A Grammatical Specific Language Impairment in Children: 
An Autosomal Dominant Inheritance?

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The aim of this study is to provide further characterization of a subgroup of so-called "Grammatical specific language-impaired (SLI)" children. The Grammatical SLI children have a persistent and disproportionate impairment in grammatical comprehension and expression of language. Previous research has indicated that their language impairment may be characterized by a domain-specific and modular language deficit. This study provides an initial investigation as to whether there is a genetic basis underlying their disorder as has been found for other forms of SLI and for SLI in general. The incidence of familial aggregation of language impairment was investigated in 12 Grammatical SLI children (aged 9:3 to 12:10). A familial language impairment (LI) history was classified as positive if one or more of the probands' relatives had a history of a speech/language or reading/writing problem which required speech therapy or any other form of remedial help. Case history information provided an initial indication that the Grammatical SLI children had a significantly higher incidence of a positive familial LI history than could be expected by chance. A questionnaire provided evidence of a positive LI history in the first-degree relatives of the SLI probands and 49 normally developing control probands. The SLI probands had a clearly and significantly higher incidence of a positive familial LI history than the control probands (77.8 vs. 28.5%, respectively). The results are consistent with a genetic basis underlying Grammatical SLI. The pattern of impairment in the SLI probands' relatives is consistent with an autosomal dominant genetic inheritance. In contrast to the control probands, the SLI probands' impaired relatives did not show a male gender bias. Thus, the gene does not appear to be sex-linked. The data indicate that further research is warranted to investigate the nature of the LI in the relatives of the Grammatical SLI probands and the genetic characteristics of this subgroup. The implications for the biological, domain-specific, and modular bases to language are discussed.

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INTRODUCTION

In recent years there has been increasing interest in investigating children with a specific language impairment (SLI). SLI children are characterized by severe problems in the development of language comprehension and expression but do not have an impairment in nonlinguistic cognitive or motor development, hearing, or emotional–social behaviour which could account for their language impairments (Benton, 1964). It is now well recognized that SLI in children is a heterogeneous disorder. This study investigates a subgroup of 12 so-called “Grammatical SLI” children. The subgroup is characterized by a persistent and disproportionate impairment in the grammatical comprehension and expression of language. The findings from previous investigations of the linguistic characteristics of this group are consistent with the view that they have an underlying domain-specific and modular language deficit (van der Lely, 1996, 1994b; van der Lely & Stollwerck, 1994). This study provides an initial investigation as to whether a genetic basis could underlie this, arguably, modular language deficit. This is explored by investigating whether the Grammatical SLI children have a familial aggregation of language impairment. The combination of the nature of the linguistic disorder and the underlying deficit in these children may provide insight into the related issues of language modularity (Fodor, 1983), language acquisition (Chomsky, 1986), and the biological basis to language (Chomsky, 1981, 1986; Pinker, 1994; Pinker & Bloom, 1990).

Specific Language Impairment in Children

SLI children often show markedly different linguistic characteristics from one another. However, many SLI children are characterized by varying degrees of morphological and grammatical impairments in the comprehension and/or expression of language (Bishop, 1994; Leonard, 1989; Leonard, McGregor, & Allen, 1992; van der Lely, 1993, 1994a; van der Lely & Harris, 1990). Within this group potentially important differences exist. For example, SLI child may present with or without severe articulatory or phonological impairment, or without an impairment in the comprehension of language. More significantly, many of the children who present with this general pattern of language impairment at 4 years of age are indistinguishable from normally developing children in later years. For example, Bishop and Edmundson (1987), in a longitudinal study, found that more than one-third of their group of children with SLI at 4 years of age (i.e., 32 out of 87 children) improved so as to be indistinguishable from control children on language measures at 5:6 years. However, for a few children in this group the language disorder persists into adulthood.

It is as yet unclear to what extent the underlying nature and cause of SLI in children with different characteristics is related. Is there one cause of SLI in children, or are we looking at distinct disorders? Is the underlying cause
of a persisting SLI different from one that resolves with time? If we are to attempt to answer these questions it is important that subgroups of SLI children are identified and investigated separately because group means may obscure potentially important findings and may be unrepresentative of any type of SLI in children.

Relatively homogeneous subgroups of SLI children have been identified. For example, Bishop and Adams (1989) identified a subgroup of semantic–pragmatic SLI children; Gopnik (1990) and Gopnik and Crago (1991) have focused on a SLI family (i.e., a family of 30 members of whom half are language impaired) and van der Lely (1993, 1994a,b) and van der Lely and Stollwerck (1994) have investigated a subgroup of Grammatical SLI children. It is this latter group which is the focus of this paper.

We have identified a small subgroup of SLI children with a persisting SLI who are characterized by a similar linguistic profile. We have termed this group Grammatical SLI to distinguish them from other subgroups. We do not claim that this subgroup is, necessarily, an autonomous subgroup from all other SLI children (although this may prove to be the case), but we propose that the Grammatical SLI children are homogeneous within this subgroup.

Grammatical SLI children have been found to be particularly impaired in areas of inflectional morphology. For example, they frequently omit tense marking and subject–verb agreement. They are also impaired in assigning theta roles in reversible sentences and in assigning pronominal reference when syntactic structural knowledge is required. However, they are not impaired in using pronominal reference appropriately when this is determined by pragmatic knowledge (van der Lely, 1993, 1994b, 1996; van der Lely & Stollwerck, 1994). These data are consistent with the view that Grammatical SLI children have a domain specific and modular language deficit. A summary of the linguistic details of the subgroup can be found in van der Lely (1994b).

