Genetics of Spoken Language Disorders

KARIN STROMSWOLD[1]

Abstract If language is the result of specialized structures in the brain and if these language-specific structures are genetically encoded, one would expect to find evidence of the heritability of language. In this article I review the results of family aggregation, pedigree, sex ratio, commingling, and segregation studies of spoken language disorders. The results of these studies provide evidence that, although spoken language disorders are genetically and behaviorally heterogeneous, genetic factors may play a substantial role in many cases of developmental spoken language disorders.

One of the fundamental questions about language acquisition is the extent to which the ability to learn language is the result of innate genetically encoded structures that are specific to language. A number of findings are consistent with there being a heritable component to language (e.g., findings about the universal properties of human language, normal children's rapid and automatic acquisition of language, the existence of a sensitive period for language acquisition, acquired and developmental disorders that specifically impair or spare language). In addition to such indirect evidence, if language is the result of genetically encoded language-specific structures, genetic studies should reveal evidence for the heritability of language. Here, I review family aggregation, sex ratio, and pedigree studies of language, emphasizing how results of these studies can be used to determine the extent to which language disorders are the result of genetic and environmental factors.

In the summer of 1994 I performed Boolean searches on the computerized bibliographic databases PsychLIT, ERIC, and Medline to cull any paper written in English that contained the letter strings *language, linguistic, articul,* or *speech* and the letter strings *hereditary, genetic, famil, twin, adoption, chromosom, linkage, pedigree, sex ratio, segregation, aggregation, DNA,* or *RNA* in its journal title, article title, key words, or abstract. Because of space limitations, twin studies and adoption studies are reviewed elsewhere (Strom-

[1]Department of Psychology and Center for Cognitive Science, Busch Campus, Rutgers University, New Brunswick, NJ 08903.

Human Biology, April 1998, v. 70, no. 2, pp. 297–324.

KEY WORDS: LANGUAGE, SPEECH, LANGUAGE DISORDERS, HERITABILITY, FAMILY AGGREGATION, SEX RATIO, P

swold 1996, 1997a). Papers on stuttering were not reviewed [see Ambrose et al. (1993)], nor were studies that investigated progressive or acquired language disorders or studies that investigated language impairments secondary to hearing impairment, motor dysfunction, neurological disorders, psychiatric disorders, or mental retardation. The reference sections of all papers deemed appropriate for inclusion were searched for additional studies not included in the computerized bibliographic databases. A few papers that were written after the computerized search were included if they met the criteria for inclusion.

Family Aggregation Studies

For many years researchers have reported that the incidence of developmental language disorders is higher in some families than in others and that such disorders often seem to run in certain families. According to Luchsinger (1970), examples of hereditary speech defects have been reported since ancient times, and by the 1880s researchers had begun to distinguish between hereditary forms of stuttering and dyslalia. If developmental language disorders are heritable, then the incidence of language disorders should be greater among relatives of children with language impairment (probands) than among relatives of children without language impairments (control subjects); more probands than control subjects should have a positive family history for language impairments (i.e., more probands should have at least one language-impaired relative) and a higher percentage of probands' relatives than control subjects' relatives should have a history of language impairment.

Included Studies. Eighteen studies of familial aggregation of developmental spoken language disorders met the inclusion criteria and contained descriptions of probands, control subjects, and family members that were complete enough to allow comparisons among the studies (Beitchman et al. 1992; Bishop and Edmundson 1986; Byrne et al. 1974; Haynes and Naidoo 1991; Ingram 1959; Lahey and Edwards 1995; Lewis 1992; Lewis et al. 1989; Luchsinger 1970; Neils and Aram 1986; Rice et al. 1998; Tallal et al. 1989a; Tomblin 1989, 1996a; Tomblin and Buckwalter 1994; Tomblin et al. 1991; van der Lely and Stollwerck 1996; Whitehurst et al. 1991).

Proband Diagnosis. In part because the studies were conducted at different times (between 1959 and 1996), the diagnoses of language impairment given to the probands varied considerably (see Table 1). Despite the differences in diagnoses, descriptions of the probands suggest that most if not all would meet current diagnostic criteria for specific developmental language disorders [e.g., Stark and Tallal (1981) and American Psychiatric Association (1994)], although the probands do not appear to suffer from the same type of

disorder. Some of the studies include children with a wide range of speech and language disorders (Ingram 1959; Neils and Aram 1986; Bishop and Edmundson 1986; Lahey and Edwards 1995), whereas other studies include children who suffer only from grammatical disorders (Rice et al. 1998; van der Lely and Stollwerck 1996) or phonological disorders (Lewis et al. 1989; Lewis 1992). [See Stromswold (1997b) for a review of classification systems for developmental language disorders.] The methods used to locate probands varied, with some studies recruiting volunteers specifically for the study (Whitehurst et al. 1991), some drawing from clinical case loads or referrals (Bishop and Edmundson 1986; Byrne et al. 1974; Haynes and Naidoo 1991; Ingram 1959; Lahey and Edwards 1995; Lewis 1992; Lewis et al. 1989; Luchsinger 1970; Neils and Aram 1986; Rice et al. 1998; Tallal et al. 1989a; Tomblin 1989; Tomblin and Buckwalter 1994; van der Lely and Stollwerck 1996), and others screening a well-defined group of children, such as all kindergarten students in a school district (Beitchman et al. 1992; Tomblin 1989, 1996a; Tomblin et al. 1991). As shown in Table 1, children were diagnosed as having a developmental language disorder based on clinical history and/or performance on standardized tests.

Family Diagnosis. What counted as evidence of language impairment in family members varied considerably from study to study (see Table 1). Some studies considered relatives with a history of only spoken language disorders to be affected, whereas other studies also included relatives who had a history of dyslexia, nonlinguistic learning disabilities, or poor school performance. Some studies investigated only whether first-degree family members were affected, whereas other studies included more distant family members. In most studies presence of language disorders in family members was determined by asking a parent. Two studies (Bishop and Edmundson 1986; Tomblin 1989) counted a family member as being language impaired only if the family member had received speech or language therapy or still had a readily apparent language impairment; one study (Lewis et al. 1989) tested siblings of probands and control subjects, and one study (Tomblin and Buckwalter 1994) tested all nuclear family members and some extended family members to determine whether they were language impaired.

Results

Family History. As shown in Table 2, all but 4 (Lewis 1992; Lewis et al. 1989; Rice et al. 1998; Whitehurst et al. 1991) of the 18 studies reported the percentage of proband children with positive family histories of impairments. In most of these studies a child was considered to have a positive family history if at least one first-degree family member (i.e., parent or sibling) was impaired. In some studies positive family history was defined as impairment in any family member, and in other studies positive family history was defined

Table 1. Family Aggregation Studies of Spoken Language Disorders

Study	*Sample Size*	*Proband Diagnosis*	*Proband Ascertainment*	*Other Family Diagnoses*
Ingram (1959)	75 probands	Developmental speech/ language disorder	Clinic records	None
Luchsinger (1970)	127 probands	Developmental speech retardation	Clinic records	None
Byrne et al. (1974)	18 severely impaired, 20 moderately impaired	Delayed speech	Clinic records	None
Neils and Aram (1986)	74 probands, 36 controls	Developmental language disorder	Clinic and school referrals (tested)	Dyslexia, stuttering, articulation
Bishop and Edmundson (1986)	34 probands, 131 controls	Developmental language disorder		None (for strict criteria)
Lewis et al. (1989)	20 probands, 20 controls	Phonologic disorder	Clinic referral (tested)	Dyslexia, stuttering, LD
Tallal et al. (1989a)	62 probands, 50 controls	SLI	Clinician and school referrals (tested)	Dyslexia, LD, school problems
Tomblin (1989)	51 proband, 136 controls	Receiving speech or language therapy	Clinician referral	Stuttering, articulation
Haynes and Naidoo (1991)	156 probands	SLI	SLI schools (tested)	None
Tomblin et al. (1991)	55 probands, 607 controls	History (5) or bottom 10% on language use questionnaire (50)	Parental report of large cohort of children	None
Whitehurst et al. (1991)	62 probands, 55 controls	Isolated expressive language delay	Newpaper advertisements (tested)	Speech, late talker, school problems
Beitchman et al. (1992)	136 probands, 138 controls	Speech, language, or voicing disorder or stuttering	Kindergarten screening test and standard tests	Dyslexia, LD, articulation
Lewis (1992)	87 probands, 79 controls	Phonologic disorder with or without language disorder	Clinician referral and clinic records (many tested)	Dyslexia, LD, stuttering, hearing loss
Tomblin and Buckwalter (1994)	26 probands	SLI	Clinic and school referral (tested)	None
Lahey and Edwards (1995)	53 probands	SLI	Clinican referral (tested)	Learning problems
Rice et al. (1998)	31 probands, 67 controls	SLI (extended optional infinitive subtype)	Clinician referral (tested)	Reading, spelling, learning
Tomblin (1996a)	534 probands, 6684 controls	SLI	Screening and diagnostic tests	
van der Lely and Stollwerck (1996)	9 probands, 49 controls	SLI (grammatical subtype)	SLI schools (tested)	Reading or writing

SLI, specific language impairment.
LD, Learning disability.

