LETTERS TO THE EDITOR

Airway Resistance Characteristics of Voice Button Tracheoesophageal Prostheses

Panje (1981) has described a surgical-prosthetic method of speech restoration for laryngectomized patients. In this method, a tracheoesophageal fistula is created on a surgical basis and a one-way valve prosthesis (Voice Button) is inserted into this opening. From a design perspective, tracheoesophageal puncture prostheses should be minimally resistive to airflow through them from the trachea to the esophagus. Decreased resistance of these devices would be expected to (a) enhance the efficiency with which esophageal voice is produced and (b) facilitate the use of a tracheal breathing valve.

In an earlier investigation (Moon, Sullivan, & Weinberg, 1983) involving Blom-Singer prostheses we have shown that two major factors (air entrance effect; air exit effect) contribute to the development of airway resistance in tracheoesophageal puncture prostheses. To date, there has been no systematic evaluation of the airway resistance offered by both components of Voice Button prostheses. The purpose of this study was to address that specific need.

PROCEDURE

The opposition Voice Button prostheses offer to the flow of air through them was calculated from the ratio between transdevice pressure and transdevice flow. Pressure measurements were made and resistance values calculated for 10 prostheses obtained directly from the manufacturer (Zomed Inc., Jacksonville, Florida).

Air Exit Effect: Four-Flapper Valve Resistance

The instrumentation used to obtain estimates of the airway resistance characteristics of the (four-flapper) valve portion of Voice Button prostheses is shown in Figure 1. Known rates of airflow were delivered to each prosthesis and the magnitudes of pressure developed at these rates were measured. Initially, the pressure buildup within the measurement system itself was measured at each flow rate (see A). During device testing, prostheses were individually coupled to the measurement system using a catheter assembly (see B). The pressure buildup at each flow rate was recorded and the system pressure buildup value for the appropriate flow rate was subtracted from this value to provide a measure of transdevice pressure.

Air Entrance Effect: Air Port Resistance

To calculate the airway resistance characteristics of this effect, some important conditions under which the prostheses are typically used were simulated. For example, a glass replica of the postlaryngectomy trachea was constructed to simulate cross-sectional area and airflow direction characteristics (Figure 2). Each device tested was inserted into a "puncture site" (P) and a retention block (R) was used to simulate tracheal occlusion. Known flow rates were introduced into this physical model and the pressure buildup was measured using a catheter-differential pressure transducer assembly (D). To measure the resistance to airflow offered solely by the air entrance port, a thin walled tube with an inner cross-sectional area equal to that of the prosthesis

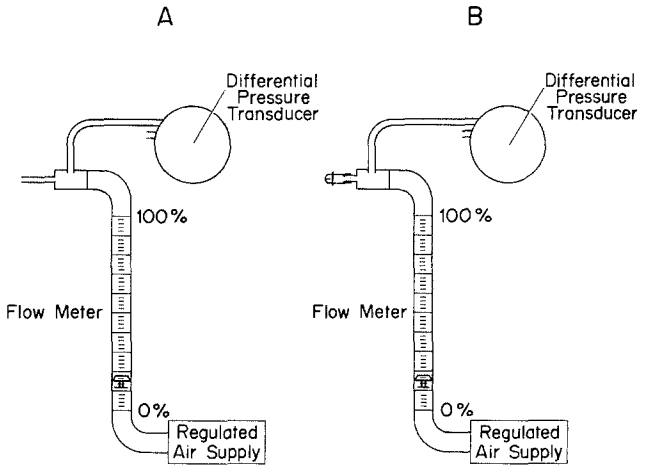

FIGURE 1. Instrumental array for measurement of airway resistance characteristics of the valve portion of the prostheses (A = measurement system alone; B = measurement system with prosthesis attached).

was inserted through the flapper valve portion of each prosthesis (see B). This procedure served to remove the influence of the flapper valve on pressure measurement and subsequent resistance calculations.

Total Device Resistance

The instrumentation shown in Figure 2 was also used to estimate the total airway resistance of the Voice Button. One procedural variation was introduced. Namely, the valve portion of the prosthesis was not opened with a catheter, but was left untouched. In this way, the influence of both the four-flapper valve and air entrance port were measured (see C).