The Underlying Etiology of SLI

The findings from recent research are consistent with a genetic basis to SLI. Some of the strongest evidence for this has been the pedigree analysis of the SLI family studied by Hurst, Baraitser, Auger, Graham, and Norell (1990) and Gopnik (1990). Hurst et al. (1990) found that half of the SLI family were language impaired. Data consistent with a genetic basis for SLI have also been found for undifferentiated groups of SLI children (Bishop & Edmundson, 1986; Haynes & Naidoo, 1991; Lewis, Cox, & Byard, 1993; Neils & Aram, 1986; Tallal, Ross, & Curtiss, 1989a,b; Tallal, Townsend, Curtiss, & Wulfeck, 1991; Tomblin, 1989). Further evidence has been provided by twin studies. Significant differences in concordance of LI in monozygotic and dizygotic twins have been found by Lewis and Thompson (1992) and in Bishop, North, and Donlan’s (1995) large study of 90 twins.
While these studies cannot rule out the possibility of some environmental factors common to family members (Rutter, Bolton, Harrington, Couteur, MacDonald, & Siminoff, 1990), this seems unlikely, as both affected and unaffected children are found in the same family (Bishop et al., 1994).

Many investigations of SLI have found a significant difference between male : female ratios, with greater numbers of males affected (Bishop et al., 1994; Haynes & Naidoo, 1991; Tomblin, 1989). However, this sex-ratio bias was not found for the SLI family (Hurst et al., 1990). This indicates that a different genetic deficit could underlie different forms of SLI in children.

Not all studies of SLI children are consistent with a genetic basis to their disorder. Whitehurst, Arnold, Smith, Fischel, Lonigan, and Valdez-Menchaca (1991) identified a subgroup of young, preschool SLI children who were delayed in their expressive language but not in their comprehension of language. The study investigated whether there was a familial history of speech, language, and school problems in the immediate family of the children. Whitehurst et al. did not find a strong familial component of language delay in their subgroup of expressive SLI children. Thus, Whitehurst et al.’s findings argue against a genetic cause for all forms of SLI children.

It is a contentious issue as to whether a genetic deficit could underlie a domain-specific and modular language deficit as is arguably found in Grammatical SLI children (cf. Bates, 1993; Bates, Dale, & Thal, 1995; Johnston, 1991; Leonard et al., 1992). However, the studies which have argued against a genetic deficit causing a modular language impairment have been based on global concepts of SLI and global measures of language, such as “comprehension vs. production” (Bates et al., 1995). Language comprehension, by its very nature, may involve both specifically modular language processing (e.g., syntactic parsing) and nonmodular central system processes (e.g., pragmatic inferential processing) (Sperber & Wilson, 1986).

Previous research has established that central cognitive deficits may be caused by genetic abnormalities (Money, 1973; Nielsen, Sorenson, & Sorenson, 1981; Stewart, 1982). However, it is less clear whether a genetic abnormality may cause a modular language deficit without other impairments in children. Bates (1993) has argued that in cases in which a genetically based disorder has been implicated underlying a language disorder, such as the family of SLI children and adults studied by Hurst et al. (1990) and Gopnik (1990), and in undifferentiated groups of SLI children (Tallal, Ross, & Curtiss, 1989a,b), the subjects also suffer from impairments in aspects of cognition and/or perception that are not specific to language (Varghha-Khadem & Passingham, 1990; Tallal, Stark, & Mellits, 1985).

It is evident that if we are to investigate whether a genetic abnormality may underlie a modular language deficit it is necessary to: (i) distinguish between measures of language which primarily tap modular language processes from central system processes when identifying the language-impaired group; (ii) investigate children whose language may be characterized...
by a modular language deficit; (iii) investigate children who do not have any other known abnormalities in nonlinguistic cognitive functioning. This study aims to meet these demands by studying a carefully selected group of Grammatical SLI children. The paper provides the first step in investigating the possibility of a genetic basis for Grammatical SLI. The incidence and pattern of familial aggregation of language impairment in this population are compared with the incidence of LI in the population at large and with a large group of age-matched control subjects.

The questions addressed in this study were: (i) do grammatical SLI children have a familial aggregation of language impairment? (ii) If so, what is the pattern of the impairment in the four family members (mother, father, brother, sister), e.g., is it consistent with a recessive or dominant genetic inheritance, and is there evidence of a sex-linked inheritance?

METHOD

Subjects

The SLI children for this study were selected from approximately 150 SLI children attending one of four residential schools specializing in the education of SLI children in England. The SLI children in the school may come from any geographical part of England, and they come from a variety of socio-economic backgrounds.

The SLI children had been diagnosed by speech and language therapists and educational psychologists as having severe and persistent difficulties with language development as measured by standardized tests of language abilities. That is, their scores fell at least $-1.5$ SD below that expected for their chronological age on some language tests. Their nonverbal cognitive abilities as measured by performance subtests of standardized IQ tests (e.g., WISC-R; Wechsler, 1974) fell within normal limits for their chronological age. In other words, their performance IQ was 85 or above. Only children who meet these criteria attend the residential school for SLI children. Further general details of the children attending the school may be found in Haynes (1992) and Haynes and Naidoo (1991).

From the 150 SLI children who attended the school, 61 of the children’s chronological ages fell between 9:0 and 13:0 years. There were 47 boys and 14 girls in this age group. Further criteria were used to exclude children who did not fall into the grammatical SLI subgroup.

Children were excluded if they showed any signs of social or emotional abnormalities or “autistic like” symptoms (e.g., ritualistic behaviors), or had been diagnosed as having semantic–pragmatic disorder (Adams & Bishop, 1989; Bishop & Adams, 1989). Children were also excluded if they had articulatory dyspraxia or severe phonological disorder, omitted final consonants (regardless of the grammatical context), or were partially unintelligible (i.e., those children classified by Haynes (1992) as having speech or speech plus SLI). Thus, this group differs from some groups of SLI subjects (e.g., Hurst et al., 1990) in that they do not have a severe articulatory-speech impairment.

