory and probable underlying mechanisms.¹⁸ We shall consider some of these before going on to consider specific approaches to treatment.

LINGUISTIC EXPLANATIONS

In general, children with RELI have particular difficulties with morphosyntax, i.e. with word inflections and the grammatical rules governing them.^{19,20} Characteristically, they have delays in understanding and applying the rules governing the correct combination of the elements of words, such as endings that mark verb tenses (e.g. *-ed*), third-person singular verbs (e.g. *I think*, *he thinks*) and plurals (e.g. *-s*), auxiliary verbs that denote tenses (e.g. *was* running, *is* running), and with determiners (*the*, *a*). Children with RELI also have deficits in correctly inferring meaning from what is said to them^{21,22} and in formulating questions.²³

Linguists have proposed theoretical explanations for these characteristic problems. One view is that such problems are specific to language, for example to linguistic modules of grammar/syntax^{24–26} or pragmatic competencies²⁷ and that they can be explained without reference to other, more general aspects of cognition. However, critics have expressed concern about the extent to which linguistic theory can provide explanations of language impairment.²⁸ Cross-linguistic studies

have also cast further doubt on the extent to which some of the explanations can be generalized to languages other than English.²⁹

COGNITIVE PROCESSING EXPLANATIONS

Children with RELI may experience difficulties in managing the cognitive functions of storage and processing where they have to complete two cognitive operations under time pressure. They also find it hard to learn new words or morphemes when processing demands are high. Linguistic explanations cannot readily account for these particular problems. This has given rise to the view that limited general processing capacity underpins the difficulties.^{30–32}

Children with RELI are also more likely to have slower reaction times across a wide range of verbal and non-verbal tasks than children with expressive problems only, and both groups have slower reaction times than typically developing peers, giving rise to a related view that their difficulties are due to a slower rate of cognitive processing.^{18,33}

Attempts have also been made to locate the underlying problems that many children with RELI experience in retaining verbal and non-verbal information at the level of working memory,³⁴ both in terms of phonological working memory deficits^{35–37} and executive functions, such as response inhibition.³⁸

Finally, children with RELI are commonly observed to have auditory processing deficits, both at the level of frequency discrimination^{39–41} and in rapid auditory processing.^{40–42} Tallal et al. refer to the latter perceptual processing deficits as ‘temporal processing difficulties’, which is in line with their hypothesis that basic temporal integration processes are important for the neural representations for units of speech and processing non-verbal tones. Within this account, children with RELI are held to process auditory information at a slower rate than their typically developing peers. They are thus disadvantaged when discriminating, sequencing, and remembering dynamic temporally cued components that are brief in duration or rapid in succession, such as speech formant transitions (e.g. separated by short inter-stimulus intervals, usually in the range of tens of milliseconds), and they require longer processing times than typically developing peers.⁴²

GENETIC FACTORS

Studies of twins indicate that genetic influences play an important part in RELI as well as in disorders of language acquisition in general.^{43–45} Genome scan studies specifically looking for linkage to specific language impairment (SLI) have either used categorical phenotypes, such as whether an individual had a diagnosis of speech and language impairment,⁴⁶ or have used quantitative measures of language ability.⁴⁷ The only linkage study that has specifically included families of probands with a strict phenotype of RELI (‘receptive language disorder’ according to research criteria of the International Classification of Diseases, 10th Revision⁵) replicated linkage to 16q and 19q.⁴⁸ This linkage was seen with the non-word repetition test as a measure of phonological working memory and not with the expressive language score as had been seen in

What this paper adds

- Receptive–expressive language impairment persists over time.
- There is a dearth of evidence from systematic reviews and randomized controlled trials for approaches to effective treatment.
- Expressive language interventions in children show promise and should be further investigated by phase II exploratory trials.