Table 2. Results of Family Aggregation Studies of Spoken Language Disorders[a]

Study	*Positive Family History*	*Frequency of Impairment*
Ingram (1959)	24% Parental history, 32% sibling history	NA
Luchsinger (1970)	36% Probands	NA
Byrne et al. (1974)	17% "Severe" probands, 55% "moderate" probands[c]	NA
Neils and Aram (1986)	46% first-degree probands, 8% first-degree controls[e]	20% all proband vs. 3% all control relatives[d]
Bishop and Edmundson (1986)	24% first-degree probands, 3% first-degree controls[e]	NA
Lewis et al. (1989)	NA	Any: 12% all probands, 2% all controls,[e] 26% vs. 5% for first-degree[d] SLI: 9% all probands; 1% all controls[c]
Tallal et al. (1989a)	77% first-degree probands, 46% first-degree controls[c]	42% first-degree probands, 19% first-degree controls[d]
Tomblin (1989)	53% first-degree probands, controls: N/A	23% first-degree probands, 3% first-degree controls[e]
Haynes and Naidoo (1991)	54% all probands, 41% first-degree probands	28% proband parents, 18% proband sibs
Tomblin et al. (1991)	35% probands, 17% controls[d]	NA
Whitehurst et al. (1991)	NA	Any: 24% probands vs. 16% controls for first-degree Speech: 12% probands vs. 8% controls for first-degree Late talker: 12% probands vs. 7% controls for first-degree School: 7% probands vs. 5% controls for first-degree
Beitchman et al. (1992)	47% all probands, 28% all controls[d]; 34% vs. 11% first-degree[e]	19% of probands had multiple affected relative, 9% of controls had multiple affected relatives[b]
Lewis (1992)	NA	LI: 15% all probands, 2% all control relatives,[e] 32% vs. 5% for first-degree relatives[e] Dyslexia: 3% all probands, 1% all control relatives,[e] 6% vs. 3% for first-degree relatives LD: 3% all probands, 1% all control relatives,[e] 6% vs. 1% for first-degree relatives[b]
Tomblin and Buckwalter (1994)	42% first-degree	Overall, 21%; mother, 15%; father, 40%; sister, 6%; brother, 24%
Lahey and Edwards (1995)	60% first-degree	Overall, 26%; mother, 26%; father, 22%; siblings, 29%
Rice et al. (1998)	NA	Any: 18% all proband relatives, 9% controls,[d] 26% vs. 13% for first-degree relatives[e] LI: 15% all proband relatives, 6% controls,[d] 22% vs. 7% for first-degree relatives[d] Other: 7% all proband relatives, 5% controls, 12% vs. 9% for first-degree relatives
Tomblin (1996a)	29% probands, 11% controls[e]	
van der Lely and Stollwerck (1996)	78% first-degree probands, 29% first-degree controls[c]	Overall: 39% probands, 9% controls[e] Mothers: 33% probands, 2% controls[d] Fathers: 38% probands, 8% controls[b] Sisters: 40% probands, 8% controls[b] Brothers: 44% probands, 19% controls

SLI, Specific language impairment.
LI, Speech or language impairment.
LD, Learning disability.
a. Significance tests are for one-tailed tests.
b. $p < 0.05$.
c. $p < 0.01$.
d. $p < 0.001$.
e. $p < 0.0001$.

as presence of impairment in either one first-degree relative or several extended relatives (Tomblin et al. 1991). If more than one measure of family history was given, first-degree family history was used to determine mean and median rates. When rates of positive family history were given for "any impairment" and for specific types of impairments [e.g., dyslexia, specific language impairment (SLI)], the rates for "any impairment" were used. If no overall rates were given, the rates for spoken language impairment were used. For Tomblin (1996a) I calculated the rate of positive family history of SLI by combining the figures for probands with isolated SLI and probands with both SLI and speech sound disorders.

For probands the reported incidence of positive family history ranged from 24% (Bishop and Edmundson 1986) to 77% (Tallal et al. 1989a), with a mean incidence of 45% and a median incidence of 39%. The incidence of positive family history was significantly greater for probands than for control subjects in all seven studies that collected data for both probands and control subjects (Beitchman et al. 1992; Bishop and Edmundson 1986; Neils and Aram 1986; Tallal et al. 1989a; Tomblin 1996a; Tomblin et al. 1991; van der Lely and Stollwerck 1996). In these seven studies the reported incidence of positive family history for probands ranged from 24% (Bishop and Edmundson 1986) to 78% (van der Lely and Stollwerck 1996), with a mean incidence of 46% and a median incidence of 35%. For control subjects positive family history rates ranged from 3% (Bishop and Edmundson 1986) to 46% (Tallal et al. 1989a), with a mean incidence of 18% and a median incidence of 11%.

Percentage of Impaired Relatives. As shown in Table 2, 11 studies reported the percentage of probands' relatives who were impaired (Haynes and Naidoo 1991; Lahey and Edwards 1995; Lewis 1992; Lewis et al. 1989; Neils and Aram 1986; Rice et al. 1998; Tallal et al. 1989a; Tomblin 1989; Tomblin and Buckwalter 1994; van der Lely and Stollwerck 1996; Whitehurst et al. 1991). If impairment rates were given for both first-degree and extended relatives, rates for first-degree relatives were used. When rates were given for "any impairment" and for specific types of impairments (e.g., dyslexia, SLI), the rates for "any impairment" were used. If no overall rates were given, the rates for spoken language impairment were used. For Lewis (1992) I calculated the rate of spoken language impairment by combining the figures given for relatives who had only spoken language impairments and for relatives who had both written and spoken language impairments.

For probands the percentage of family members who were impaired ranged from 20% (Neils and Aram 1986) to 42% (Tallal et al. 1989a), with a mean impairment rate of 28% and median impairment rate of 26%. For control subjects the percentage of family members who were impaired ranged from 3% (Neils and Aram 1986) to 19% (Tallal et al. 1989a), with a mean impairment rate of 9% and a median impairment rate of 7%. The incidence of impairment was significantly higher among proband relatives than among

control relatives in seven of the eight studies that made such a comparison. Whitehurst et al. (1991) found that first-degree proband relatives were more likely to be impaired than first-degree control relatives on all measures, but none of the differences were significant. Whitehurst et al. (1991) also found that for extended families control relatives were significantly *more* likely than proband relatives to suffer from "any problem," school problems, or late talking but not speech problems. It is unclear what to make of the discrepancy between first-degree and extended family members, but it should be noted that this study was the only study to reveal such a finding.

Effect of Criteria on Rates of Impairment. Although all the studies revealed that probands were more likely to have a positive family history of impairments than control subjects and that probands' relatives were more likely to be impaired than control subjects' relatives, the reported rates varied considerably. Part of this variance is probably due to heterogeneity of the probands included in the studies. Although descriptions of the probands suggest that they met inclusionary and exclusionary criteria for specific developmental language impairments (Stark and Tallal 1981), as indicated in Table 1, these criteria are sufficiently broad that children with very different phenotypic profiles were included in the reviewed studies. Given the degree of phenotypic heterogeneity among the probands, even if all the probands suffered from genetic language disorders, the children were almost certainly genetically heterogeneous. Genetic heterogeneity is likely to affect rates of family affectedness because genetic factors may play a greater role in the expression of some language disorders than in others.

