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SPECIFIC LANGUAGE IMPAIRMENT IN CHILDREN

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OVERLOOKED BY PUBLIC HEALTH: SPECIFIC LANGUAGE IMPAIRMENT

Mabel L. Rice, University of Kansas highlights specific language impairment and why it often goes unrecognised as health disorder

The human language faculty has received centuries of interest by scholars of different disciplines, including philosophers, linguists, psychologists, educators, among others. It is widely admired as a remarkable gift by nature to humans, regarded as a unique characteristic of humans among the many species of living things. Children around the world acquire their native language without explicit teaching, even though languages come in many different forms – 6,000 different languages are estimated to be in use today. Just as walking is expected for young children everywhere, so is talking in sentences that can be understood by adults. The language-learning task in front of babies is now known to be incredibly complex, which makes this universal achievement even more impressive and at the same time, because it is universal, often thought to be simple because babies can do it.

Raising awareness for specific language impairment

The fact that it is so easy for most children obscures the fact that it is selectively difficult for some children. Children with Specific Language Impairment (SLI) are sometimes described as having the most common, but unrecognised, developmental disorder of childhood (and probably adulthood, too, given new outcome data). The point of this article is to bring SLI to the attention of policy makers, experts, and opinion leaders in public health, medicine, and education, as a largely unrecognised yet high impact disorder of childhood that persists into adulthood, creates high costs to societies and life-long frustrations or shame to the affected persons. It is undoubtedly a crucial but often misunderstood barrier to personal self-actualisation in life.

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According to the National Institute of Deafness and Other Communicative Diseases (NIDCD) in the U.S., Specific language impairment is defined as "a language disorder that delays the mastery of language skills in children who have no hearing loss or other development delays." Population-based studies report 7-10% of 5-year-old children have SLI, making it the most common manifestation of language impairments in children and the most common early childhood disorder, more common than Attention Deficit/Hyperactivity Disorder (ADHD) and autism combined. Children with SLI are at high risk for lower academic achievement relative to age peers, to encounter difficulties establishing social relationships and to end their education at high school completion[1,2]. Language

impairments are associated with increased health costs starting in early childhood and approaching the teen years[3]. Modelled outcomes from 5 to 34 years shows increased risk of unemployment for children with a history of SLI[4]. Girls with a history of SLI are almost 3 times more likely to experience sexual abuse as adolescents or young adults than girls without SLI[5]. Multiple studies report that only about 25% of children with SLI receive treatment when they enter school, around age 5 years.

Why SLI has a low profile in healthcare sectors

Although there is increasing reason to view SLI as an important public health issue, it has a very low profile in public health forums. An important exception is the recent inclusion of the goal to increase the proportion of children with language disorders who receive intervention services as part of the Healthy People 2020 initiative in the U.S., developed by the Office of Disease Prevention and Health Promotion. In the press of life-threatening diseases around the world that threaten the well-being of children and their families, it could be argued that SLI would be a "mild" disorder that could be deferred for a later time when more resources are available. On the other hand, it is clear that in modern societies the ability to use language well is increasingly important for all sorts of life interactions. It is crucial for the effective use of electronic media, understanding the many documents of the modern world, mastering an academic curriculum, advocating for oneself whether in childhood disputes or threat situations, in applying for a first job, health literacy, and engaging in the interactions of commerce. As indicated by the few studies of long-term outcomes of children with SLI, economic risk is likely to be part of the scenario.

At the level of scientific inquiry, overlooking the presence of language impairments can complicate interpretation of outcomes of studies of children with autism, ADHD, or low nonverbal intelligence, who may or may not have language impairments[6,7]. For example, in paediatric studies evaluating diseases such as HIV and associated treatments, recent detailed studies of language impairments

show that language impairment is the most common adverse outcome[8] and those predictors are not the same for children who have language impairments selectively or in combination with hearing loss or low nonverbal intelligence[9-11].

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The point here is that public policy expertise is vital for a better understanding of the social and economic consequences of SLI and possible treatment options. Conversely, a better understanding of SLI is vital for the necessary studies of public policy that bear on this important condition. This paper is the first in a series of papers to appear in subsequent issues of Open Access Government. Future instalments will address these questions: How is the language of children with SLI different from typical children? How does SLI compare to Hearing Loss, Speech Sound Disorder, Intellectual impairment or ADHD? What causes SLI? What are the social consequences of SLI? What should public policy experts consider for future investigations and ways to pursue effective treatments?

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SPECIFIC LANGUAGE IMPAIRMENT: WHAT DO WE KNOW?