the SLI Consortium, 2002 genome scan study. Non-word repetition also gives the best discrimination between parents of affected children and non-affected families.⁴⁹ However, the relationships between the phenotypic markers, the genotype, and the clinical condition of SLI are complex. Although non-word repetition deficits can behave as a marker in individuals whose earlier language difficulties resolved and be present among wider family members affected by RELI,⁴⁵ they are not in themselves sufficient to give rise to SLI. Five-year-olds who have weak phonological working memory skills can also be found in typical populations without having SLI.⁵⁰ Thus there appear to be additive risk factors such as syntactic deficits and/or auditory temporal deficits; the evidence suggests that the former are heritable and the latter environmentally determined.⁴³

COMMENTS

One feature common to these competing theoretical accounts is that in general they have arisen from the study of within-child variables in experimental cohorts varying in selection criteria¹⁸ with little in the way of exploration of proximal and distal external variables. There may also be overlap between the accounts. For example, RELI may have a high heritability because it represents the most severe form of SLI and has a genotype that has an impact on correlated cognitive processes that mediate linguistic processing.

INTERVENTIONS FOR RELI

Evidence from systematic reviews

Recent systematic reviews of the literature report evidence of the effectiveness of speech and language therapy interventions for expressive language outcomes for children with SLI that cannot be accounted for by low IQ, behaviour or emotional problems or hearing or neurological impairments.^{3,51} However, the picture for receptive language outcomes is more problematic owing to a dearth of evidence, particularly from randomized controlled trials (RCTs), the effects of early remission,⁴ lower incidence of RELI relative to specific expressive language delay, and variability in the criteria for eligibility for recruitment.

Law et al. in an early review⁵¹ identified only five studies with receptive language outcomes that met the eligibility criteria of controlled studies of effects of intervention upon children in the age range 0 to 7 years with ‘primary’ speech and language delay (akin to SLI but not based upon formal psychometric discrepancy criteria). Four of these studies involved children aged 36 months or younger. This raises issues about the reliability of the test scores because measures obtained from preschool children are particularly susceptible to the influence of factors associated with development, such as short attention span and distractability, levels of activity, and prob-

lems in engaging with an unfamiliar test administrator.⁵² In addition, few of the participants in these studies had RELI, with receptive language outcomes reported for children receiving intervention for specific expressive language delay. It is thus unclear whether these interventions would be beneficial for children with RELI.

In Law et al.'s more recent review,³ only two studies met inclusion criteria for interventions targeted on receptive language (here, receptive syntax) with 'no treatment' control groups, both reporting non-significant effects. In the first of these studies Glogowska et al.⁵³ reported a non-significant standardized effect size (SES) of 0.19 (95% CI -0.12 to 0.51) from a sample of 155 preschool children (71 receiving treatment and 84 controls). In the second study, Law et al.⁵⁴ reported a non-significant SES of -0.45 (95% CI -1.18 to 0.28) from a sample of 38 preschool children (28 treatment and 10 controls).

Evidence from recent randomized control trials

Evidence from four recent large-scale RCTs not thus far included in published systematic reviews report interventions for children with RELI. Three of these studies investigated interventions based upon underlying auditory processing deficits. The fourth was based upon existing models of language therapy in the UK.

Cohen et al.⁵⁵ reported the findings from a multicentre, intention-to-treat RCT performed in Scotland with blind assessment of outcomes to determine the effectiveness of the Fast ForWord-Language program.⁵⁶ This is a computer-based intervention that utilises auditory processing theory³⁹ and uses games with signal-processed modified speech designed to compensate for underlying auditory temporal processing difficulties. The participants ($n=77$) were aged between 6 and 10 years, and were monolingual English speakers with a diagnosis of RELI. Average scores for both receptive and expressive language on the Clinical Evaluation of Language Fundamentals (CELF-3^{UK}),⁵⁷ a standardized language test, were -2SDs below the mean. The children were randomized into one of three groups: a group receiving Fast ForWord-Language, a comparison group receiving ongoing language therapy, and a second comparison group who played educational computer games with unmodified speech. Outcomes were measured at 9 weeks' post-baseline assessment and at 6 months' follow-up by qualified SLTs not otherwise involved in the project who were blind to the children's research group. The results revealed no significant additional benefit from playing the Fast ForWord-Language games 5 days a week for 6 weeks under parental supervision for 90 minutes each day relative to the first control group (SES -0.04 [95% CI -0.59 to 0.52] for receptive language) nor relative to the computer-games control group, who played commercially available educational computer games without modified speech on the same schedule. This trial did not support auditory processing deficits as a general explanation of severe RELI, although this was a particularly impaired cohort of children.