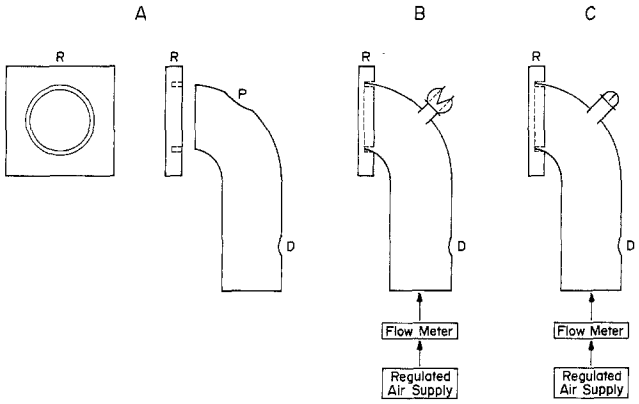

FIGURE 2. Instrumental array for measurement of airway resistance characteristics of air entrance port and overall prosthesis resistance (A = simulated trachea and retention block; B = air entrance port resistance measurement system; C = overall prosthesis resistance measurement system).

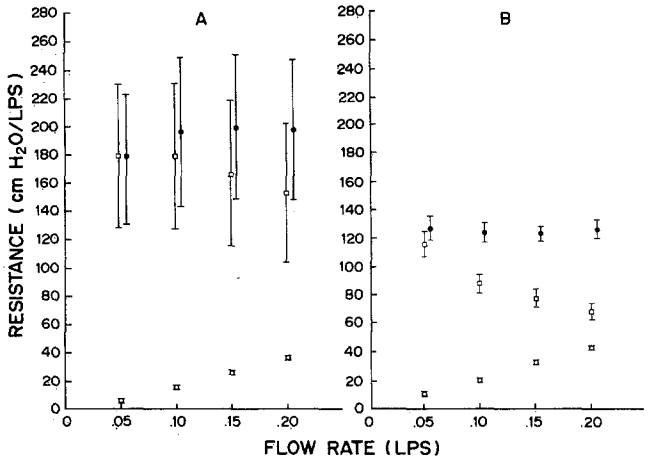

FIGURE 3. Resistance to airflow of Voice Button prostheses (A) and Blom-Singer prostheses (B).

RESULTS

Airway resistance properties of Voice Button prostheses are summarized in Figure 3A. These data illustrate mean resistance and standard deviation values for 10 devices. Each mean data point represents the average of three resistance calculations for 10 devices. A nonsignificant ($p > .01$) repeated measures effect was obtained for this body of data, indicating that the airway resistance for individual devices calculated on the basis of repeated, independent trials did not differ significantly from trial to trial.

Air Exit Effect: Four-Flapper Valve Resistance

First, consider the airway resistance offered by the valve portion of the prosthesis (open squares). Airway resistance for this component decreased as a function of flow rate. Average resistance values ranged from 179.4 cm H_2O/LPS at 0.05 LPS to 152.7 cm H_2O/LPS at 0.20 LPS. The variability, expressed in terms of standard deviation, was about 30% of the mean across the four sampled flow rates. These results conform with the predicted device response. Namely, the valves opened more widely with increasing air flow rates through them.

Air Entrance Effect: Air Port Resistance

Next, consider the airway resistance offered by the air entrance component (open circles). Airway resistance for this component increased as a function of increasing air flow rate. Average resistance values ranged from 4.9 cm H_2O/LPS at 0.05 LPS to 35.8 cm H_2O/LPS at 0.20 LPS. Variability in airway resistance expressed in terms of standard deviation was approximately 7% of the mean across the four sampled flow rates.

The increase in air port resistance with increasing flow rate was expected. Since the size of the air entrance port remains constant and airflow through the entrance port has a turbulent component, the pressure drop across the air port of Voice Buttons can be described as a quadratic function. Hence, the pressure drop is proportional to a constant times the square of velocity through the port ($p = k\,\dot{v}^2$).

Total Device Resistance

Finally, consider the total airway resistance offered by these devices (closed circles). Total resistance represents the combined, relative influence of the air entrance and exit effects. Total airway resistance ranged from 177 cm H_2O/LPS at 0.05 LPS to approximately 200 cm H_2O/LPS at each of the remaining air flow rates.