Mabel L Rice, University of Kansas details how the language of children with specific language impairment differs from typical children

A recent paper in this publication introduced the condition of Specific Language Impairment (SLI) as a largely unrecognised yet high impact common disorder of childhood (7-10% of children) that persists into adulthood and warrants further consideration by public health experts. SLI is a language disorder that delays the mastery of language skills in children who have no hearing loss or other developmental delays. Recent studies reveal the ways in which language development in children with SLI is not the same as unaffected children, yet also shows many of the same strengths.

"SLI is a persistent language disorder that is evident early in development and has many similarities to younger language profiles with steady gains in language, levelling off in adolescence. This makes it likely that a child with SLI will become an adult with weak language skills, particularly in some, but not all, parts of the grammar."

These 3 findings are crucial to understanding the differences and similarities across the age span of 1 to 20 years of age. To sort this out, we need to consider the developmental arc of children's acquisition of language, from first words as toddlers, through childhood, and into adulthood. We also need to consider different dimensions of language; single words appear first, followed by simple sentences that adhere to grammatical rules. Some features of the adult grammar are relatively late-appearing in English-speaking children and those features are especially late for children with SLI.

Finding 1: Children with SLI are likely to be late language learners

Most children start producing words between 1 and 2 years of age, and then follow an accelerated rate of language acquisition. As shown in Figure 1, children with SLI can be delayed by 1 or even 2 years in this early start-up period. Studies of preschool children report that the language of 5-year-old children with SLI resembles that of 3-year-old typically developing children, a notable delay at a time of rapid change. Yet when their language system begins to grow, it does so at a rate and pattern of change much like that of younger children. Children with SLI seem prepared to learn language in much the same way as other children, only with a delayed start. Because the rate and pattern of change in children with SLI parallels that of unaffected children, they are not likely to 'catch up' to their age peers. Yet when children with SLI reach preadolescence they, like typically developing children, slow their rate of language acquisition and then level off into adulthood, the stable end-state for much of language development[1].

Finding 2: Delayed vocabulary development can follow the pattern of late start + similar growth trajectories

Not all children with SLI have vocabulary deficits beyond the preschool years, but if so, the pattern holds over many years. Vocabulary growth is

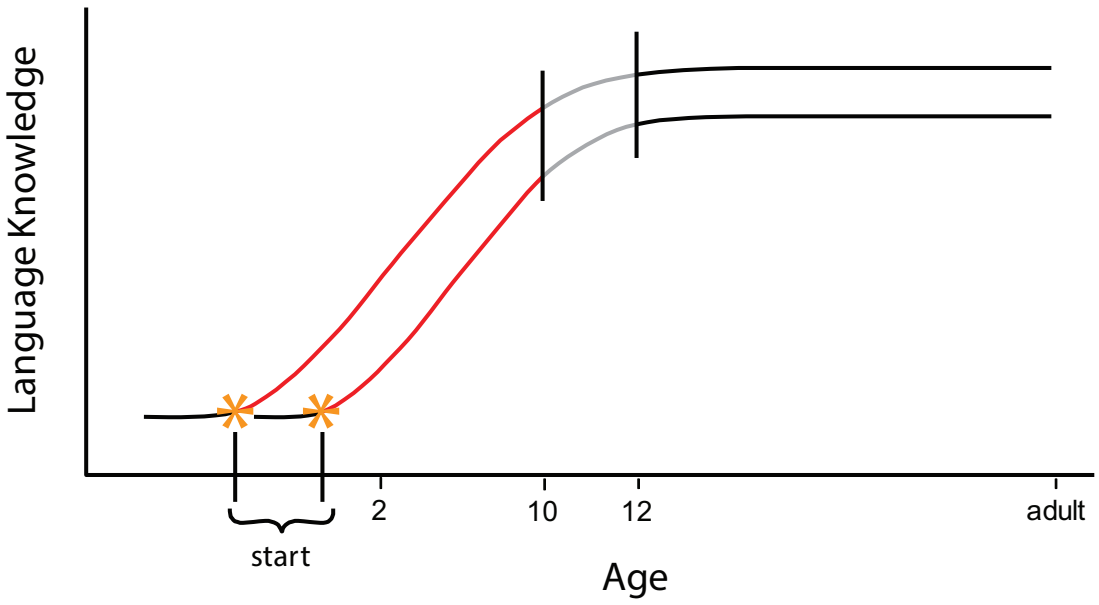

Figure 1: Developmental Trajectory of Language Growth from Start to Adulthood

shown in Figure 2[2]. This figure charts children's understanding of words, from 4 to 20 years of age, in a study that tested the same children annually. The SLI group is at the bottom 15% for their age and, in this study, the typical group is in the top 15%. At 4 years the SLI children, on average, know fewer words than children at higher levels for their age. This is not surprising; what is of interest is that the children at the low end learn new words at the same rate over time as the comparison group, but they don't close the gap. Instead, for both groups the rate of new word learning markedly slows with age, beginning at 10-12 years of age and maintaining a slower rate into adulthood. Although before 4 years of age girls tend to have larger vocabularies than boys, this study found girls to have a slower rate of word acquisition than boys in adolescence, which left a marked disadvantage for girls with lower levels of vocabulary in the crucial time of education for preparation into the work place or higher education.