The children were then assessed on a battery of standardized and nonstandardized language tests which tapped a range of comprehension abilities and expressive language abilities. The choice of the tests was determined by their reliability in identifying SLI children and their frequent use in research, rather than by any adherence to theoretical notions on which the tests were based. The tests provided a standardized measure of language abilities in relation to the children’s chronological ages. There were six standardized language tests used for selection purposes: The British Picture Vocabulary Scale (BPVS) (Dunn, Dunn, Whetton, & Pintilie, 1982), a test of comprehension of single word vocabulary; the Test for Reception Of Grammar (TROG) (Bishop, 1983), a multichoice test of understanding a range of grammatical structures
in sentences; the Naming Vocabulary subtest from the British Ability Scales (NV-BAS) (Elliott, Murray, & Pearson, 1978), which tests the ability to name pictured objects; the Grammatical Closure subtest from the Illinois Test of Psycholinguistic Abilities (GC-ITPA) (Kirk, McCarthy, & Kirk, 1968), which tests production of morphology; and the Action Picture Test (APT) (Renfrew, 1988) and the Bus Story (revised version; Renfrew, 1991), which provide measures of grammatical structure and semantic content of expressive language. Although the scoring on these latter two tests may be criticized on a theoretical basis (e.g., their definitions of linguistic criteria) they do seem to identify the subgroup of SLI children that we are interested in (see also Bishop & Edmundson, 1987, for the tests’ prognostic value). The expressive responses from the APT and Bus story were audio-recorded on a Sony DAT recorder using an Electret condenser microphone (ECM-959), positioned approximately 20 cm to the side of the child’s mouth. Detailed transcriptions were made from these recordings. The recordings provided a further means of checking the children’s articulatory ability and intelligibility. Three of the children did not meet the articulation ability criterion and were eliminated from the group.

A grammatical and morphological versus lexical–semantic distinction was made broadly and was taken to reflect modular versus central system abilities. Children were only included in the study if they generally showed a greater impairment in comprehension and expression of grammatical and morphological abilities than their lexical–semantic (vocabulary) abilities and ‘‘information content’’ as measured on the six tests. The absolute scores (i.e., standard deviations (SD) or, if SD were not available for the test, equivalent age scores) were judged to be of lesser importance than the pattern of the scores across the tests. This is because when testing using standardized tests of language, SLI children may show only a mild or insignificant impairment in their overall scores as the tests often tap a range of structures, some of which cause difficulties for the children and some of which do not. Although this causes problems for the modular–central processing distinction, such a distinction can still be made broadly in relation to the tests. The test scores may also be influenced by language remediation therapy which may have provided the child with a nonlinguistic strategy to achieve a correct response. Therefore, the following details must be taken as a guide only, and all test scores should be judged in relation to the child’s previous history of performance on the test in question.

The SLI children included in the subgroup generally scored at least \(-1.5\) SD (though some scored up to \(-2.5\) SD) below that expected for their chronological age on the test of grammatical comprehension and up to \(-5.5\) SD on the test of expressive morphology. The children generally had an equivalent age score at least 3 years below their chronological age on tasks tapping grammatical ability, but many age equivalent scores fell well below this minimum criterion. For example one SLI child, MP, who had a chronological age of 12:10, had an equivalent age score for ‘‘subordinate clauses’’ on the Bus Story of 4:2 years. Full details of the test scores for each SLI child in this group can be found in Appendix A.

Two further nonstandardized criteria were used which have proved to be good indicators of children with Grammatical SLI. The first was that the children were to show inflectional morphological errors of subject–verb agreement and tense marking. To be included in the group the children had to omit the third person singular number agreement on the verb (e.g., \(He\) go _to work_) and/or make tense errors in the story-telling task (e.g., \(He\) _jumping_) at least 15% of the time (see van der Lely, 1996, for further details).

Second, the children were to have an impairment in the comprehension of semantically reversible sentences. A picture pointing test comprising 48 reversible sentences designed to test active, full passive, and short progressive passive sentences (e.g., \(The\ fish\ is\ being\ eaten\)), and short ambiguous (potentially adjectival) sentences (e.g., \(The\ fish\ is\ eaten\)) was administered (see van der Lely, 1993, for further details). Children were only included in the subgroup if they made some ‘‘reversal errors’’ on the full passive sentences (i.e., assigned the subject and object to the agent and theme thematic roles, respectively). Previous research has shown that reversal errors in assigning thematic roles to syntactic structures in full passive sentences are a reliable and consistent finding with Grammatical SLI children (van der Lely & Dewart, 1986; van der Lely & Harris, 1990; van der Lely, 1994a). Normally developing children of 7:5 to 8:9 performed near ceiling on this test of active and passive sentences and had a mean
Subject Details: Chronological Ages and Raw Scores from the Four Standardized Tests for the Grammatical SLI Children and Three Groups of Younger Control Children Developing Language Normally

<table>
<thead>
<tr>
<th>Subjects</th>
<th>SLI children (N = 12)</th>
<th>LA1 controls (N = 12)</th>
<th>LA2 controls (N = 12)</th>
<th>LA3 controls (N = 12)</th>
<th>Summary of analysis between groups</th>
</tr>
</thead>
<tbody>
<tr>
<td>Chronological age</td>
<td>11.2 (1.1)</td>
<td>5.9 (0.4)</td>
<td>6.11 (0.4)</td>
<td>7.11 (0.5)</td>
<td></td>
</tr>
<tr>
<td>Range</td>
<td>9.3–12:10</td>
<td>5.5–6.4</td>
<td>6.5–7.4</td>
<td>7.5–8.9</td>
<td></td>
</tr>
<tr>
<td>TROG</td>
<td>13.08 (1.78)</td>
<td>12.58 (2.35)</td>
<td>16.00 (1.75)</td>
<td>17.33 (1.23)</td>
<td>LA1 = SLI* &lt; (LA2 = LA3)</td>
</tr>
<tr>
<td>GC-ITPA</td>
<td>20.00 (3.56)</td>
<td>21.25 (3.16)</td>
<td>26.25 (4.08)</td>
<td>28.91 (2.19)</td>
<td>LA1 = SLI* &lt; (LA2 = LA3)</td>
</tr>
<tr>
<td>BPVS</td>
<td>78.83 (8.93)</td>
<td>56.25 (8.91)</td>
<td>71.67 (9.71)</td>
<td>80.00 (9.62)</td>
<td>LA1 &lt;* SLI = (LA2 &lt;* LA3)</td>
</tr>
<tr>
<td>NV-BAS</td>
<td>17.91 (1.17)</td>
<td>15.67 (1.61)</td>
<td>17.17 (1.27)</td>
<td>17.50 (0.90)</td>
<td>LA1 &lt;* SLI = (LA2 = LA3)</td>
</tr>
</tbody>
</table>

* Significant at p < .050.