Total airway resistance for individual devices ranged from 93 cm H_2O/LPS to 317 cm H_2O/LPS, indicating that overall resistance was 2.6–9 times that offered by the normal larynx during vowel production (Smitheran & Hixon, 1981).

COMMENTS

Average resistance values (range 153–179 cm H_2O/LPS) for the valve portion of Voice Button prostheses examined here were substantially less than those established previously. For example, Weinberg (1982) calculated the airway resistance offered by the valve portion of 4 Voice Button prostheses. He reported a range of 285–440 cm H_2O/LPS in this small sample of devices. In an attempt to identify the origin of this discrepancy, measurements were made of the valve slit lengths in prostheses used in both the present project and in the Weinberg (1982) study. The results of this analysis revealed that, on the average, valve slits of prostheses used in the present project were 24% longer than those of values used in the Weinberg (1982) project. This increase in slit length (i.e., valve flapper dimensions) would be expected to permit increased valve opening and to lower airway resistance. The Voice Buttons used in both of these projects were obtained directly from the manufacturer. Devices used in the Weinberg (1982) project were obtained in 1981, whereas the devices used in the present project were obtained in 1983. We have not been able to determine whether the increase in average slit length measured in more recently acquired devices represents sampling bias or intentional alteration in the manufacturing process and design of Voice Button prostheses.

Airway resistance characteristics of the Voice Button are compared with those of Blom-Singer tracheoesophageal prostheses in Figure 3. Resistance properties for both devices were calculated on an identical basis using the same equipment in our laboratory. These data show that the total opposition to airflow through Voice Button devices exceeded that offered by Blom-Singer devices. Overall mean resistance of Voice Button prostheses was 1.5 times that of Blom-Singer prostheses. The difference in the resistive load of Voice Button prostheses can clearly be attributed to differences in the design of the air exit (valve) portion of these devices. Resistance values calculated for the air entrance port were roughly comparable (see Figure 3 for details).

The comparative data illustrated in Figure 3 also show that Voice Button performance was considerably more variable. In an attempt to identify the origin of this variability we did two things. First, we carefully inspected devices on a direct and microscopic (25×) basis. Visual inspection was used to determine whether the four-flapper valves were "cut on center." Second, we measured the lengths of slits bounding each of the flappers on each of the devices tested to determine whether the boundaries of each of the four flappers of a given valve were of equal length.

These observations revealed that flapper valves of 7 of the 10 Panje devices were not "cut on center." As indicated previously, each prosthesis has a four-flapper valve and each flapper is bounded by two slits. Measurement of slit lengths revealed that none of the prostheses had boundaries (four slits) of equal length. In only 7 of the 40 (10 devices × 4 flappers) flappers measured were the slits bounding a given flapper of equal length. These observations suggest considerable variation in the dimensions of a critical component of Voice Button prostheses which may explain the variability in performance noted in these devices.

As indicated earlier, all tracheoesophageal prostheses should, from a design perspective, be minimally resistant to airflow through them from the trachea to the esophagus. This opposition to airflow constitutes one important resistive load laryngectomized patients must overcome during voice/speech production.

The magnitude of such loads should be minimized to reduce the respiratory effort required to produce voice/speech and to facilitate the use of a tracheal breathing valve.

The results of this project reveal that Voice Button prostheses offer more resistance to airflow than Blom-Singer devices and that the performance of Voice Button prostheses is substantially more variable. These observations suggest (a) the need for modification in prosthetic design and performance and (b) the need to carefully study patient response to these variations in device characteristics.

Jerald Moon
Bernd Weinberg
Purdue University
West Lafayette, Indiana

REFERENCES

MOON, J. B., SULLIVAN, J., & WEINBERG, B. (1983). Evaluation of Blom-Singer tracheoesophageal puncture prostheses performance. *Journal of Speech and Hearing Research, 26,* 459–464.

PANJE, W. R. (1981). Prosthetic vocal rehabilitation following laryngectomy: The Voice Button. *Annals of Otology, Rhinology, and Laryngology, 90,* 116–120.

SMITHERAN, J. R., & HIXON, T. J. (1981). A clinical method for estimating laryngeal airway resistance during vowel production. *Journal of Speech and Hearing Disorders, 46,* 138–146.