A recent large-scale RCT performed in the USA⁵⁸ also investigated the effectiveness of Fast ForWord-Language.

Participants included children with RELI but their progress could not be distinguished among children with specific ELI.

Bishop et al.⁵⁹ addressed issues relating to both auditory temporal processing and to limited general processing capacity explanations of RELI in an RCT involving 36 participants aged 8 to 13 years. The children had scores of less than -1SD on standard measures of language. Participants with RELI failed to benefit from a computer training program for comprehension of grammatical constructions to help sentence comprehension (SES 0.04 [95% CI -0.2 to 0.28]). The findings once again fail to support auditory processing deficits as a general explanation of RELI within the range of their study. They would, however, be compatible with a general limited processing capacity explanation and suggest that a more individualized, contextualized approach may be preferable for children with RELI, in contrast to the computer-based rote-learning approach used on the study.

Boyle et al.^{60,61} investigated the effectiveness of current language-therapy practices based upon meta-analyses of published studies.^{51,62} The participants in their RCT were 161 children aged 6 to 12 years who had persistent primary receptive and/or expressive language impairment with no reported marked hearing loss and no moderate/severe articulation/phonology/dysfluency problems or who otherwise required individual SLT work. Eighty-six of the children had RELI (defined using a threshold criterion of CELF-3^{UK} Receptive Language⁶⁰ standard score ≤ 81 and non-verbal IQ scores of >75) and 75 had specific expressive impairment. They were randomized to one of five conditions, which were as follows: (1) Individual, direct project therapy: SLT working individually with a child ($n=34$, 20 with RELI); (2) Group direct, project therapy: SLT working with a small group of children ($n=31$, 17 with RELI); (3) Individual, indirect project therapy: a trained SLT assistant working individually with a child ($n=33$, 17 with RELI); (4) Group, indirect project therapy: a trained SLT assistant working with a small group of children ($n=32$, 18 with RELI); and (5) Control group (who received existing community-based services; $n=31$, 14 with RELI).

Project therapy was delivered three times per week for 15 weeks, in 30- to 40-minute sessions, and those in the comparison group received their ongoing therapy regime. The therapy focused on comprehension monitoring, vocabulary development, grammar, narrative, and developing language learning strategies. All post-baseline measures were blind-assessed by qualified SLTs not otherwise involved with the project.

There was no significant difference between the four modes of project therapy but children with specific expressive impairment made greater gains in both receptive and expressive language than those with RELI (all p values <0.025). Further, although the children receiving project therapy made significant overall gains in expressive language ($p=0.031$), there was only a modest and non-significant intervention effect for receptive language scores relative to the comparison group for the subgroup of children with RELI (SES 0.25 [95% CI -0.32 to 0.82]). However, the impact of the small numbers involved on the statistical power of this comparison (14 in the

comparison group and 72 receiving project therapy) should be noted.

Recent phase I and small-scale trials including children with established RELI also suggest vocabulary development as a promising intervention. Direct teaching of vocabulary was effective with four children aged 10 to 11 years (Easton et al.⁶³) and two children aged 8 to 9 years using criterion-referenced measures,⁶⁴ and as effective as narrative intervention in developing language skills with a cohort of 54 secondary school children with RELI.⁶⁵ 'Traditional' therapy including vocabulary teaching was as effective at encouraging eight children over 8 years old with severe RELI to use a mental visualization strategy to aid their comprehension of oral narratives.⁶⁶ Interestingly, mental imagery training itself produced a significant improvement in the responses of children with RELI to literal questions about a short narrative.⁶⁶ Furthermore, a small-scale RCT found that developing semantic definitions of verbs was as effective as syntactic-semantic shape coding on criterion-referenced measures of verb argument structure for 27 children aged 11 to 16 years with severe RELI attending a specialist residential school.⁶⁷