From the original group of 61 SLI children, 12 children (19.6%) met the selection criteria for inclusion in the subgroup of Grammatical SLI children. There were 10 boys and 2 girls. Thus, unlike the SLI family reported by Hurst et al. (1990) and Gopnik (1990), who showed equal numbers of impaired males and females, this subgroup has a preponderance of males. On the surface, the gender bias of the sample of SLI children in this study concurs with data from undifferentiated groups of SLI children (e.g., Tallal et al., 1989a,b). However, the gender bias may be taken to reflect the bias in the sample from which the Grammatical SLI children were selected: there were 47 boys and 14 girls.

It is evident from the small number of children who met the criteria of Grammatical SLI that the prevalence of this very specific persisting language impairment is low. The findings from this study indicate that in the relatively few children in whom SLI persists, only a few (less than 20%) may be classified as having Grammatical SLI.

The subgroup of children in this study had a mean chronological age of 11:3 (range 9:3–12:10). Table 1 provides a summary of the overall subgroup’s subject details compared with three groups of younger normally developing children on four of the standardized language tests.

**FAMILIAL HISTORY OF LANGUAGE IMPAIRMENT**

To investigate whether there was evidence of a familial aggregation of language impairment in the selected subgroup of 12 Grammatical SLI children, familial history of language impairment was determined in two ways: (1) data were collected from interviews with the SLI children’s parents, and (2) a questionnaire was sent to the parents of the SLI children and a large group of age-matched control children who were developing language normally.
Interview Data

The interview data aimed to assess whether the incidence of a general or nonspecified LI in the relatives of the Grammatical SLI probands was greater than could be expected. In order to do this, two estimates were made. One was related to the prevalence of language impairment (LI) in the general population: For this study, LI was estimated to be found in 4% of the population. This was felt to be a conservative estimate as previous research has reported a prevalence of LI in 3% of first-degree relatives of children developing normally (Neils & Aram, 1986; Tomblin, 1989). However, other studies have reported that SLI occurs in approximately 1 in 1000 children (Stevenson and Richman, 1976). This lower prevalence of SLI may reflect differences in the definition of SLI.

The second estimate considered the average size of the extended family for each proband for which information about their language development and abilities may be known. An estimate of 12 family members was made. This was erring on the side of caution as the average number of children per family in the United Kingdom is less than 2.5, and for at least some family members it could be expected that language development and history would not be known. Thus, the probability of finding 1 person in 12 people with a language impairment (LI) is \( p = .387 \). Therefore, if the prevalence of LI in this sample was random, we would expect that there would be 4.644 probands with a positive familial history of LI. In other words, the probability of nobody reporting a positive familial LI history is \( p = 1 - (.96^{12}) \).

Procedure. For each child a formal interview was routinely carried out with the mother or both parents on admittance to the school for SLI children. Details of any speech, language, reading, or writing difficulties and whether or not remediation had been received for any family members of the proband was recorded in the case history notes. Data for the extended family of each proband were available. For the purposes of this study, first-degree relatives (mother, father, siblings) and second-degree relatives (uncles, aunts, and cousins) were included. Grandparents were not included as these data were only available for some of the SLI probands.

A positive family history of language impairment was defined as a reported speech, language, reading, or writing problem which required remedial help. Reading and writing problems were included in the definition as recent evidence has shown that impairments in reading and writing may be indicative of an underlying language disorder (Hulme and Snowling, 1992). Relatives with general developmental delay or acquired impairment in cognitive or motor abilities were not included.

Results. For the 12 Grammatical SLI children, 9 children had a positive familial history of language impairment. There were a total of 13 relatives of the probands with a history of language impairment. Seven of these were first-degree relatives and 6 were second-degree relatives.
Based on the estimates of general LI in the population and the size of the extended family given above, the data from first and second degree relatives were further analyzed. A $\chi^2$ test revealed that the incidence of a positive familial history was significantly more than could be expected by chance ($\chi^2 = 6.665, p < .01$).

However, caution should be made in the interpretation of these data. Although, in the context of this study and previous studies, the estimates of the family size and the incidence of LI in the population are reasonable, they do not have to vary very much for a different picture to emerge. For example, if the incidence of a positive LI history was 11% in the population one may expect to find 9 out of 12 probands with a positive LI history by chance. These factors emphasize the difficulties of using data of this nature and indicate that clinical impressions of a high incidence of familial LI history can be misleading.

A more reliable method of obtaining preliminary evidence of positive familial history of LI is to use a questionnaire. This has the advantage that control data can be obtained from the relatives of children developing normally and compared with the grammatical SLI probands. Therefore, a survey using a questionnaire was carried out.

**Questionnaire**

The comparison between general LI in the relatives of the SLI and age-matched probands aimed to provide more detailed information about the pattern of LI impairment as well as the incidence of general LI in the two groups. In an attempt to assure reliability of the data obtained from the questionnaire, only information about first-degree relatives was sought.

**Procedure.** A questionnaire with a cover letter and a stamped envelope for the reply was sent to the parents of each of the 12 Grammatical SLI children. The questionnaire was also sent to the parents of 150 normally developing children (control probands) to obtain comparable data of the general prevalence of a positive familial history of LI in the UK population.

The control children were selected from two schools; one in central London and one in a rural town. All the children from these two schools whose chronological ages fell within the age range of the SLI children (i.e., 9:3 to 12:10 years) were included in the group.

A sample questionnaire which had been filled in was enclosed to try to avoid any ambiguity of what was required. The parents were required to write down the child’s name and to list all first-degree relatives (parents and siblings) and answer four questions for the child and each listed family mem-

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1 However, note that the data from the questionnaire to be reported below revealed an 8.62% incidence of SLI in the population sample. Therefore, based on these data, 9 out of 12 SLI probands with a positive familial LI history is still significantly different from chance.
ber on a prepared form. The questions were: (1) Has she/he had any speech or language problems? (2) Has she/he ever received any speech or language therapy? (3) Has she/he had any reading or writing problems? (4) Has she/he ever had any other help for a speech, language, reading, or writing problem? The parent had to circle “yes/no/don’t know” answers as appropriate. An additional column for “Any other comments?” was provided.