WEINBERG, B. (1982). Airway resistance of the Voice Button. *Archives of Otolaryngology, 108,* 498–500.

Received April 15, 1983
Accepted April 6, 1984

Fragile X Syndrome: Its Relations to Speech and Language Disorders

A recently identified chromosome abnormality has been associated with some forms of X-linked inheritance of mental retardation in males. Speech and language disorders are thought to be a major concomitant of this type of mental retardation. In this paper we describe linguistic and clinical aspects of this syndrome and suggest some of its implications for research in communication disorders.

A BRIEF HISTORY

It has been known since the nineteenth century that there are a greater number of intellectually impaired males than females, with approximately 25% more males (Lehrke, 1974). A variety of studies (summarized by Lehrke, 1974) have confirmed the presence of a greater proportion of males than females among the mentally retarded, in both the general and institutional populations. In 1943, Martin and Bell reported a pedigree in which 11 males within two generations in one family were affected with nonspecific retardation, that is, retardation with no associated physical disorders or anomalies. Several later reports (Allan, Herndon, & Dudley, 1944; Dunn, Renpenning, & Gerard, 1963; Renpenning, Gerrard, Zaleski, & Tabata, 1962) identified similar families, and it was shown that the retardation followed an X-linked pattern of inheritance. X-linked recessive inheritance patterns (such as hemophilia and color blindness) are those in which a nonaffected carrier mother passes a mutant gene on one of her two X chromosomes to 50% of her sons, who are then affected with the condition, and to 50% of her daughters, who may be unaffected carriers and continue the X-linked inheritance pattern. In these conditions, girls presumably are not affected or perhaps mildly affected, because some of the defect carried on the X chromosome with the mutant gene they received from their mother is compensated for by information on the normal X chromosome they received from their father. When boys, on the other hand, receive an affected X chromosome from their mother, the Y chromosome from the father does not provide any compensatory information, and therefore the defect associated with the mutant gene on the one X chromosome is fully expressed.

X-linked mental retardation has been found to be quite common. Turner and Turner (1974) estimated that X-linked genes could be responsible for 20% or more of the moderately retarded male population. This form of intellectual impairment may, then, be second only to Down's syndrome in prevalence.

In 1969, the first finding of structural abnormalities of X chromosomes in the males of a family with an X-linked pattern of inheritance of mental retardation was reported (Lubs, 1969). The tips of the long arm of these X chromosomes appeared pinched or constricted. All four retarded males in three generations in the family studied demonstrated this abnormal X chromosome, as did the mother of the two brothers in the most recent generation. This family remained an intriguing but unreplicated observation for 7 years, until reports of similar findings emerged from France and Australia (Giraud, Ayme, Mattei, & Mattei, 1976; Harvey, Judge, & Wiener, 1977). Sutherland (1977) was the first to realize that in order to observe the marker X chromosome, it was necessary to grow cells to be tested in a medium deficient in folate, one of the B vitamins. Even under the most favorable conditions, though, the marker site never appears in all cells; rarely does the proportion of cells showing the marker exceed 50%. Sutherland suggested the use of the term *fragile site* to refer to this X chromosome abnormality, although some geneticists prefer the less specific term *marker*. Although Turner and Opitz (1980) have proposed the existence of several types of X-linked mental retardation based on a combination of clinical and cytogenetic criteria, the term *fragile X syndrome* as it is commonly used refers to those forms of X-linked mental retardation in which this marker X chromosome is identified, using the culture conditions known to elicit it, in the cells of affected males. Not all families with X-linked patterns of inheritance of mental retardation exhibit these marker X chromosomes when tested (Herbst, Dunn, Dill, Kalousek, & Krywanink, 1981), and it is not yet clear whether or not the fragile X and "nonfragile X" forms of X-linked retardation represent distinct syndromes. The significance of the marker X chromosome, that is, the means by which it affects intellectual development, is completely unknown. The facts that (a) the marker appears only under particular culture conditions and (b) it never appears in all cells are observations that puzzle geneticists and make it difficult to know whether fragile X syndrome is truly a unique form of X-linked mental retardation or simply a cytogenetic artifact.