Finding 3: Tense and agreement marking on verbs is a grammar requirement likely to be difficult for children with SLI

This is evident in the use of forms of auxiliary or copula BE, as in "He is happy" and "Are the girls here?" as well as Auxiliary DO, as in "Does he want a cookie?" past tense, as in "Patsy walked home" and third person singular, as in "He wants a cookie." The persistent problem is that children with SLI, on average, omit these parts of the sentence as if the requirement were optional instead of obligatory. Figure 3 shows how persistent this tendency is in children with SLI[3]. As expected, between the ages of 6 and 16 years typically developing children recognise in judgement tasks that omission of these parts of the grammar render a sentence ungrammatical. Even at 6 years this is an easy task. Yet children with SLI, on average, persist in considering omissions as optional and OK for grammar. It is as if they are stuck with an immature form of the grammar into adolescence.

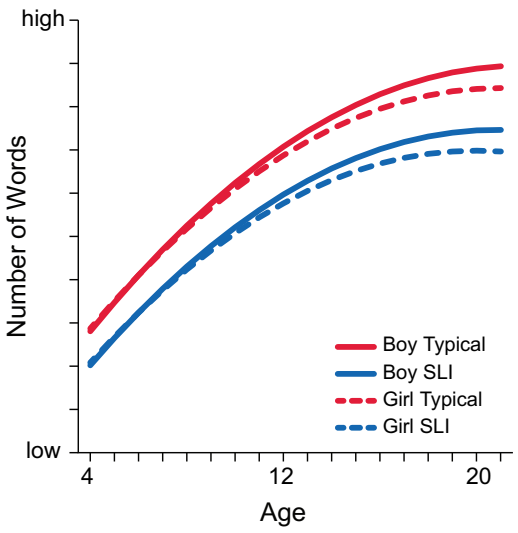

Figure 2: Growth of Word Comprehension from Childhood to Adulthood

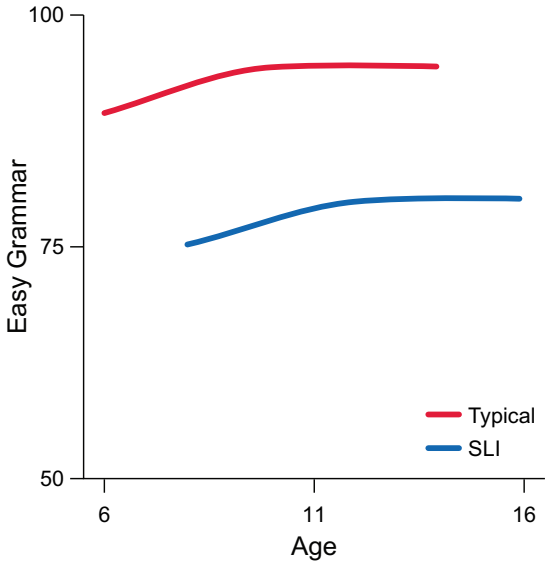

Figure 3: SLI Children's Judgment of Omitted Tense and Agreement Markers in Childhood

Interpretation: SLI is a persistent language disorder that is evident early in development and has many similarities to younger language profiles with steady gains in language, levelling off in adolescence. This makes it likely that a child with SLI will become an adult with weak language skills, particularly in some, but not all, parts of the grammar. Much of the weakness in language can be hidden under compensatory social skills, and thereby goes undetected as a likely contributor to poor reading skills or avoidance of social interactions in adolescents and adults. Current studies investigate genetic influences on the causal pathways, as inherited language abilities can drive the strong language growth trajectories, yet selective inherited differences operative at key times could account for persistent and unresolved delays[1].

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Funding references:
NIH: R01DC001803, R01DC005226, P30DC005803, R42DC013749, P30HD00528 and T32DC000052.

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WHAT CAUSES
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S SPECIFIC LANGUAGE IMPAIRMENT? COGNITIVE IMPAIRMENTS ARE NEITHER OR SUFFICIENT

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Children's language starts out very simply and grows with age. Yet some children do not keep up with their age peers. This is a developmental condition known as "language impairments," which sometimes but not always exists when children have other developmental disorders, such as hearing loss or cognitive impairments. The cause of children's language impairments is not known. People often draw upon the observed overlap with other obvious developmental disorders such as hearing loss, intellectual impairments, autism spectrum disorder (ASD), Down syndrome, or Fragile X to conclude that language impairments share the same underlying cause. This assumption, although widespread, is off the mark in important ways and can create misleading impressions of children with language impairments and their families.