To increase the return of the questionnaires a reminder letter was sent home with the probands 10 days after the original questionnaire had been sent and the probands’ teachers verbally reminded the children that their parents should return the questionnaire.

Results. For the SLI probands, 9 of the 12 questionnaires (75%) were returned. For the three SLI children whose questionnaires were not returned the above case history information had revealed that one had a positive familial history of LI and two a negative familial history of LI. The data for the SLI probands included two half-sisters and three half-brothers of one of the probands (i.e., the father’s children by another marriage). One of these half-brothers had a positive history of reading and writing problems. There were a total of 31 family members for which data were available, i.e., the yes or no answers had been appropriately circled. This gave an average number of approximately 3.5-first degree relatives for each proband.

For the control probands, 72 of the 150 questionnaires (48%) were returned. Twenty-three of these questionnaires could not be used. This was largely because of some reference to speech/language or reading/writing problems or omission of this information for the control proband. The problems noted for the proband, however, were often minor, e.g., “had speech therapy for a lisp.” Data for the remaining 49 questionnaires were analyzed.

A familial history of LI was considered to be positive if it was reported that a particular family member had a history of a speech or language problem and speech therapy, a reading or writing problem and remedial reading or writing help, or a history of speech, language, reading, and writing problems for which they had received remedial help. A summary of the results of the questionnaire for the SLI probands and the control probands can be found in Tables 2 and 3, respectively.

Tables 2 and 3 indicate that on virtually every measure (e.g., total proportion of positive familial LI history, overall proportion of positive speech/language history or reading/writing history), the SLI probands’ first-degree relatives show a substantially higher incidence of a positive LI history. The following analyses confirmed these observed differences between the Grammatical SLI probands and control probands.

Twelve of the 31 relatives of the Grammatical SLI probands had a positive family history (38.7%). For the control probands, 15 of the 174 relatives (8.62%) had a positive LI history. Thus, there was proportionally almost 4.5 times as many relatives in the SLI proband families who had a positive LI history than in the families of the control probands.
TABLE 2
Results of the Questionnaire: Familial History of Language Impairment for First Degree Relatives of the Grammatical SLI Children

<table>
<thead>
<tr>
<th>Relative</th>
<th>Total reported number</th>
<th>Number affected</th>
<th>Speech/language</th>
<th>Reading/writing</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mothers</td>
<td>9</td>
<td>3 (33.3%)</td>
<td>3 (33.3%)</td>
<td>3 (33.3%)</td>
</tr>
<tr>
<td>Fathers</td>
<td>8</td>
<td>3 (37.5%)</td>
<td>0 (0%)</td>
<td>3 (37.5%)</td>
</tr>
<tr>
<td>Sisters</td>
<td>5</td>
<td>2 (40%)</td>
<td>2 (40%)</td>
<td>0 (0%)</td>
</tr>
<tr>
<td>Brothers</td>
<td>9</td>
<td>4 (44%)</td>
<td>0 (0%)</td>
<td>4 (44%)</td>
</tr>
<tr>
<td>Total</td>
<td>31</td>
<td>12 (38.7%)</td>
<td>5 (16.13%)</td>
<td>10 (32.26%)</td>
</tr>
</tbody>
</table>

Note. Number of questionnaires returned, 9 (75%); number of probands with a positive speech and language history, 4 (44.5%); number of probands with a positive reading and writing history, 6 (66.7%); total number of probands with a positive family history of LI, 7 (77.8%). A positive history of LI is a reported speech/language impairment plus speech therapy, and/or a reading/writing impairment plus remedial help.

For the 9 SLI probands for which data were available, 7 (77.8%) had a positive family history. That is, 7 of the probands had one or more first-degree relatives with a positive LI history. For the 49 control probands, 14 (28.57%) had a positive familial history. A $\chi^2$ test was carried out to compare the proportions of the incidence of a positive familial history between the two groups. This revealed a highly significant difference, reflecting the higher incidence of a positive familial LI history for the SLI probands, $\chi^2 = 20.83$, $p = .000005$.

It was noted that only one (2.04%) of the control probands had more than one member of the family with a positive LI history. In this family both of

TABLE 3
Results of the Questionnaire: Familial History of Language Impairment for First Degree Relatives of the Control Children

<table>
<thead>
<tr>
<th>Relative</th>
<th>Total reported number</th>
<th>Number affected</th>
<th>Speech/language</th>
<th>Reading/writing</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mothers</td>
<td>49</td>
<td>1 (2.04%)</td>
<td>1 (2.04%)</td>
<td>1 (2.04%)</td>
</tr>
<tr>
<td>Fathers</td>
<td>48</td>
<td>4 (8.33%)</td>
<td>1 (2.08%)</td>
<td>3 (6.25%)</td>
</tr>
<tr>
<td>Sisters</td>
<td>40</td>
<td>3 (7.50%)</td>
<td>1 (2.50%)</td>
<td>2 (5.00%)</td>
</tr>
<tr>
<td>Brothers</td>
<td>37</td>
<td>7 (18.92%)</td>
<td>5 (13.51%)</td>
<td>3 (8.12%)</td>
</tr>
<tr>
<td>Total</td>
<td>174</td>
<td>15 (8.62%)</td>
<td>8 (4.60%)</td>
<td>9 (5.17%)</td>
</tr>
</tbody>
</table>

Note. Number of questionnaires returned, 72 (48.0%); number of usable questionnaires, 49 (32.66%); number of probands with a positive speech and language history, 7 (14.29%); number of probands with a positive reading and writing history, 7 (14.29%); number of probands with a positive family history of LI, 14 (28.57%).
the probands' parents were hearing impaired. This accounted for the one mother in this group who had a positive LI history. For the SLI group, 3 (33.3%) of the probands had more than one member with a positive LI history.