Variations among the studies in family member diagnoses had a profound impact on the rates of family affectedness. As expected, studies that were more restrictive about what counted as evidence of language impairment in family members found lower rates of positive family history of language impairment and lower percentages of language-impaired relatives for both probands and control subjects. For example, Tomblin (1989) found that 23% of language-impaired children's first-degree relatives had received speech therapy, whereas only 3% of first-degree relatives of nonimpaired control children had received speech therapy. Using much less stringent criteria for impairment, Tallal et al. (1989a) found that 42% of the first-degree relatives of children with SLI had a history of speech or language disorder (excluding isolated articulation disorders), dyslexia, learning disability, or poor school performance (having to repeat a grade or exhibiting below-average performance in reading, writing, or math), whereas 19% of first-degree relatives of well-matched control subjects had a similar history.

Unfortunately, Tallal et al. (1989a) did not provide the percentages of proband and control relatives who specifically had a history of speech or language problems. The extent to which Tallal et al.'s (1989a) figures are inflated by the inclusion of relatives with nonlanguage impairments ...

estimated by comparing the affectedness rates for parents when liberal and conservative criteria of affectedness are used. When liberal criteria of affectedness are used, 40% of proband parents and 20% of control parents were affected, whereas only 25% of proband parents and 10% of control parents had a history of language impairment. The percentage of proband parents with a history of language impairment in the Tallal et al. (1989a) study is similar to that found in other studies (Ingram 1959; Neils and Aram 1986; Tomblin 1989; Lewis 1992; Lewis et al. 1993; Tomblin and Buckwalter 1994; Haynes and Naido 1991; Beitchman et al. 1992), although the percentage of control parents with a history of language impairment is higher in the Tallal et al. (1989a) study than in other studies (Tomblin 1989; Lewis 1992).

The extent to which the rates of familial affectedness are increased when the criteria for affectedness are made less stringent can also be seen by comparing the familial affectedness rates within a study. For example, in their study of children with severe phonological disorders, Lewis et al. (1989) found that only 9% of all proband relatives (both nuclear and extended) and 1% of all control relatives had a history of developmental speech or language disorders, but when relatives with dyslexia, stuttering, or unspecified learning disabilities were also considered affected, 12% of all proband relatives and 2% of all control relatives were affected. In a study of a different group of children with phonological disorders Lewis (1992) found that 26% of probands' first-degree relatives and 4% of control subjects' first-degree relatives had a history of developmental speech or language disorders. When relatives with dyslexia or learning disabilities were also considered affected, the percentage of affected first-degree proband relatives rose to 33%. Rice et al. (1998) found that 22% of probands' first-degree relatives and 7% of control subjects' first-degree relatives had a history of speech or language disorders, but when relatives with reading, spelling, or learning problems were also considered affected, impairment rates rose to 26% and 13% for proband and control relatives, respectively.

Nuclear versus Extended Families and Rates of Impairment. As expected, the number of probands and control subjects with a positive family history was less when only first-degree relatives were considered than when all relatives were considered. With the exception of the control children in Whitehurst et al.'s (1991) study, in all studies the percentage of affected proband and control relatives was greater when only first-degree relatives were considered than when all relatives were considered. This probably reflects that people are more aware of developmental language disorders in members of their immediate family than in more distant relatives [see Lewis (1992)].

Effect of Method of Ascertainment and Rates of Impairment. Tomblin (1989) argued that using parental reports to determine whether family members are impaired may inflate the differences between probands and control subjects because parents of language-impaired children may be more aware of relatives' history of language impairment. Tomblin (1989) also argued that parental reports about subjective measures (e.g., were relatives difficult to understand as children?) are more likely to be biased than parental reports of more objective measures (e.g., did relatives require speech therapy?). Despite these concerns, in studies that counted the same conditions as evidence of a history of language impairment, the familial affectedness rates were similar regardless of whether the affectedness of relatives was determined by testing relatives (Lewis et al. 1989; Tomblin and Buckwalter 1994) or by asking objective or subjective questions about language status.

Discussion

Ascertainment Biases. Although familial aggregation data suggest that some language disorders have a genetic component, several alternatives must be considered. One possibility is that the higher rate of language impairment among proband relatives than among control relatives is the result of how probands were selected. For example, parents with language-impaired children who have a strong family history of language impairment may be more likely to volunteer for genetic studies of language impairment than parents of language-impaired children who do not have a family history of language impairment. Although this possibility cannot be eliminated, the fact that all the studies yielded similar results (even though the methods used to recruit subjects varied) suggests that selection bias is not solely responsible for the higher rate of impairment among proband relatives. A second possibility is that parents of language-impaired children may be more likely to know of and report language impairments in relatives than parents of children who do not have language impairments. Again, this is unlikely to be the explanation because rates of impairment reported by parents were similar to those found through direct testing.

Deviant Linguistic Environment Hypothesis. A third possibility is that children with language-impaired parents or siblings are more likely to be linguistically impaired because they are exposed to deviant language [the deviant linguistic environment hypothesis (DLEH)]. Some studies have reported that mothers are more likely to use directive speech and less likely to use responsive speech when talking to their language-impaired children than mothers speaking to normal children [e.g., Conti-Ramsden and Friel-Patti (1983)]. However, as numerous researchers have pointed out, children's language impairments may cause mothers to use simplified speech rather than vice versa; mothers of language-impaired children may use directive speech because they cannot understand their impaired children and their impaired children cannot understand them if they use more complicated language. Furthermore, within a fairly wide range linguistic environment appears to have

little or no effect on language acquisition by normal children [e.g., Heath (1983)].

The DLEH makes a number of specific predictions about the patterns of impairments that are likely and unlikely to occur in families. If impoverished linguistic environments cause language impairments to be concentrated in certain families, then the most severely linguistically impaired children should come from families with the highest incidence of language impairment. Contrary to this prediction, Byrne et al. (1974) found that children with profound language impairments were *less* likely to have positive family histories of language impairment than children with moderately language impairments. [However, Byrne et al. (1974) also found that compared with moderately impaired subjects, severely impaired subjects had more physical deformities, suffered more pre- and perinatal complications, and had more illnesses in early childhood. This is consistent with severe impairments more often being the result of pre- or postnatal events or congenital impairments and moderate language impairments more often being familial. In effect, these data suggest that there are (at least) two sources of impairment.] Also contrary to the DLEH, Lahey and Edwards (1995) found that impairment rates were greater among relatives of children who had isolated expressive language impairments than among relatives of children who had both receptive and expressive language impairments. Tallal et al. (1991) found no differences in the language abilities of children who did and did not have a positive family history of language disorders, although their study included only children who had both expressive and receptive delays.

If exposure to deviant language causes children to become language impaired, then language-impaired children should have the same type of impairment as their relatives. Contrary to this prediction, Neils and Aram (1986) found that 38% of parents who reported a history of a speech and language disorder said that the disorder they had was different from their child's disorder. In a longitudinal study of the speech and language status of children of 24 probands with speech or language disorders and 28 control subjects, Felsenfeld et al. (1995) found that children of probands performed significantly worse on all tests of articulation and expressive language and were significantly more likely to have received articulation treatment than children of control subjects. They found no evidence, however, that probands' children made the same types of phonological errors as their parents.

Sometimes children outgrow their spoken language impairments and exhibit no overt signs of spoken language impairment as adults. If the DLEH is correct, then parents with a history of spoken language impairment who are no longer impaired should be no more likely to have language-impaired children than parents with no such history. Contrary to this prediction, Neils and Aram (1986) found that a third of the probands' parents who had a history of a spoken language disorder no longer suffered from the disorder as an adult. In a prospective study of early spoken language deficits in children who were later diagnosed as dyslexic, Scarborough (1990) found that on tests of spoken language normal readers from dyslexic families did not perform like their dyslexic siblings but rather performed like normal readers from normal families. This suggests that the early language deficits of dyslexic children do not stem from being reared in dyslexic families.

Contrary to Scarborough's (1990) findings, Lewis et al. (1989) found that in their study of children with severe phonological disorders siblings of probands were more like their impaired siblings than they were like normal control subjects on tests of speech and reading (but not language). One possible explanation for the differing results is that they are due to differences in the proband populations (children with reading disorders versus children with phonological disorders). A more likely explanation is that, whereas Scarborough (1990) excluded any proband siblings who had written or spoken language disorders, Lewis et al. (1989) did not exclude siblings who were themselves impaired. Lewis et al. (1989) reported that 17% (5 of 30) of proband siblings were receiving speech therapy at the time of the study, and it is possible that the inclusion of 5 impaired siblings caused the performance of proband siblings to be more like that of probands than that of normal controls.