Instead, the acquisition of language follows a distinct pathway. This is evident in documentation of selective sparing of language in children with cognitive impairments and selective impairment of language in children's development, most notably in children with Specific Language Impairment (SLI). In other papers in this e-book, articles describe this most common, but unrecognized, developmental disorder of childhood, which most likely persists into adulthood for many of the affected children. Best estimates are that 7-10% of children who have no hearing loss or other developmental delays show language impairments at school entry, around 5 years of age[1,2].

Another paper in this e-book describes the ways in which the language of children with SLI is not the same as unaffected children, yet also shows many of the same strengths. Important features of SLI are that children are not likely to outgrow it, are likely to encounter difficulties in learning to read and likely to struggle in other academic endeavors. People can assume the children may not be very bright, or are simply poorly motivated, assumptions that only add to the frustration of children with undiagnosed SLI.

Selective sparing of language acquisition in cases of very limited cognitive abilities

To unpack the relationship between general cognitive ability and language ability, let us begin with cases of selective sparing of language in persons with very limited cognitive abilities. Such children have been documented in the literature for a long time and are of interest to scholars of the origins of human language abilities. For example, one well documented phenomenon is the "cocktail party syndrome" for some patients (adults or children) with spina bifida and/or hydrocephalus, in which subnormal intelligence co-exists with excessive talking comprised of superficial content but well-formed and sometimes quite complex grammar[3]. A detailed case study is reported of an adolescent female referred to as "D.H." She functioned in everyday life situations at the level of significant cognitive impairments. Yet in her spontaneous conversation her language appeared to be normal, with appropriate syntax, vocabulary and interactive topic maintenance. The conclusion of the linguists who studied her was that she had skillful use of all aspects of language, in spite of her cognitive impairment,

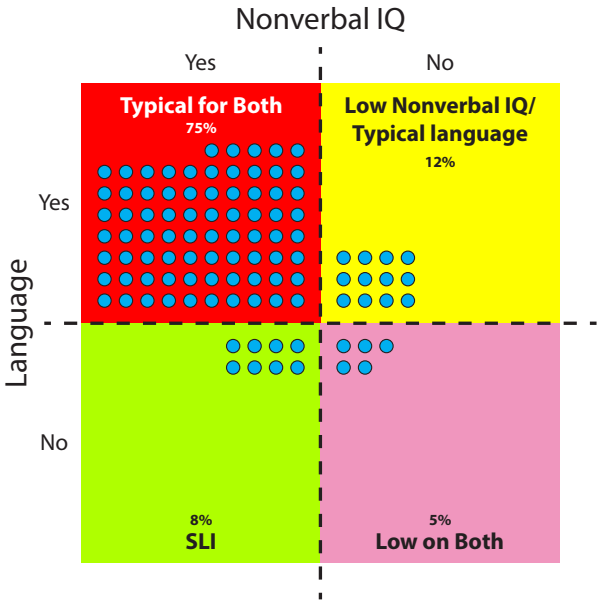

Figure 1: Distribution of children according to language and nonverbal IQ scores

posing a puzzle that persists to the present[3].

Another well documented case history describes a young man with limited cognitive abilities who had extraordinary linguistic prowess. He was a native speaker of English, with a diagnosis of brain damage of an unknown kind. His performance IQ scores were between 42 and 75 (100 is expected) and his verbal IQ scores were between 89 and 102 (also with 100 as expected). As a young adult, he lived in a sheltered community with assistance with daily living activities. He had a remarkable ability to translate from and communicate in a large number of languages, with some knowledge of Danish, Dutch, Finnish, French, German, Modern Greek, Hindi, Italian, Norwegian, Polish, Portuguese, Russian, Spanish, Swedish, Turkish and Welsh. His linguistic abilities were studied in great detail by linguists hoping to capture the underlying properties of universal grammar that were spared in his condition[4].

Children with ASD are often thought of as having language impairments and limited cognitive abilities. Yet the recent revision of the Diagnostic and Statistical Manual of Mental Disorders: DSM-5[5] revised the diagnostic criteria for ASD to demote language impairments from a centrally defining characteristic to a "specifier" condition. This means that ASD can be officially diagnosed in children with or without language impairments, and with or without intellectual impairments. Instead, deficits in social communication and social behavior are crucial to diagnosis, as are restrictive, repetitive patterns of behavior, interests, or activity. In other words, the DSM-5 criteria recognize that classic language impairments are not intrinsically interconnected with social communication deficits or repetitive behaviors, and are not diagnostic of ASD.