Further analyses were carried out to assess the overall pattern of impairment in the different family members and to see whether the increased incidence of a positive LI history was evident for each of the family members (i.e., mother, father, brother, sister).

For the SLI probands, 3 (33.3%) of the mothers had a positive LI history in comparison to 1 (2.04%) of the mothers of the control probands who had a positive LI history. This difference was significant, $\chi^2 = 11.596, p < .001$. For the fathers, 37.5 and 8.33% of the SLI and control probands, respectively, had a positive LI history. Again, analysis revealed that this difference was significant, $\chi^2 = 5.333, p < .025$.

Significantly more of the SLI probands' sisters than control probands' sisters (40 vs. 7.5%) were also found to have a positive LI history, $\chi^2 = 4.753, p < .05$. Although 44% of the SLI probands' brothers had a positive LI history in comparison to only 18% of the control probands' brothers, this difference was not significant, $\chi^2 = 2.592, p > .05$. This result reflects the increased incidence of a positive LI history of the brothers for the control probands.

A comparison of the numbers of brothers and sisters with a positive history for the SLI probands (see Table 2) shows a similar proportion of impairment in male and female siblings. In contrast, Table 3 indicates that the control probands had a gender bias toward more impaired boys than girls, which is frequently reported for the normal population (Vogel, 1990). A further analysis confirmed that there was a significant gender bias between the impaired brothers and sisters among the control probands, $\chi^2 = 7.294, p < .01$. Thus, these data from the control probands concur with previous findings which have shown that in the normal population and in undifferentiated groups of SLI children more boys may have speech/language and or reading/writing problems than girls (Tallal et al., 1989b; Vogel, 1990).

An analysis of the total proportion of males versus females with a positive LI history for the SLI and control probands revealed a significant gender ratio difference between the two groups, $\chi^2 = 3.938, p < .05$. This reflects the generally higher number of impaired males than females in the control probands' parents and siblings which is not evident for the grammatical SLI probands. This point will be taken up further under Discussion.

Finally, it was noted for the SLI probands that all three mothers who had a positive family history reported both speech-language and reading-writing problems. These three mothers also reported long histories of speech therapy. In contrast, only reading-writing problems were reported for the fathers. This male/female difference may reflect a bias in the method of collecting the data. Typically, it was the mother who filled in the questionnaire. How-
ever, the same male/female bias in reading–writing/speech–language problems was reflected in the sibling data. The four brothers with a positive family history had reading–writing impairments, while the two sisters had speech–language impairments. Clearly, further investigations are warranted to ascertain whether the male family members have or had evidence of speech–language problems.

**DISCUSSION**

This study was concerned with extending our existing knowledge concerning familial aggregation of LI in SLI probands. Previous research has focused largely on undifferentiated groups of SLI children. This study is the first (to our knowledge) to investigate whether there is evidence of familial aggregation of LI in a subgroup of Grammatical SLI children who appear to have a modular language impairment. The incidence of a positive familial history of the Grammatical SLI probands and age-matched control probands who were developing language normally was compared.

The results of the questionnaire revealed that the SLI probands had a significantly higher incidence of one or more first-degree relatives with a positive familial LI history than the control probands; 77.8% of the SLI probands had a positive familial LI history in comparison to only 28.57% of the control probands. It should be noted, however, that questionnaire data are subject to selection bias. For example, it is possible that the parents of control children in which there was a familial LI history may have been less likely to respond. In addition, having a child with SLI may make parents more likely to be aware of LI in siblings and other family members.

However, the findings from the questionnaire are supported by the case history information. A significantly higher positive familial history than could be expected, based on the incidence of general LI in the population, was revealed. There were very few cases in which a positive familial history was reported in only one of the two data sets for a particular SLI child. In one case a positive familial LI history was revealed by the questionnaire which had not been previously reported (two brothers with reading and writing problems). The questionnaire revealed a further two sisters and one father who had a LI history in addition to other LI family members which had been reported in the case history notes.

Thus, the data from this study are consistent with a genetic basis underlying Grammatical SLI in children. However, the data do not give any indication as to the nature of the LI in the SLI probands relatives. This point will be taken up later under General Discussion.

The results of the analysis of the incidence of a positive familial LI history in the four family members (mothers, fathers, brothers, and sisters) provide further insight into the possible nature of the genetic inheritance of Grammatical SLI in children. Specifically, from these data we may distinguish be-
patterns of LI that are consistent with recessive and dominant genetic inheritance, and between autosomal and sex-linked inheritance.

The pattern of familial impairment found in this study for the grammatical SLI probands provides an initial indication that the genetic inheritance is an autosomal dominant inheritance. For the SLI probands, it was either the mother or the father who had a positive history, rather than both parents; that is, 70.58% of the SLI probands had one affected parent. In addition, it could be expected that if Grammatical SLI was caused by an autosomal (non-sex linked) dominant genetic abnormality then approximately 50% of the siblings would have a positive LI history. The results from the questionnaire show that close to 50% (i.e., approximately, 43%) of the probands’ siblings have a positive history.

It would appear from the results that the gene(s) which may cause grammatical SLI is not sex linked. An equal incidence of a positive history was generally found for the overall male and female first-degree relatives and there was not a significant difference between the incidence of LI brothers and sisters among the SLI probands. These data are particularly striking when compared with the findings for the control probands and previous studies of undifferentiated groups of SLI children. The data from the control probands in this study showed a significant gender bias with more males than females with a LI history. Tomblin (1989) found a very high rate of impaired brothers (39.9%) in comparison with impaired sisters (16.67%). A similar gender ratio bias was reported by Haynes and Naidoo (1991) and was found generally in Bishop et al.’s (1995) twin study. Thus, the findings from this study, although preliminary, suggest that a different genetic basis could underlie Grammatical SLI and some other forms of SLI.