In the case of young preschool children, Camarata et al.⁶⁸ found that a treatment group of 21 children with an mean age of 31 months with RELI made significantly greater gains in receptive language in response to an intervention focused on expressive grammar than a randomly allocated comparison group of six children (mean age 37.6mo; $p < 0.05$, SES 1.07). The intervention consisted of twice-weekly individual sessions of an hour for 12 weeks using imitation, modelling, and conversational re-casting approaches targeted on improving production of grammar. Further investigation of such transfer effects with young children would be of interest, although it should be noted that the eligibility criteria of 1SD below expected levels on standardized measures of both expressive and receptive language would have resulted in the recruitment of some children with less severe levels of impairment. It is also unclear to what extent these problems are likely to persist over time.

Mapping practice onto theory

The extent to which professional practice with children with RELI maps on to underlying theory has been investigated in a recent survey of qualified SLTs in the UK, focusing on practice with children aged 5 to 11 years.⁶⁹ The findings revealed that children with RELI are seen as a priority and receive extensive services that reflect diverse practice. This includes interventions targeted on specific deficits or based upon published programmes/frameworks for practice, behavioural approaches to teaching vocabulary, and sentence comprehension. Meta-cognitive activities (e.g. training to think about communication) were widely used, in particular with older children: nearly 80% of all activities reported with those aged 11 years compared with some 20% of reported activities for those aged 5 years. Underlying theory did not appear to be regarded as important for informing intervention, and SLTs placed more emphasis on the presenting problems associated with the child's deficit. This begs the question of the use of

current theory for informing interventions and of the effectiveness of disseminating research findings to practitioners. However, the low number of respondents ($n=56$) should be noted.

DISCUSSION

There are relatively few published controlled intervention studies (most of which are based on monolingual English-speaking populations) and an overall lack of evidence for approaches to effective treatment for children with RELI. Given that auditory processing interventions, and regimes based upon intensive delivery of existing therapy in particular, have not thus far proved very effective with this group, there is a case for investigating the use of further approaches. There are two complementary directions for such research. Phase II exploratory trials⁷⁰ to determine the feasibility of promising impairment-based interventions, for example therapy regimes for vocabulary development,^{71–74} and 'transfer' effects between expressive language intervention and receptive language gains in the case of young children with RELI, are needed to guide future full-scale RCTs, which are difficult to resource and populate. In addition, given the persistent nature of RELI, there is a pressing need to investigate 'enabling' interventions; i.e. interventions that support children in coping with RELI and maximize academic attainment.

The intractable nature of RELI also suggests that interventions that help children cope with ongoing receptive difficulties will be needed, to ensure that the children have the opportunity to experience facilitating communication environments and learn coping strategies, as the linguistic demands of literacy, education, and work increase.⁷⁵ This will mean that functional communication goals and interventions aimed at increasing participation will be needed over extended periods of time.

Many children with reading comprehension failure have comorbid difficulties in oral language comprehension,⁷⁶ but these may go unrecognized in schools because they may have no overt problems in speech, phonological processing, or word reading accuracy.⁷⁷ Investigating and identifying RELI among children with educational failure affords an opportunity for appropriately tailored literacy materials and instruction, as well as reciprocal teaching.⁷⁸ Advice and guidance can encourage classroom and official talk that is sensitive to linguistic processing limits and welcomes requests from the children for clarification when they do not understand. Children can also be encouraged to develop the executive and self-regulation skills used in planning, goal setting, monitoring, and completing tasks, applying working memory, sustaining attention, and inhibiting impulses,⁷⁹ and to manage their emotional states⁸⁰ in order to aid learning and understand what is socially acceptable in their school and adult communities.

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