If the DLEH is correct, then one would expect that all children with SLI would have at least one close relative with a language impairment. Contrary to this prediction, in the reviewed studies an average of 54% of the language-impaired children had no impaired first-degree relatives. If the DLEH is correct, birth order might affect how likely a child is to exhibit a language disorder (e.g., because later born children get proportionally less linguistic input from parents). The birth order data are mixed. Tomblin et al. (1991) found that birth order did not have an effect on the severity or likelihood of developing language disorders, whereas Lahey and Edwards (1995) found that last-born children were more likely to be language impaired than first-born or middle-born children. In most societies mothers typically have the primary responsibility for child rearing. Thus the DLEH predicts that the correlation of language status should be greatest between mother and child. Contrary to this prediction, Tomblin (1989) found that among the family relations he studied (e.g., mother-child, father-child, male sibling–child, female sibling–child), the relationship was weakest between mother and child. In addition, Rice et al. (1998) found that the ratio of impaired fathers to impaired mothers was 4.5:1; Tomblin and Buckwalter (1994) found a ratio of 2.7:1; Neils and Aram (1986) found a ratio of 1.4:1, and Tallal et al. (1989a), Whitehurst et al. (1991), and Lewis (1992) all found that the ratio was approximately 1:1.

Modes of Transmission

The most definitive method for investigating the genetic basis of familial language disorders is to determine which genes are responsible for the

language disorders found in affected families [see Brzustowicz (1998) (this issue)]. To date, no linkage analyses have been published for spoken language disorders. Despite the lack of genetic studies, if behavioral studies reveal that the pattern of family members who are and are not impaired is consistent with a particular model of genetic transmission, this supports the notion that language disorders are genetically transmitted.

Single Major Locus Models. For genetic disorders with a single major locus the observed ratios of affected and unaffected relatives can be compared with those expected for a particular Mendelian transmission pattern (autosomal dominant, autosomal recessive, X-linked recessive). If we assume that expressivity and penetrance rates approach 100% and that the spontaneous mutation rate approaches 0%, simple Mendelian genetics dictates that, if a disorder is autosomal dominant and if an affected person mates with an unaffected person, on average half of the offspring will be affected and these affected offspring will in turn transmit the disorder to half of their offspring. Unlike autosomal dominant disorders, which should not skip generations, autosomal recessive disorders may (e.g., an impaired grandparent and grandchildren, but normal parents). If a disorder is autosomal recessive and two (unaffected) carriers mate, then a quarter of their offspring will be affected and three-quarters will be unaffected. Of the unaffected offspring, on average, two-thirds will be carriers. If these carriers mate with noncarriers, none of their offspring will be affected but half will be carriers. In the rare cases in which a carrier mates with an affected individual, half of the resulting offspring will be affected. Of those offspring that are not affected, half will be carriers.

With X-linked recessive traits, if a female who is unaffected but is a carrier for a condition mates with an unaffected male, half of the sons will have the condition and half of the daughters will be carriers for the condition. If an unaffected male mates with a female who is not a carrier (and does not have the trait), half of the daughters will be carriers and none of the sons will be affected. If a female who exhibits an X-linked recessive trait mates with an unaffected male, all of the sons will have the condition and all of the daughters will be carriers for the condition. The only way an affected male and an unaffected female can transmit an X-linked recessive disease is if the female is a carrier. In such rare cases half of the male and female offspring will be affected.

Multifactorial Polygenic Models. Complex behavioral traits such as language may result from several genes interacting in complex ways with each other and the environment (multifactorial polygenic). Genetic models may require additional parameters (e.g., rate of spontaneous mutation, probability of ascertainment, penetrance and expressivity of a trait). For example, epidemiological studies indicate that even within families language impairments

can be expressed in different ways, with some family members exhibiting severe global deficits in spoken and written language and others exhibiting only moderate deficits in written language. Incomplete penetrance is also possible, and some family members may have the genotype for speech or language disorders but exhibit no observable sign of a spoken or written language disorder.

The probable existence of genetic and behavioral heterogeneity for language disorders means at least three distinct relationships could exist between genotypes and behavioral phenotypes. It is possible (although unlikely) that there is a one-to-one relationship between genotypes and phenotypes, with each genotype causing a distinct type of language disorder. According to such an account, for each behaviorally distinct language disorder there is a distinct disorder at the genotype level. Alternatively, there might be a one-to-many mapping between genotypes and phenotypes, with a single genetic disorder resulting in many behaviorally distinct types of language disorders. For example, one child with a genetically encoded articulation disorder might respond by refusing to talk at all, whereas another child with the same genetic disorder might speak and make many articulation errors. Last, there may be a many-to-one mapping between genotypes and phenotypes with many distinctive genetic disorders resulting in the same type of linguistic disorder. For example, many researchers have noted that English-speaking children with developmental language disorders frequently omit functional or grammatical category morphemes, such as articles and determiners, auxiliary verbs, and verbal and nominal inflections (Leonard 1998; Stromswold 1997b). Some of these children may have a genetically encoded articulation disorder that causes them to omit functional morphemes because they are typically pronounced rapidly. Some may have a genetically encoded disorder affecting processing of rapid auditory input that causes them to have difficulty acquiring unstressed, short-duration functional morphemes. Others might omit functional morphemes because they have a syntactic deficit and functional morphemes convey grammatical information.

In addition, the genotype-phenotype relationship is not necessarily static. Longitudinal studies reveal that even within an individual a developmental language disorder can manifest itself in different ways at different ages (Aram et al. 1984; Silva et al. 1987).

Family Aggregation. Results of family aggregation studies may provide insight into the mode of transmission of language disorders. For example, if the incidence of a disorder is low (say, 5%) and the disorder is autosomal dominant, then most probands will have exactly one affected parent and, on average, half the probands' siblings will be impaired. [Assuming that impaired people are equally likely to mate with impaired people as unimpaired people are, if the incidence of a disorder is 5%, then the probability of having 2 affected parents is $0.05^2 = 0.0025$. But see Gilger (1991) for evidence of

assortative mating.] If the disorder is autosomal recessive, then most probands will have two (unaffected) carriers for parents, and one-quarter of the proband's siblings will be impaired.

The rate of affectedness among probands' parents in the family aggregation studies reviewed here ranged from 19% to 71% (mean rate = 31.6%, median rate = 25.5%), with 9 of the 12 studies having rates of 19% and 28%. Lewis et al. (1993) reported that 7% of the probands in their study had two affected parents, Lahey and Edwards (1995) reported that 10% of their probands had two affected parents, and Tallal et al. (1989a) reported that 19% of their probands had two affected parents. The fact that in most of the studies most of the probands did not have an affected parent suggests that there is considerable genetic heterogeneity (i.e., many cases of familial language impairments do not involve autosomal dominant transmission), the rate of spontaneous mutation is high, or there is incomplete penetrance of language disorders. For the reviewed studies the rate of affectedness among probands' siblings ranged from 15% to 49% (mean rate = 30.3%, median rate = 28.5%). Thus for most of the reviewed studies the sibling rates are more consistent with spoken language disorders being genetically heterogeneous or autosomal recessive than with autosomal dominant transmission (although the rates are compatible with autosomal dominant transmission with low penetrance or expressivity).

Pedigree Analyses. Pedigree analyses involve rejecting possible modes of transmission based on which family members in a pedigree are affected, and the greater the number and types of genetic relationships that exist in a pedigree, the more likely it is that particular models can confidently be rejected [see Pauls (1983)]. One problem with pedigree studies is that the techniques used to test different modes of transmission are extremely sensitive to errors in phenotypic characterization. Misclassifying even a small number of unaffected family members as affected or vice versa can dramatically affect how likely various modes of transmission are deemed to be. This is particularly problematic for pedigree studies of developmental language disorders because of the complex relationship between genotype and phenotype.

Several studies have reported a number of kindreds with extremely large numbers of severely affected family members (Arnold 1961; Gopnik 1990; Hurst et al. 1990; Lewis 1990) in which transmission seems to be autosomal dominant with variable rates of expressivity and penetrance. Hurst et al. (1990) performed a pedigree analysis on the same family studied by Gopnik (1990) and concluded that the mode of transmission in this family was a single autosomal dominant gene with near 100% penetrance. Samples and Lane (1985) performed a similar analysis on a family in which the parents were first cousins. Neither parent reported any written or spoken language difficulties, yet all six of their offspring had a severe developmental language disorder. Based on these findings, Samples and Lane concluded that the most likely mode of transmission in this family was a single autosomal recessive gene.