Documentation of independence of language impairments and cognitive impairments in population-based samples of children

Population-based studies recruit a large sample of children representative of the full range of children in the population. When all children in the sample are measured, it is possible to identify children with SLI and children who have low nonverbal IQ but do not have language impairments. Either of these groups are likely to be unidentified by educators or other special service providers if there is a strong assumption of causal overlap for cognitive impairments and language impairments in children without other obvious developmental disabilities indicative of brain damage or other syndromic conditions. The evidence required to evaluate this possibility is expensive to obtain because assessments should be individually administered to each child for domains of language and nonverbal cognitive abilities. This is time-consuming and requires well trained data collectors, in addition to experts in experimental design and quantitative analyses. One such study[2,6] is of interest here because measures from the same 5-year-old children are reported for general language assessment, nonverbal IQ, a grammar marker[7], and speech disorders[8]. Children were excluded from the sample if they had neurological disorders, clinical syndromes, and/or hearing loss. The outcomes can be presented as percentages of children who fall into four groups based on levels of language ability and nonverbal IQ: 1) Typical or above in both language and nonverbal IQ; 2) Low levels of both language and nonverbal IQ; 3) Typical language and low nonverbal IQ; and

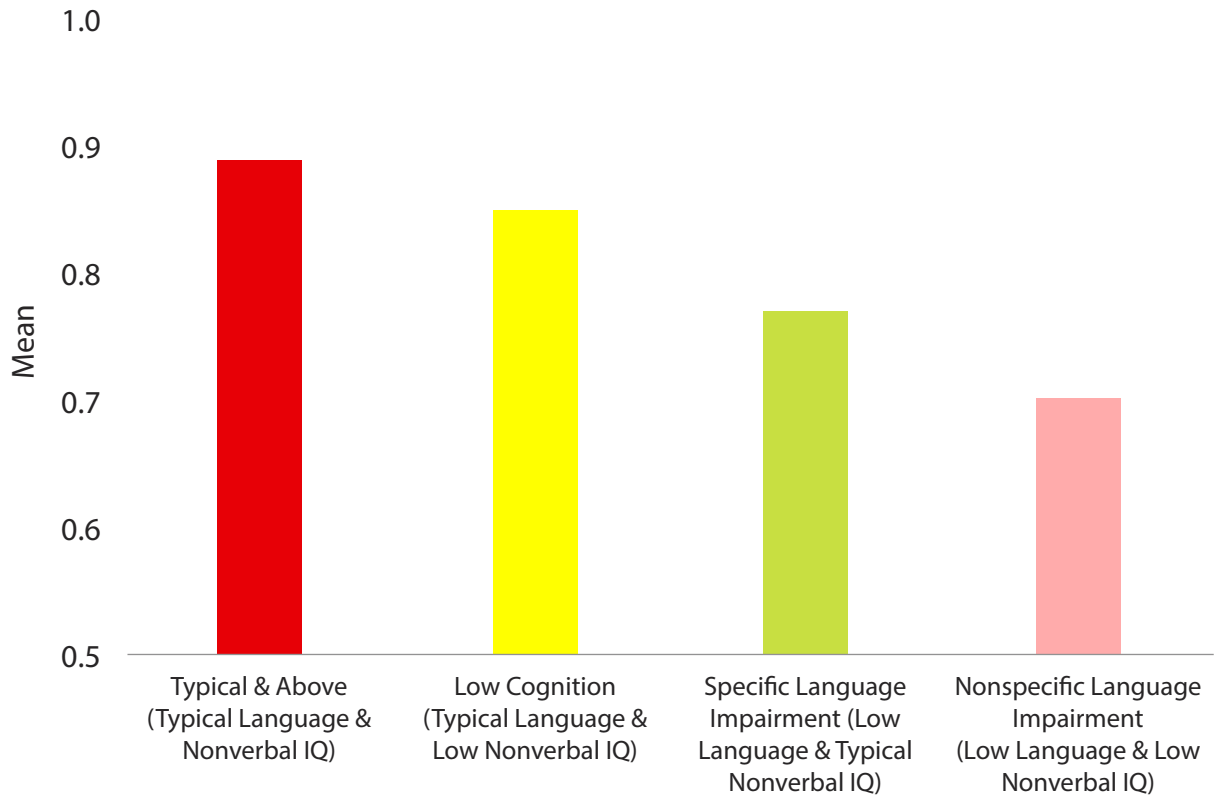

Figure 2: Grammatical Tense Marking

4) Low language and typical or above nonverbal IQ (i.e., a profile consistent with SLI). See another paper[9] for more details about the assessments in the study.

The results are displayed in Figure 1. Two cells, indicated in red and pink, are expected to collect all the children if language and nonverbal IQ are tightly associated in a shared causal pathway. Instead, we see nontrivial exceptions in the off cells: Children with low nonverbal IQ but nevertheless typical or above language scores (~12%) (shown in yellow cell), and children with SLI (low language with typical or above nonverbal IQ) (~8%) (green cell). The expected red cell of "typical children" captures 75% of the sample; the expected pink cell of low language and low nonverbal IQ captures 5% in this sample which excluded children with neurological disorders. So roughly 20% of the children show a profile inconsistent with the assumption that low nonverbal abilities cause SLI. The conclusion is that low nonverbal IQ levels are neither necessary nor sufficient for language impairments in children.