General Discussion

The findings from this study support previous research which has shown a familial aggregation of LI in undifferentiated groups of SLI probands (Bishop & Edmundson, 1986; Neils & Aram, 1986; Tallal et al., 1989a,b; Tomblin, 1989) and the SLI family (Hurst et al., 1990; Gopnik, 1990). This study extends these previous findings. The data are consistent with a genetic basis underlying the form of language impairment characterized by Grammatical SLI. The results from this study for the subgroup of Grammatical SLI children may be contrasted with the subgroup of expressive SLI children who did not appear to have a positive familial history (Whitehurst et al., 1991). The difference between the findings from Whitehurst et al. and this study highlights that the same surface behavior in expressive language (e.g., an impairment in inflectional morphology) may be found in different subgroups of SLI children who may not be characterized by the same underlying deficit.

There are also interesting differences between the results of this study and
those of previous studies in which the findings are consistent with a genetic basis to SLI. The data tentatively suggest that a different genetic inheritance may underlie different forms of SLI.

This may be partially revealed in a comparison of the results from this study with those of Haynes and Naidoo (1991). The Grammatical SLI children in this study were a subgroup of the SLI population studied by Haynes and Naidoo. For this global population of SLI children, Haynes and Naidoo found that 41% of the children had a positive history. Further analysis revealed that the Grammatical SLI probands had a significantly higher incidence of a positive history than the overall SLI population from which they were selected ($\chi^2_{(1)} = 5.547, p < .02$).

Stronger evidence that the genetic basis underlying Grammatical SLI is different from some other forms of SLI is provided by the apparent autosomal pattern of inheritance found for this subgroup. While the lack of a sex-linked inheritance concurs with Hurst et al.’s (1990) investigation of the SLI family, it contrasts with the greater number of LI males in both the control population in this study and many previous investigations of mixed groups of SLI probands.

The results from this study, like those of Gopnik and Crago (1991), support the notion of a genetic disorder underling the acquisition of grammatical knowledge which is needed for normal language development. There is, however, some dispute as to whether the SLI family studied by Gopnik and Crago (1991) and by Vargha-Khadem and Passingham (1990) suffer from nonlinguistic cognitive deficits which can explain or are associated with their language impairments (Bates, 1993). In contrast to this SLI family, no nonlinguistic deficits have been revealed for the Grammatical SLI children in our study. Thus, the linguistic impairment of these subjects does not appear to be associated with any other disorder. This makes the data from the present study particularly interesting as the findings are consistent with the possibility of a genetic basis underlying a modular language deficit.

However, caution is expressed with the interpretation of the findings from this study. It should be stressed that this investigation is a first step in exploring a possible genetic basis to Grammatical SLI. While the results are consistent with a genetic explanation, they are not conclusive. From the data so far, environmental explanations, although unlikely (see Bishop et al., 1995) cannot be ruled out (cf. Rutter et al., 1990; Lewis et al., 1993).

The data from this study do not identify the nature of the LI exhibited by the SLI probands’ relatives. It would be of interest to know whether the LI of the relatives could also be classified as a Grammatical SLI. Further investigations of the LI relatives of the Grammatical SLI probands using specific tests of grammatical knowledge are required. Such investigations, providing valuable evidence of the relationship of the LI of the Grammatical SLI probands and their relatives, may help to determine whether we are
indeed looking at distinct disorders in the SLI population, or which forms of SLI are genetically related.

In addition, it should be noted that the numbers of Grammatical SLI children in this study were small. The data from this investigation indicate that further research is warranted with both Grammatical SLI children and other carefully defined subgroups of SLI children to substantiate the findings from this investigation. For example, genetic linkage studies using genetic markers may be employed alongside an investigation of the linguistic nature of the LI in SLI probands’ relatives. Such research may provide further evidence to validate the proposal that there is a genetic basis to Grammatical SLI in children.

Finally, this investigation of Grammatical SLI children, which is consistent with an autosomal dominant genetic inheritance underlying their disorder, although preliminary in nature, provides an exciting source of evidence supporting a biological, domain-specific, and modular basis to language.
## APPENDIX A

Raw Scores, z Scores, or Standard Scores and Equivalent Age Score for the Matching and Selection Language Tests for Individual SLI Children

<table>
<thead>
<tr>
<th>Subject</th>
<th>Chronological age</th>
<th>BPVS Raw score (z score)</th>
<th>Equivalent age</th>
<th>TROG Raw score (z score)</th>
<th>Equivalent age</th>
<th>NV-BAS Raw score (SS)</th>
<th>Equivalent age</th>
<th>GC-ITPA Raw score (z score)</th>
<th>Equivalent age</th>
</tr>
</thead>
<tbody>
<tr>
<td>JW</td>
<td>9:3</td>
<td>60 (−1.7)</td>
<td>6:5</td>
<td>10 (−2.2)</td>
<td>5:3</td>
<td>17*</td>
<td>7:9</td>
<td>17 (−3.7)</td>
<td>6:0</td>
</tr>
<tr>
<td>WL</td>
<td>9:5</td>
<td>72 (−0.9)</td>
<td>7:9</td>
<td>12 (−1.7)</td>
<td>5:9</td>
<td>17*</td>
<td>7:9</td>
<td>18 (−3.8)</td>
<td>6:3</td>
</tr>
<tr>
<td>JS</td>
<td>9:10</td>
<td>89 (0.0)</td>
<td>9:9</td>
<td>13 (−1.5)</td>
<td>6:0</td>
<td>19*</td>
<td>&gt;7:11</td>
<td>17 (−4.6)</td>
<td>6:0</td>
</tr>
<tr>
<td>AZ</td>
<td>10:3</td>
<td>72 (−1.3)</td>
<td>7:9</td>
<td>12 (−1.9)</td>
<td>5:9</td>
<td>19*</td>
<td>&gt;7:11</td>
<td>16 (−5.5)</td>
<td>5:10</td>
</tr>
<tr>
<td>RJ</td>
<td>10:11</td>
<td>76 (−1.4)</td>
<td>8:2</td>
<td>16 (−0.8)*</td>
<td>9:0</td>
<td>19*</td>
<td>&gt;7:11</td>
<td>16*</td>
<td>5:10</td>
</tr>
<tr>
<td>AZ</td>
<td>11:0</td>
<td>72 (−1.7)</td>
<td>7:9</td>
<td>12 (−2.1)</td>
<td>5:9</td>
<td>18*</td>
<td>&gt;7:11</td>
<td>24*</td>
<td>7:11</td>
</tr>
<tr>
<td>CT</td>
<td>11:11</td>
<td>86 (−1.1)</td>
<td>9:0</td>
<td>13 (−2.2)</td>
<td>6:0</td>
<td>18*</td>
<td>&gt;7:11</td>
<td>21*</td>
<td>7:0</td>
</tr>
<tr>
<td>SB</td>
<td>12:0</td>
<td>90 (−0.7)</td>
<td>9:5</td>
<td>15 (−1.6)</td>
<td>8:0</td>
<td>17*</td>
<td>7:9</td>
<td>24*</td>
<td>7:11</td>
</tr>
<tr>
<td>AT</td>
<td>12:1</td>
<td>80 (−1.6)</td>
<td>9:0</td>
<td>13 (−2.2)</td>
<td>6:0</td>
<td>16*</td>
<td>6:3</td>
<td>17*</td>
<td>6:0</td>
</tr>
<tr>
<td>BS</td>
<td>12:2</td>
<td>78 (−1.8)</td>
<td>8:5</td>
<td>12 (−2.5)</td>
<td>5:9</td>
<td>20*</td>
<td>&gt;7:11</td>
<td>22*</td>
<td>7:3</td>
</tr>
<tr>
<td>AW</td>
<td>12:2</td>
<td>84 (−1.5)</td>
<td>9:3</td>
<td>16 (−1.2)*</td>
<td>9:0</td>
<td>17*</td>
<td>&gt;7:11</td>
<td>22*</td>
<td>7:3</td>
</tr>
<tr>
<td>MP</td>
<td>12:10</td>
<td>87 (−1.4)</td>
<td>7:9</td>
<td>13 (−2.2)</td>
<td>6:0</td>
<td>18*</td>
<td>&gt;7:11</td>
<td>26*</td>
<td>8:6</td>
</tr>
</tbody>
</table>