Pedigree studies can be used to evaluate predictions made by the hypothesis that the tendency for language disorders to run in families is the result of the deviant linguistic environments in such families (i.e., the DLEH) rather than the result of a genetically transmitted disorder. According to the DLEH, within a family with a high incidence of language impairment, one would not expect to find any family members with completely normal language. However, even in families with a high incidence of language impairment, there are often family members whose language develops normally despite extensive exposure to deviant language. For example, in the 3 families studied by Arnold (1961), 9 of 24 members of family 1, 20 of 32 members of family 2, and 17 of 29 members of family 3 were not language impaired (see Figures 1a–1c). In the family studied by Hurst et al. (1990) and Gopnik (1990) (Figure 1d) 14 of the 30 family members had normal language. Lenneberg (1967) reported that Luchsinger studied a family in which 6 of 14 family members were not language impaired (Figure 1e). In a family studied by Brewer [also reported by Lenneberg (1967)] 14 of 30 family members were unaffected. If nonbiological relatives (i.e., relatives by marriage) are included, 15 of 28 members of Arnold's (1961) family 1, 20 of 32 members of Arnold's (1961) family 2, 27 of 39 members of Arnold's (1961) family 3, 13 of 21 members of Luchsinger's family, 30 of 47 members of Brewer's family, and 20 of 36 members of the family studied by Hurst et al. (1990) and Gopnik (1990) were not language impaired.

Even for an autosomal dominant disorder the only way all offspring could regularly be affected is in the extremely rare instance in which at least one of the affected parents happened to have two defective genes. The DLEH, on the other hand, predicts that within a nuclear family all siblings should either be language impaired or normal. Contrary to this prediction, familial aggregation studies and most pedigree studies of language disorders reveal that language-impaired children usually have some siblings that are language impaired and some siblings that are not language impaired. For example, this is true for all three of the families studied by Arnold (1961), three of the four families studied by Lewis (1990), and the families studied by Luchsinger (Figure 1e), Brewer (Lenneberg 1967), and Stromswold (1994) (Figure 1f).

Consider Arnold's (1961) family 1 in which both grandparents had language impairments. Despite the fact that both parents were language impaired, only two of their five offspring were language impaired. Both of the language-impaired offspring were female and married nonimpaired husbands. One of the impaired daughters had three male offspring, none of whom were affected. The other daughter had six language-impaired offspring (three males, three females) and one normal (female) offspring. If the DLEH is correct, it is unclear how three of the five offspring of two language-impaired individuals could develop normal language. It is also unclear how the unaffected granddaughter

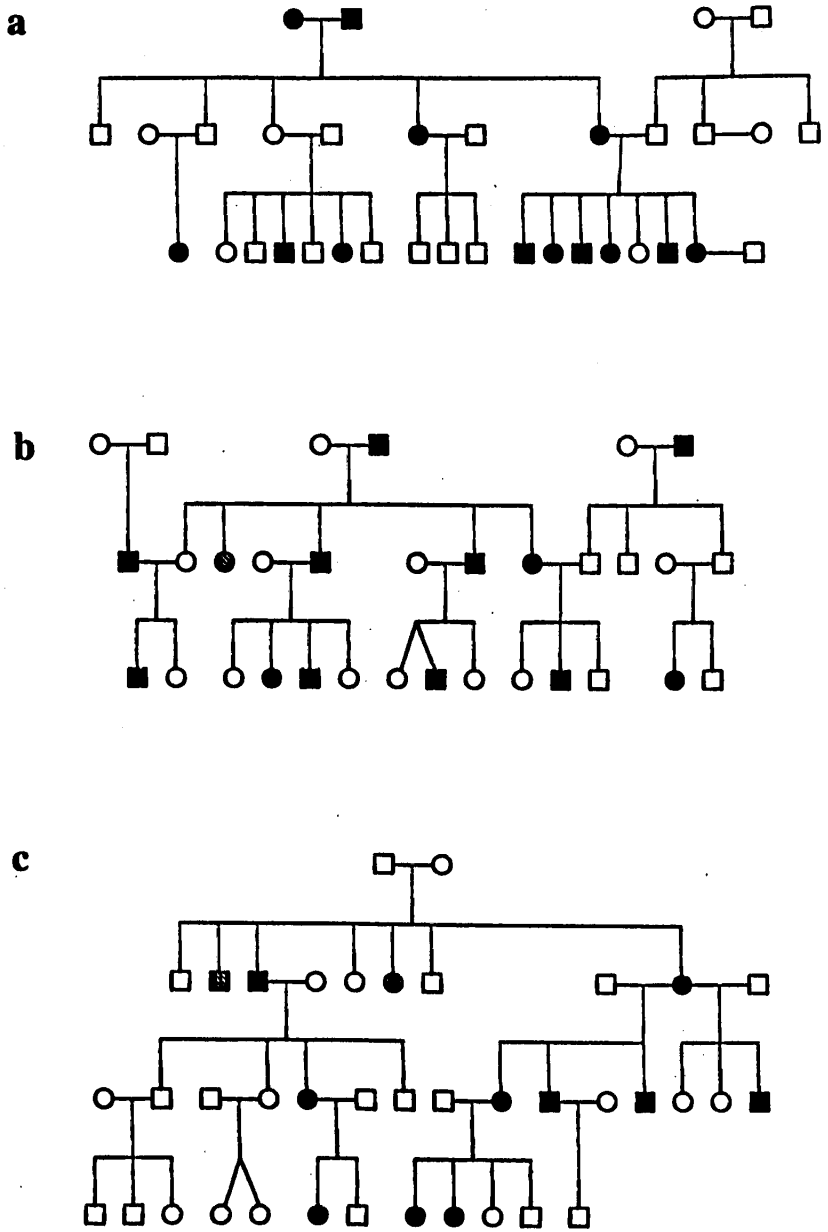

Figure 1. Pedigrees of families with high incidences of language impairment. (a) Arnold (1961) family 1; (b) Arnold (1961) family 2; (c) Arnold (1961) family 3; (d) Gopnik (1990) and Hurst et al. (1990) family; (e) Luchsinger family (Lenneberg 1967); (f) Stromswold (1994) family. Circles, females; squares, males; diamonds, sex unknown. Solid, affected; open, unaffected; hatched, possibly affected.