On a more detailed level, there is evidence in support of selective sparing of certain properties of the grammar. English sentences have a requirement for grammatical tense marking (also called "finiteness marking") for a full clause. This is especially evident in questions with DO, such as "Where does he go?" The form of DO is required to mark tense and agreement with the subject, without any contribution to the meaning of the sentence. Other indicators of grammatical tense marking are copula and auxiliary forms of BE, third person singular -s, regular and irregular forms of past tense[10]. Young English-speaking children are likely to use these forms optionally where they are required in sentences for some time before they consistently use them, a period that persists for children with SLI[11,12]. Thus, there is reason to think of this grammatical requirement as "weak" in some children. The results are shown in Figure 2, which reports the mean percentage correct for grammatical tense marking. Five years

of age is when typically developing children are reaching full mastery, with a group mean of .90 or 90% use in obligatory contexts as shown in the red bar of the figure. The Low Cognition group is not statistically significantly different, with a mean of .86 (yellow bar). On the other hand, the SLI group scores statistically significantly lower, with a mean of .78 (green bar), and the Low/Low group is lowest, with a mean of .71 (pink bar). The pattern across the groups provides further support to the possibility of selective sparing of grammar in children with low nonverbal IQ even when they do not have classic clinical syndromes associated with developmental disorder, as documented in the cases above. Thus, the grammar marker of SLI is not diagnostic of low nonverbal IQ.

The assumption that low nonverbal IQ causes SLI could be related to the ways in which children who do not match our expectations can remain undetected. The cases of selective sparing of language can cause people to assume a child has robust nonverbal IQ when this is not true, thereby leading to frustration for the child in school where a lack of nonverbal ability can be attributed to low motivation or poor study habits. On the other hand, children with SLI often go undetected in part because they develop compensatory ways of avoiding situations that call for more language ability than they have. This can be apparent in their avoidance of advocating for themselves in childhood disputes with peers, or avoiding verbal participation in class activities. Another way they remain invisible is that most children with SLI do not have clinically significant speech disorders, even at school entry. In the study reported in Figures 1 and 2, 98.2% of the children had developmentally appropriate speech in a measure adjusted for mild misarticulations, such as a frontal lisp, that may be evident at 5 years but are likely to be outgrown by 7 years of age. The rate of speech impairments in children with SLI was estimated at 0.51%, suggesting that speech disorders and language impairments are likely to appear independently in young children. The rate of speech impairments in the low/low group was 0.77%; in the low nonverbal/typical or above language group the rate was 0.5%. A recent population study also reported high levels of speech ability in children with language disorders[1] although the groupings were not defined out in the same ways as the study reported here, i.e., in the prevalence estimates the low nonverbal IQ group included children who did not have language impairments.

The conclusion is that the common assumption that children with SLI are not very bright is not warranted. Instead, children with SLI can have normal or above nonverbal IQ levels and children with low nonverbal IQ levels can have language abilities as expected for their age. Further, these non-confirming conditions/cells are not rare, comprising about 20% of the population. Language can be selectively impaired or selectively spared. This means scientists need to search for two possible causal pathways that sometimes intersect, instead of one common cause. It also means the common assumption needs to be suspended when encountering children likely to have SLI.

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WHAT THE LANGUAGE OF TWINS CAN TELL US ABOUT CHILDREN WITH SPECIFIC LANGUAGE IMPAIRMENT

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One unexplained kind of language impairment in children is known as Specific Language Impairment (SLI). As the name implies, children with SLI have language impairment "specifically", i.e., with no other conditions that are known to cause language impairment. The children with SLI do not have overt neurodevelopmental disorders, hearing impairments or other syndromes such as autism spectrum disorder (ASD) or Down syndrome. The causes of SLI are a puzzle, given that language acquisition is very robust in most children despite great variations in other dimensions of children's development and great variations in the ways in which families around the world raise their children. It is not necessary for families to explicitly "teach" their children the grammar of their languages; instead, children seem to spontaneously "pick it up" when they are toddlers and quickly increase their abilities thereafter. Yet the children with SLI start their language systems later and are at risk for persistent low language abilities into adulthood.

The cause of SLI is unknown, although there is growing evidence pointing toward the likelihood of inherited risk for SLI. Twin children provide an informative "natural experiment" for evaluating possible inherited factors contributing to SLI. Here I will summarize the logic of twin studies and outcomes of recent studies that support possible inherited factors on the causal pathway for language impairments such as SLI. I will also note how twins' language acquisition differs from that of single-born children.