Note. BPVS, British Picture Vocabulary Score; TROG, Test of Reception of Grammar; NV-BAS, Naming Vocabulary, British Ability Scales; GC-ITPA, Grammatical Closure subtest from Illinois Test of Psycholinguistic Abilities; SS, Standard Score.

* On the basis of the scores for RJ and AW, they would not have been included in the group. However, previous scores on this test for both children showed a greater deficit in relation to their BPVS scores. It appeared that the TROG score obtained above represented a sudden improvement on this test. This may have resulted from the remedial help they were receiving at this time which was directed at improving the performance on particular structures which were assessed in this test.

* SS or z score not available.
<table>
<thead>
<tr>
<th>Subject</th>
<th>Chronological age</th>
<th>Info (age)</th>
<th>Sent. length (age)</th>
<th>Subclause (age)</th>
<th>Total % Full passives</th>
<th>Test of Active and Passive Sentences</th>
</tr>
</thead>
<tbody>
<tr>
<td>JW</td>
<td>9:03</td>
<td>28 (6:1)</td>
<td>14 (8:2)</td>
<td>2 (4:8)</td>
<td>34 (6:9)</td>
<td>23 (5:3)</td>
</tr>
<tr>
<td>WL</td>
<td>9:05</td>
<td>23 (5:1)</td>
<td>10 (6:4)</td>
<td>1 (4:2)</td>
<td>26.5 (4:2)</td>
<td>20 (4:3)</td>
</tr>
<tr>
<td>JS</td>
<td>9:10</td>
<td>29 (6:4)</td>
<td>11 (6:10)</td>
<td>1 (4:2)</td>
<td>33.5 (6:6)</td>
<td>26 (6:3)</td>
</tr>
<tr>
<td>AZ</td>
<td>10:03</td>
<td>24 (5:3)</td>
<td>13 (7:10)</td>
<td>2 (4:8)</td>
<td>28 (4:8)</td>
<td>20 (4:3)</td>
</tr>
<tr>
<td>RJ</td>
<td>10:10</td>
<td>27 (5:10)</td>
<td>8 (4:7)</td>
<td>1 (4:2)</td>
<td>34.5 (7:0)</td>
<td>22 (5:0)</td>
</tr>
<tr>
<td>AZ</td>
<td>11:00</td>
<td>22 (4:11)</td>
<td>11 (6:10)</td>
<td>1 (4:2)</td>
<td>34.5 (7:0)</td>
<td>25 (6:0)</td>
</tr>
<tr>
<td>CT</td>
<td>11:11</td>
<td>33 (7:4)</td>
<td>12 (7:4)</td>
<td>2 (4:8)</td>
<td>38 (8:5)</td>
<td>24 (5:9)</td>
</tr>
<tr>
<td>SB</td>
<td>12:00</td>
<td>20 (4:7)</td>
<td>12 (7:4)</td>
<td>3 (5:10)</td>
<td>35.5 (7:6)</td>
<td>23 (5:3)</td>
</tr>
<tr>
<td>AT</td>
<td>12:01</td>
<td>29 (6:4)</td>
<td>11 (6:10)</td>
<td>1 (4:2)</td>
<td>34.5 (7:0)</td>
<td>26 (6:3)</td>
</tr>
<tr>
<td>BS</td>
<td>12:02</td>
<td>30 (6:7)</td>
<td>11 (6:10)</td>
<td>2 (4:8)</td>
<td>35 (7:3)</td>
<td>26 (6:3)</td>
</tr>
<tr>
<td>AW</td>
<td>12:02</td>
<td>25 (5:5)</td>
<td>9 (5:7)</td>
<td>2 (4:8)</td>
<td>35 (7:3)</td>
<td>25 (6:0)</td>
</tr>
<tr>
<td>MP</td>
<td>12:10</td>
<td>32 (7:1)</td>
<td>9 (5:7)</td>
<td>1 (4:2)</td>
<td>35 (7:3)</td>
<td>28 (6:9)</td>
</tr>
</tbody>
</table>

Note. Info, information score; subclause, number of subordinate clauses; (age), equivalent age score.
REFERENCES