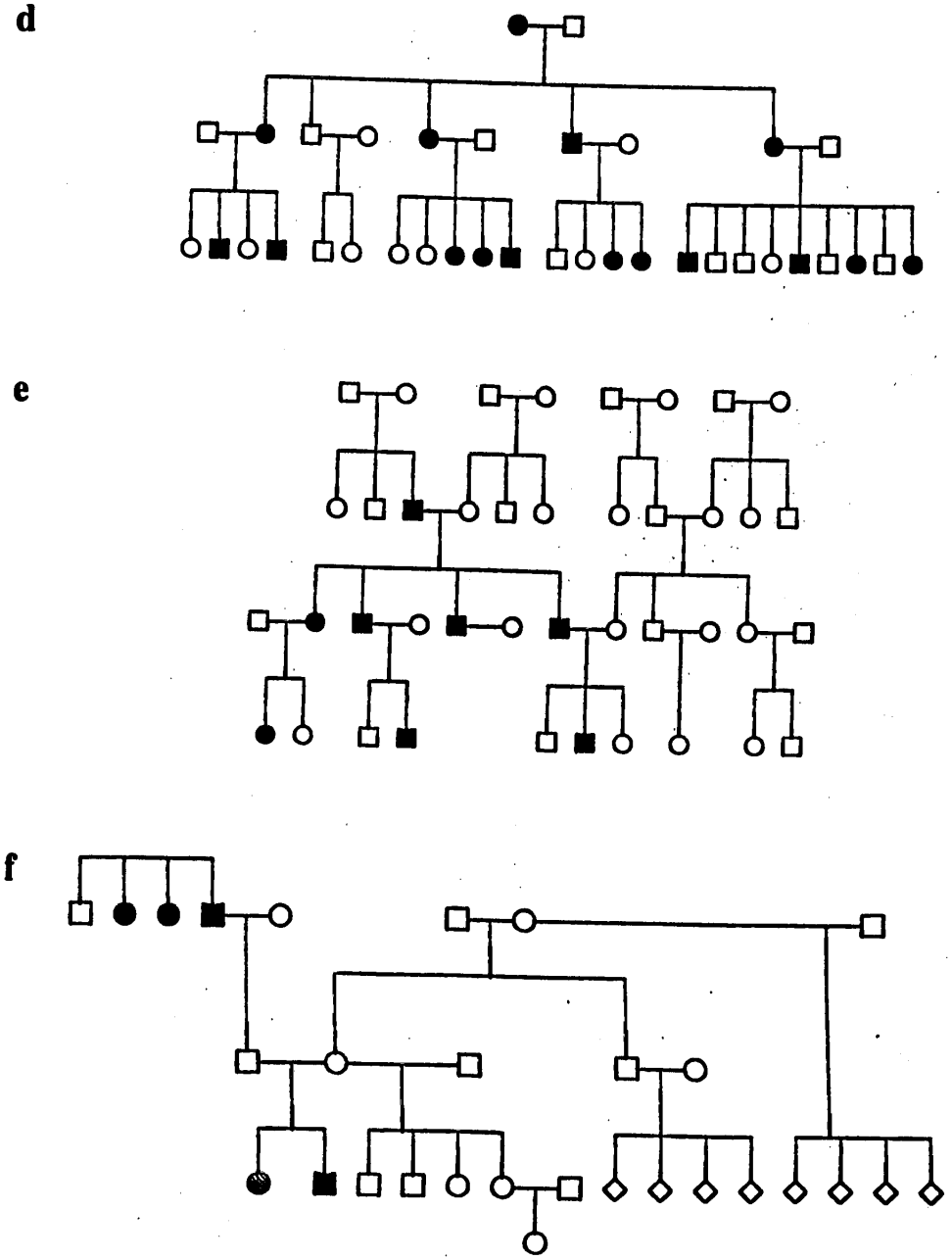

Figure 1. Continued.

managed to develop normal language despite exposure to deviant language from six language-impaired siblings, a language-impaired mother, two language-impaired grandparents, a language-impaired aunt, and three language-impaired cousins.

The DLEH predicts that one will not find families in which the language impairment skips a generation. In Arnold's (1961) family 1 and family 2 there are instances of language impairment skipping a generation. Particularly revealing is Arnold's family 1, in which both grandparents are affected. Of the three offspring that were not language impaired, one did not have any children. The other two unimpaired offspring both had at least one child with a language impairment (Arnold 1961). Another example of a language impairment skipping a generation can be seen in a male child with severe developmental dyspraxia (see Figure 1f) (Stromswold 1994). Although both of his parents have normal language, his paternal grandfather is severely dysfluent and his two paternal great aunts are mildly dysfluent.

Sex Ratio Studies. The incidence of language disorders is often reported to be greater for males than for females. However, a cursory review of the family pedigrees shown in Figure 1 indicates that language disorders may be transmitted from father to son and that unaffected fathers may have affected daughters, suggesting that spoken language disorders are *not* the result of an X-linked recessive trait. Sex ratios can also be used to investigate whether language disorders are X-linked recessive. For an X-linked recessive trait to be expressed in a woman, the recessive allele must be present on both of a woman's X chromosomes, whereas for a man to express the trait, the recessive allele needs to be present only on the man's one X chromosome. Thus, if language disorders are X-linked recessive, the male:female ratio should be $n{:}n^2$, where n is the frequency of the disordered allele. For example, if the X-linked recessive allele is found on 10% of X chromosomes, the male:female sex ratio will be 10:1. If the X-linked recessive allele is found on 5% of X chromosomes, the ratio will be 20:1.

As shown in Table 3, of the 18 family aggregation studies of spoken language disorders reviewed here, all but 2 (Bishop and Edmundson 1987; Byrne et al., 1974) included information about the sex of probands or probands' affected and unaffected relatives. The male:female sex ratio for probands in these studies ranged from a low of 1.3:1 (Tomblin 1996a) to a high of 5.9:1 (Whitehurst et al. 1991), with most studies reporting sex ratios between 2:1 to 3:1. In the eight studies that reported the sex ratios of affected siblings the ratios ranged from 1:1 (Rice et al. 1998; van der Lely and Stollwerck 1996; Whitehurst et al. 1991) to 4:1 (Tomblin and Buckwalter 1994), with the rest reporting sex ratios between 1:1 and 2.5:1. In the nine studies that reported affected parental ratios impaired parent sex ratios ranged from 1:1 (Tomblin 1989; van der Lely and Stollwerck 1996; Whitehurst et al. 1991) to 4.5:1 (Rice et al. 1998), with seven studies reporting ratios between 1:1

Table 3. Sex Ratios

Study	*Sex Ratios for Probands and Controls (M:F)*	*Impaired Siblings Sex Ratio (M:F)*	*Impaired Parent Sex Ratio (M:F)*
Ingram (1959)	Probands, 2.6:1	NA	NA
Luchsinger (1970)	Probands, 2.2:1	NA	NA
Neils and Aram (1986)	NA	Probands, 1.3:1; controls, NA	Probands, 1.4:1; controls, NA
Lewis et al. (1989)	Probands, 1.9:1; controls, 1.9:1	Probands' affected relatives, 1.5:1; controls' affected relatives, 1.9:1	
Tallal et al. (1989a)	Probands, 2.5:1; controls, 1.0:1	Probands, 1.4:1; controls, 0.6:1	Probands, 1.2:1; controls, 1.1:1
Tallal et al. (1989b)	Probands, 2.4:1	65% of sibs were male (71% for affected sibs, 61% for unaffected sibs)	
Tomblin (1989)	Probands, 2.9:1; controls, N/A	Probands, 2.4:1; controls, NA	Probands, 1.0:1; controls, NA
Haynes and Naidoo (1991)	Probands, 4.8:1	Probands, 3.6:1	NA; 47% of sibs male
Tomblin et al. (1991)	Probands, 2.0:1	NA	NA
Whitehurst et al. (1991)	Probands, 5.9:1; controls, 3.2:1	Probands, 1:1; controls, 0.8:1	Probands, 1.0:1; controls, 2.7:1
Beitchman et al. (1992)	Probands, 1.9:1; controls, 1.8:1	NA	NA
Lewis (1992)	Probands, 1.6:1; controls, 1.6:1	LI probands, 1.8:1	LI probands, 1:1
Tomblin and Buckwalter (1994)	NA	Probands, 4:1	Probands, 2.7:1
Lahey and Edwards (1995)	Probands, 1.8:1	NA	Probands, 1.18:1
Rice et al. (1998)	Probands, 1.8:1	Probands, 0.9:1	Probands, 4.5:1
Tomblin (1996a)	Probands, 1.3:1	NA	NA
van der Lely and Stollwerck (1996)	Probands, 4:1; controls, NA	Probands, 1.1:1; controls, 2.53:1	Probands, 1.1:1; controls, 4.2:1

LI, Speech or language impairment.

and 1.5:1. Even the most extreme sex ratio for probands (6:1) (Whitehurst et al. 1991) or affected siblings (4:1) (Tomblin and Buckwalter 1994) are less than would be expected if language disorders were X-linked recessive.

If the higher incidence of language disorders in males than females is not the result of X-linked recessive transmission, why are males more likely to be language impaired than females? One possibility is that the greater ratio of impaired males is the result of an ascertainment bias. A number of studies of dyslexia that used systematic sampling rather than sampling from clinic populations have found no significant difference in the incidence of dyslexia among boys and girls (DeFries and Gillis 1993; Shaywitz et al. 1990; Wadsworth et al. 1992). Thus for dyslexia the elevated male:female ratio may reflect that boys are more likely to be referred to therapy than are girls.

The data for spoken language impairments are mixed. Through systematic screening of 849 kindergarten children, Tomblin (1996b) identified 16 boys and 9 girls as being language impaired based on performance on a battery of tests, whereas in a later study a systematic screening of 7218 kindergarten children revealed that 8% of boys and 6% of girls had spoken language disorders (Tomblin 1996a). Furthermore, once probands were removed from the analyses (thereby removing any ascertainment biases), the sex ratios for spoken and written language disorders typically ranged between 1:1 and 1.5:1. In their meta-analysis of studies that examine sex differences in verbal ability, Hyde and Linn (1988, p. 64) concluded that "the gender difference in verbal ability is currently so small that it can effectively be considered zero."