The case of twins

Twin children are paired in their development before they are born, sharing their mother's uterus; they are born at the same time and share their home environments as they are raised and grow through childhood. Nature provided essential differences across pairs of twins, in the form of two kinds of pairs. Monozygotic (MZ) twins, sometimes called "identical" because they tend to look alike, begin as a single fertilized egg that splits into two, i.e., the two children develop from one zygote, which splits and forms two embryos. Dyzgotic (DZ) twins, sometimes called "fraternal" because of their "brotherly" or sibling status, develop from two different eggs, each fertilized from its own sperm cell. Thus, MZ twins can be thought of as "duplicates" genetically whereas DZ twins are siblings who happen to be the same age. MZ twins are the same sex; DZ twins may or may not be the same sex.

Two pairs of twins are illustrated in Figures 1 and 2. Following common scientific conventions for showing pedigree relationships, Figure 1 depicts twin MZ girls, where the circles represent females (males are represented by squares), and the connected diagonal lines above their heads indicate MZ connectedness. In contrast, the DZ twin girls are female siblings without a connecting line between the two pointing lines. In these figures the level of language abilities for each child are indicated by the letter "L" within their heads. The MZ twin pair shares a large black "L" indicating robust language ability. The DZ twin pair has two different language symbols; one twin has a small red "L" instead of the expected large black letter, indicating one of the girls has low levels of language ability. Presence of the same trait, such as language

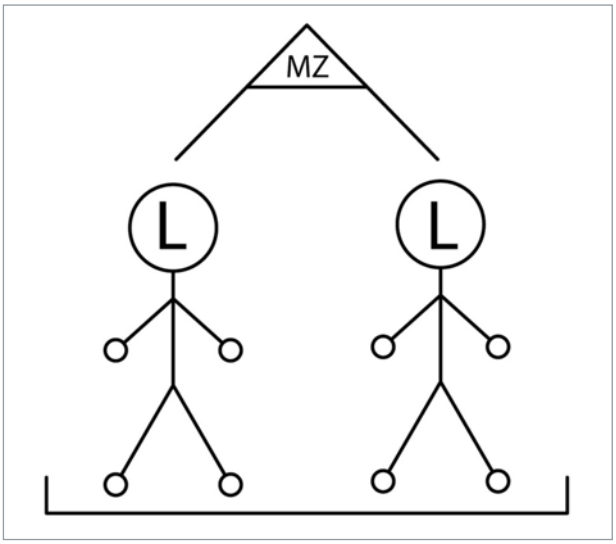

Figure 1: Monozygotic female twins concordant for robust language abilities

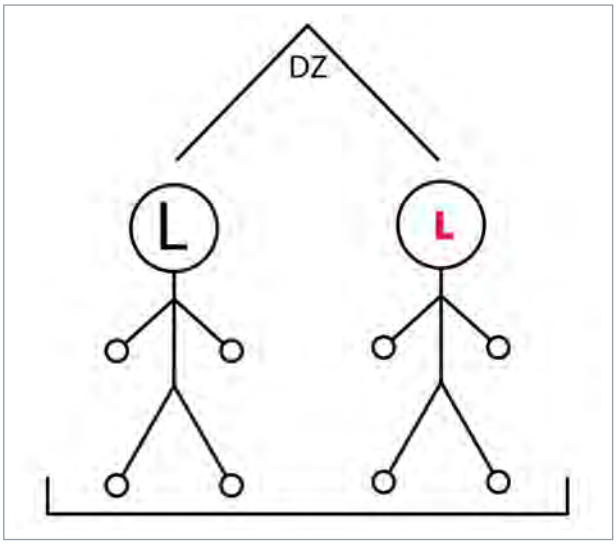

Figure 2: Dyzygotic female twins discordant for language ability

ability, in both members of a twin pair is known as "concordance."

The logic of twin behavioral research

Methods of behavioral genetics research build on the notion of concordance to estimate the sources of similarity and differences within twin pairs. This is represented by the equation h2+c2+e2=1.00, with "h" for heritability, "c" for environmental effects common to the twin pair, such as the resources of the home, and "e" for environmental effects unique to each twin, such as an episode of meningitis during infancy. Larger values for h2 are interpreted as increased likelihood of inherited influences on the trait, although the exact molecular genetic mechanisms are not revealed by the method.

Possible "twinning" effects on language acquisition

At the outset of language acquisition, twins can be compared to single-born children in the acquisition of early language milestones. An open question is whether twins may lag their age peers at the outset of language acquisition. A twinning effect could be related to the additional care required by two babies instead of one. If so, the expected outcomes would be as depicted in Figure 3, such that, irrespective of the kind of twin pairs, twin children could score lower on early vocabulary acquisition than same age singleton children.