A second possibility for the sex difference in language impairment is that males and females might be equally likely to have language-disordered genotypes but that the expression of these genotypes is sometimes fatal in females. Tallal et al.'s (1989b) finding that 65% of the siblings of probands were male (71% of affected siblings, 61% of unaffected siblings) is consistent with such an account.

A third possibility for the greater incidence of impaired males is that males and females may be equally likely to have the genotypes that correspond to language disorders but that males may be at greater risk of expressing the disorder [e.g., Tallal et al. (1989a,b)]. In order for a female to express the language disorder phenotype, she may require a greater genetic load for language disorder than that required for a male to express the language disorder phenotype. A prediction entailed by this account is that, if a female is phenotypically language impaired, then she should be more likely to transmit the impairment to her offspring. A second prediction entailed by this theory is that, if female probands must receive a higher genetic load to express language impairments, female probands should have a greater number and density of impaired relatives.

The data are mixed with regards to these predictions. Consistent with the first prediction, Whitehurst et al. (1991) found that probands' mothers were significantly more likely than control subjects' mothers to be affected or to have been late talkers, and Tallal et al. (1989) found that probands' mothers were more likely than control subjects' mothers to have a language, reading, or writing disorder. Consistent with the second prediction, Lewis (1992) found that female probands had significantly more relatives with learning disabilities and global language impairments than male probands. Lewis et al. (1993) found that the percentage of affected family members was higher in families ascertained through female probands (15% of all family members, 39% of first-degree relatives) than in families ascertained through male probands (8% of all family members and 24% of first-degree relatives). Tallal et al. (1989b) reported that female probands had 1.6 times as many affected siblings as male probands. Beitchman et al. (1992) found no significant difference in the rate of positive family history for male and female probands, but they did find that significantly more female probands than male probands had more than one impaired relative. Lahey and Edwards (1995) found that a higher percentage of female probands' siblings were impaired than male probands' siblings (42% vs. 22%, respectively, $p < 0.1$).

On the other hand, contrary to the genetic load theory, Luchsinger (1970) found 20 cases of paternal line inheritance and only 12 cases of maternal line inheritance. Tomblin (1989) found no significant difference in the frequency of language impairment among relatives of female and male probands. Haynes and Naidoo (1991) found no significant difference in the rate of positive family history for male and female probands, and Rice et al. (1998) found that the sex of the proband did not significantly affect the percentage of nuclear or extended family members that were impaired (for nuclear families overall affectedness rates were 29% and 21% for male and female probands, respectively).

In summary, although Tallal et al. (1989b) has argued that the sex ratio data for SLI are most consistent with an autosomal dominant transmission with greater penetrance through mothers than fathers, at this point the sex ratio data from many studies conflict and this hypothesis cannot be said to be supported by the preponderance of the data. Rather, the higher incidence of language disorders among males than among females is probably due in large part to ascertainment biases.

Commingling Analyses. Multifactorial polygenic transmission results in a quantitative trait with a normal distribution, whereas single major locus transmission is more likely to generate a multimodal distribution, with each mean representing a distinct subpopulation. Maximum-likelihood estimates can be used to test the fit of different models with additional parameters (i.e., means) added only if they yield a better fit of the data. To date, only one commingling analysis of SLI data has been performed. Tomblin and Buckwalter (1994) analyzed 647 children's performance on TOLD-2 grammatical completion and picture identification (vocabulary) subtests. For vocabulary

the best model had a single mean. For grammatical completion the best model had two populations. One subpopulation included 80% of the children, with the other 20% of the children making up a second subpopulation with a higher mean. Tomblin and Buckwalter (1994) failed to find evidence of a low-performing group, indicating either that too few children with SLI were in the sample or that SLI represents the low end of normal ability.

Segregation Analyses. Once additional parameters are added to models of transmission, simple estimates of affectedness ratios are not sufficient to determine the mode of transmission. Rather, data from nuclear families must be used to estimate the values of parameters of a specific model. Segregation analyses can be used to give maximum-likelihood estimates of the parameters, and statistical tests can be used to test specific modes of transmission. One serious limitation of segregation analysis is that the technique assumes that all the nuclear families under investigation suffer from the same genetic disorder and that they represent a random sample of the population under investigation. Lewis et al. (1993) analyzed the pedigrees of 45 probands with a history of moderate to severe speech or language disorder. Eight hundred eighty-eight first-, second-, and third-degree family members were included in the pedigrees. To perform the segregation analysis, Lewis et al. split the 45 kindreds into 276 nuclear families. The hypothesis that there was neither multifactorial polygenic nor single major locus transmission was rejected at the 0.05 level. However, based on the analyses, Lewis et al. were unable to distinguish between single major locus and multifactorial polygenic transmission, but they concluded that in most cases multifactorial polygenic transmission is the most likely mode of inheritance of developmental language disorders.

Tomblin and Buckwalter (1994) analyzed the extended families (1004 family members) of 26 probands with SLI. Although the sample size was too small to permit a full segregation analysis. Tomblin and Buckwalter reported that a comparison of the expected and observed number of affected siblings (Penrose 1953) indicated that the most likely mode of transmission is multifactorial polygenic.

Summary and Future Directions

A comprehensive review of family aggregation studies indicates that spoken language disorders run in certain families and that this may be due, at least in part, to heritable factors. Based on the reviewed results, it is fairly safe to say that in most cases familial language disorders are the product of both genes and the environment. However, given the types of studies reviewed, it is difficult to quantify the extent to which genetic versus environmental factors play a role in the development of language disorders (among behavioral studies twin and adoption studies are better suited for quantifying

the relative importance of nature and nurture). The results of segregation, pedigree, commingling, and sex ratio studies suggest that spoken language disorders are etiologically heterogeneous and may be autosomal recessive or dominant or may involve complex interactions between genes and the environment. In addition to genetic heterogeneity, other factors make investigations of the genetics of language disorders difficult. The expression and penetrance of spoken language disorders are rarely complete. Furthermore, even within a family (whose impaired members presumably share the same language disorder genotype), there is often behavioral heterogeneity, with the type of language disorder varying from one impaired member to another and from one age to another.

Even though most children can develop language normally in a wide range of linguistic environments, it is possible that there is a synergistic effect between genetic and environment factors, with children who are genetically at risk for developing language disorders being particularly sensitive to subtly impoverished linguistic environments. If this is the case, family aggregation, twin, and pedigree studies (but not adoption studies) may overestimate the relative importance of genetic factors because children who are genetically at risk are also more likely to be exposed to deviant linguistic environments since language disorders run in families. Because most of the reviewed studies did not rigorously assess or control for the cognitive abilities of proband and control subjects, it is possible that language disorders are due (at least in part) to genetic factors but that these genetic factors are not specific to language.

One final caveat is that, even if there are language disorders that appear to be (partially) due to genetic factors specific to language, it is possible that such genetic factors play no appreciable role in the variation in linguistic abilities among unimpaired people. Even if we discover genotypes associated with language disorders (or genotypes associated with being a linguistic savant), the observed variation in language abilities for normal children may be due exclusively to environmental factors (such as education) or genetic factors (such as IQ) that are not specific to language. [Consider the following analogy: Although deviant alleles exist that cause people to have too few or too many red blood cells (anemia and polycythemia, respectively), the variation observed in hematocrit among "normal" people may be due to factors that are not genetic, such as diet, elevation, and presence of other diseases.]

Although it is possible for a single genotype to result in different linguistic profiles and, conversely, for different genotypes to result in similar profiles, researchers should attempt to limit behavior heterogeneity among their subjects. Doing so should increase the likelihood of identifying the genotypes associated with language disorders and hence lead to advances in early diagnosis. Success at identifying the genotypes for language disorders (e.g., through linkage studies) should also lead to progress in identifying the environmental factors that lead to the expression of language disorders. This in turn might lead to advances in the remediation of language disorders.

Acknowledgments This research was supported by grants from the John Merck Foundation on the Biology of Developmental Disabilities and the Johnson & Johnson Foundation. I am grateful to members of the audience at the 20th Boston University Conference on Language Development (Stromswold 1996) for their helpful comments. I also thank Dorothy Bishop, Jenny Ganger, Jeffrey Gilger, Bruce Tomblin, Heather van der Lely, and two anonymous reviewers for helpful discussion and/or for providing copies of unpublished manuscripts.