Outcomes of recent twin studies

Twinning effects. Recent studies document twinning effects in the language acquisition of 24-month-old twins 1 and again at 4 and 6 years of age2. The outcomes are depicted in Figure 4. The "X" in the figures show that, instead of the expected score of 100 (or, 50th percentile) for their age, the children score a standard score of 90 (about the 25th percentile). Also, the figure indicates that the DZ twin children, on average, score somewhat higher than the MZ twins, a statistically significant difference that resolved by 6 years of age. These outcomes were found across multiple measures of language and speech development at each age level. These findings do not support the notion that a twinning effect is due to the burdens of raising two children because one kind of twin pairs was less likely to experience twinning effects than the other kind, although the child-raising burden posed by two babies would be the same for both kinds of pairs. Instead, the findings point toward a possible zygosity effect based on differences between the kinds of twin pairs, differences perhaps biological in nature. Twinning effects decreased slightly from 4 to 6 years, with a statistically significant improvement, moving toward the age-level expectations, although the gap is not yet closed with the age peers at age 6 years.

Heritability outcomes. Across studies, heritability

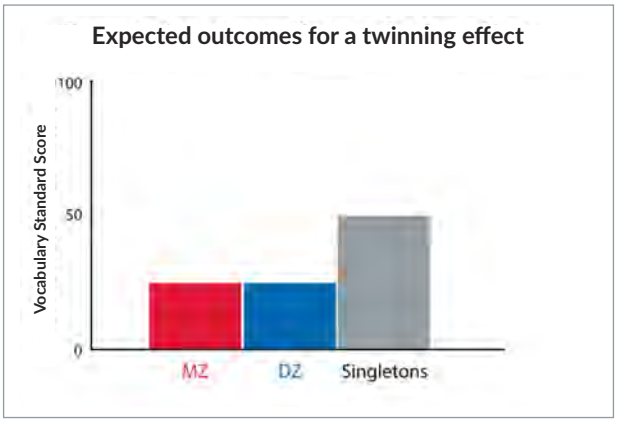

Figure 3: Expected outcomes for a twinning effect

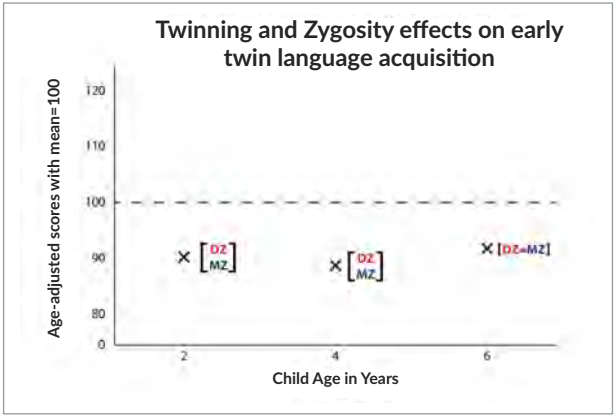

Figure 4: Twinning and zygosity effects on early twin language acquisition

estimates tend to increase with age, and to vary across the dimensions of language outcomes. For the full sample of twins, at two years of age, heritability is estimated as .25 for vocabulary, .52 (boys) and .43 (girls) for early grammar, and .22 for combining words[1]. At four years of age, heritability is estimated in the range of .10-.73, with the highest estimates for speech and mean length of utterance2. At six years of age, the h2 range is .54-.92 with the highest for grammar and speech[2]. These heritability estimates were from models adjusting for possible effects of perinatal status and external factors such as parental income.

Heritability outcomes also tend to be higher when studying children at the same age level and dividing the group into two, comparing twins in the typical or above range versus children who score below age peers. This can be called "heritability of language impairment" in samples of twins screened for children with clinical diagnosis of neurodevelopmental disabilities. A recent study of 16-year-old twins reported substantial heritability for language impairment[3]. For a grammar task, the estimates for genetic influence on low performance levels ranged from .36 with the criterion of the lowest 10% of the group to .74 with the lowest 5% of the group.

Lessons from twin studies for understanding SLI

Inherited influences on language acquisition in young children are suggested by heritability estimates from studies of twin children, although the exact molecular genetics are not yet identified. Although environment plays a role in language acquisition, the possibility of stronger inherited effects for children with levels of language substantially below age expectations further adds to the support of likely inherited influences in the causal pathways for SLI. Studies of twins suggest that heritability increases with age and is stronger for some dimensions of language than others, with grammatical abilities perhaps more heritable than vocabulary, for example. An important caveat from twin studies is that it is very important to be mindful of possible twinning effects on language in young twin children, effects which differ from the condition of SLI. On the other hand, a better understanding of the causes of twinning effects on language could help reveal characteristics of cortical development that guide language acquisition in young children or that are immature in children with SLI.

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