Received 11 December 1996; revision received 3 October 1997.

Literature Cited

Ambrose, N.G., E. Yairi, and N. Cox. 1993. Genetic aspects of early childhood stuttering. *J. Speech Hear. Res.* 36:701–706.

American Psychiatric Association. 1994. *Diagnostic and Statistical Manual of Mental Disorders,* 4th ed. Washington, DC: American Psychiatric Association.

Aram, D.M., B.L. Ekelman, and J.E. Nation. 1984. Preschoolers with language disorders: 10 years later. *J. Speech Hear. Res.* 27(2):232–244.

Arnold, G.E. 1961. The genetic background of developmental language disorders. *Folia Phoniatr.* 13:246–254.

Beitchman, J.H., J. Hood, and A. Inglis. 1992. Familial transmission of speech and language impairment: A preliminary investigation. *Can. J. Psychiatr.* 37(3):151–156.

Bishop, D.V., and A. Edmundson. 1986. Is otitis media a major cause of specific developmental language disorders? *Br. J. Disorders Commun.* 21:321–338.

Brzustowicz, L.M. 1998. Molecular genetic approaches to the study of language. *Hum. Biol.* 70(2):325–345 (this issue).

Byrne, B.M., L. Willerman, and L.L. Ashmore. 1974. Severe and moderate language impairment: Evidence for distinctive etiologies. *Behav. Genet.* 4(4):331–345.

Conti-Ramsden, G., and S. Friel-Patti. 1983. Mothers' discourse adjustments to language-impaired and non-language-impaired children. *J. Speech Hear. Disorders* 48:360–367.

DeFries, J.C., and J.J. Gillis. 1993. Genetics of reading disability. In *Nature, Nurture, and Psychology,* R. Plomin and G.E. McClearn, eds. Washington, DC: American Psychological Association, 121–145.

Felsenfeld, S., M. McGue, and P.A. Broen. 1995. Familial aggregation of phonological disorders: Results from a 28-year follow-up *J. Speech Hear. Res.* 38:1091–1107.

Gilger, J.W. 1991. Differential assortative mating found for academic and demographic variables as a function of time of assessment. *Behav. Genet.* 21:131–150.

Gopnik, M. 1990. Feature-blind grammar and dysphasia. *Nature* 344:715.

Haynes, C., and S. Naidoo. 1991. *Children with Specific Speech and Language Impairment.* London, England: MacKeith Press.

Heath, S.B. 1983. *Ways with Words: Language, Life, and Work in Communities and Classrooms.* New York: Cambridge University Press.

Hurst, J.A., M. Baraitser, E. Auger et al. 1990. An extended family with a dominantly inherited speech disorder. *Dev. Med. Child Neurol.* 32:347–355.

Hyde, J.S., and M. Linn. 1988. Gender differences in verbal ability: A meta-analysis. *Psychol. Bull.* 104:53–69.

Ingram, T.T.S. 1959. Specific developmental disorders of speech in childhood. *Brain* 82:450–467.

Lahey, M., and J. Edwards. 1995. Specific language impairment: Preliminary investigation of factors associated with family history and with patterns of language performance. *J. Speech Hear. Res.* 38:643–657.

Lenneberg, E.H. 1967. *Biological Foundations of Language.* New York: Wiley.

Leonard, L. 1998. *Children with Specific Language Impairment.* Cambridge, MA: MIT Press (in press).

Lewis, B.A. 1990. Familial phonological disorders: Four pedigrees. *J. Speech Hear. Disorders* 55:160–170.

Lewis, B.A. 1992. Pedigree analysis of children with phonology disorders. *J. Learn. Disabil.* 25(9):586–597.

Lewis, B.A., N.J. Cox, and P.J. Byard. 1993. Segregation analysis of speech and language disorders. *Behav. Genet.* 23(3):291–297.

Lewis, B.A., B.L. Ekelman, and D.M. Aram. 1989. A familial study of severe phonological disorders. *J. Speech Hear. Res.* 32:713–724.

Luchsinger, R. 1970. Inheritance of speech deficits. *Folia Phoniatr.* 22:216–230.

Neils, J., and D.M. Aram. 1986. Family history of children with developmental language disorders. *Percept. Mot. Skills* 63:655–658.

Pauls, D.L. 1983. Genetic analysis of family pedigree data: A review of methodology. In *Genetic Aspects of Speech and Language Disorders,* C.L. Ludlow and J.A. Cooper, eds. New York: Academic Press, 139–147.

Rice, M.L., K.J. Haney, and K. Wexler. 1998. Family histories of children with SLI who show extended optional infinitives. *J. Speech Lang. Hear. Res.* (in press).

Samples, J., and V. Lane. 1985. Genetic possibilities in six siblings with specific language learning disorders. *Am. Speech Lang. Hear. Assoc.* 27:27–32.

Scarborough, H.S. 1990. Very early language deficits in dyslexic children. *Child Dev.* 61(6):1728–1743.

Shaywitz, S.E., B.A. Shaywitz, J.M. Fletcher et al. 1990. Prevalence of reading disability in boys and girls: Results of the Connecticut Longitudinal Study. *J. Am. Med. Assoc.* 26:998–1002.

Silva, P.A., S. Williams, and R. McGee. 1987. A longitudinal study of children with developmental language delay at age three: Later intelligence, reading, and behavior problems. *Dev. Med. Child Neurol.* 29:630–640.

Stark, R.E., and P. Tallal. 1981. Selection of children with specific language deficits. *J. Speech Hear. Disorders* 46:114–122.

Stromswold, K. 1994. Language comprehension without language production: Implications for theories of language acquisition. Paper presented at the Boston University Conference on Language Development, Boston, Massachusetts.

Stromswold, K. 1996. The genetic basis of language acquisition. In *Proceedings of the 20th Annual Boston University Conference on Language Development,* A. Stringfellow, D. Cahana-Amitay, E. Hughes et al., eds. Somerville, MA: Cascadilla Press, v. 2, 736–747.

Stromswold, K. 1997a. The heritability of language: A review of twin and adoption studies. Unpublished.

Stromswold, K. 1997b. Specific language impairments. In *Behavioral Neurology and Neuropsychology,* T.E. Feinberg and M.J. Farah, eds. New York: McGraw Hill, 755–772.

Tallal, P., R. Ross, and S. Curtiss. 1989a. Familial aggregation in specific language impairment. *J. Speech Hear. Disorders* 54:167–173.

Tallal, P., R. Ross, and S. Curtiss. 1989b. Unexpected sex ratios in families of language/learning impaired children. *Neuropsychologia* 27(7):987–998.

Tallal, P., J. Townsend, S. Curtiss et al. 1991. Phenotypic profiles of language impaired children based on genetic/family history. *Brain Lang.* 41:81–95.

Tomblin, J.B. 1989. Familial concentrations of developmental language impairment. *J. Speech Hear. Disorders* 54:287–295.

Tomblin, J.B. 1996a. The big picture of SLI: Results of an epidemiologic study of SLI among kindergarten children. Paper presented at the Symposium on Research in Child Language Disorders, Madison, Wisconsin.

Tomblin, J.B. 1996b. Genetic and environmental contributions to the risk for specific language impairment. In *Towards a Genetics of Language*, M. Rice, ed. Mahwah, NJ: Eribaum Associates, 191–210.

Tomblin, J.B., and P.R. Buckwalter. 1994. Studies of genetics of specific language impairment. In *Specific Language Impairments in Children*, R.V. Watkins and M.L. Rice, eds. Baltimore, MD: Paul H. Brookes Publishing, 17–34.

Tomblin, J.B., J.C. Hardy, and H.A. Hein. 1991. Predicting poor-communication status in preschool children using risk factors present at birth. *J. Speech Hear. Res.* 34:1096–1105.

van der Lely, H., and K. Stollwerck. 1996. A grammatical specific language impairment in children: An autosomal dominant inheritance? *Brain Lang.* 52:484–504.

Wadsworth, S.J., J.C. DeFries, J. Stevenson et al. 1992. Gender ratios among reading-disabled children and their siblings as a function of parental impairment. *J. Child Psychiatr. Psychol.* 33(7):1229–1239.

Whitehurst, G.J., D.S. Arnold, M. Smith et al. 1991. Family history in developmental expressive language delay. *J. Speech Hear. Res.* 34:1150–1157.