Clinical features. Turner and Frost (1980) have presented a clinical picture of fragile X syndrome. Despite the generally nonspecific character of their retardation, males affected with fragile X syndrome have been found to show some features in common, most of which consist of mildly excessive growth. Head circumference may be large in infancy, ears are usually longer than normal, lower jaw may be somewhat large. These features are not present in affected males in all fragile X families, though. Testicular enlargement, or macroorchidism, is almost universally seen in postpubertal males (Escalante, Grunspun, & Frota-Pessoa, 1971; Howard-Peebles & Stoddard, 1979; Sutherland & Ashforth, 1979; Turner, Eastman, Casey, McLeay, Procopis, & Turner, 1975), although the size of the testes in prepubertal boys with fragile X syndrome is more variable.

The degree of retardation among males affected with fragile X syndrome can vary widely, although the majority function in the moderately retarded range whereas others are mildly or severely impaired. The incidence of fragile X syndrome in profoundly retarded individuals has not yet been investigated. Further, a few males with the marker X chromosome have been reported to show normal intelligence (Daker, Chidiac, Fear, & Berry, 1981; Webb, Rogers, Pitt, Halliday, & Theobald, 1981), but these reports are controversial. Fragile X syndrome has been identified in a number of ethnic groups throughout the world.

Although females with the fragile X chromosome are generally

unaffected carriers, at least one third appear to be mildly retarded or learning disabled (Jacobs et al., 1980; Turner, Brookwell, Daniel, Selikowitz, & Zilibowitz, 1980). When an X-linked pattern of inheritance is established in a family by means of a pedigree study, mothers of males with fragile X syndrome are considered obligate carriers. Many of these women do not, however, exhibit the marker X when chromosome studies are done.

In addition to general mental retardation, most of the males affected with fragile X syndrome show delayed motor and speech development. Speech has been reported to be particularly disordered (Allan et al., 1944; Deroover, Fryns, Parloir, & VanDenBerghe, 1977; Dunn et al., 1963; Lehrke, 1974; Martin & Bell, 1943; Renpenning et al., 1962; Snyder & Robinson, 1969; Yarborough & Howard-Peebles, 1976). Speech in affected males has been described as "perseverative," having a characteristic "litany-like" intonation, "jocose," "limited," and "defective."

The specificity of the verbal disabilities in fragile X syndrome is not yet clear. Howard-Peebles, Stoddard, & Mims (1979) studied verbal skills in 13 individuals from four families showing an X-linked pattern of inheritance of mental retardation. Only one of these families exhibited fragile X syndrome. All subjects were found to have language deficiencies on the Utah Test of Language Development (Mecham, Rex, & Jones, 1969). On the Illinois Test of Psycholinguistic Abilities (Kirk, McCarthy, & Kirk, 1968), all were found to have more strengths in nonverbal than verbal channels. The family with the fragile X chromosome differed from the others in being somewhat stronger in auditory reception and somewhat weaker in manual expression. Articulation errors identified in the subjects by means of the Goldman-Fristoe Test of Articulation (Goldman & Fristoe, 1969) resembled those seen in Down's syndrome children and in nonimpaired children during the developmental period. The authors concluded that all subjects in the study showed a generalized, rather than a specific, language disability and that the tools used showed little promise of differentiating a verbal disability unique to fragile X syndrome. Herbst et al. (1981) found "perseverative speech" in affected males from six families with X-linked mental retardation, three of whom expressed the fragile X marker. All were found to have fluent conversation but showed difficulty in conversing about suggested topics, defining words, and producing sentences to express thoughts appropriate to their mental age. Those with the marker generally showed somewhat poorer performance on the Peabody Picture Vocabulary Test (Dunn, 1965) than those without, but there were some exceptions.

The subjects of these studies were generally mentally retarded adults who had been living for some time in institutions. Because most studies have been conducted in institutions for the retarded where few children under the age of 14 are now in residence, few reports of young boys with fragile X syndrome are available. One observation that has been made in children and adolescents with fragile X syndrome is that some show autistic features (Brown et al., 1982; Meryash, Szymanski, & Gerald, 1982). Some young males with fragile X syndrome are reported to be hyperactive as well, but by adulthood these behavioral features become less marked. Adults with fragile X are generally considered well-behaved and easily manageable residents of institutions for the retarded.

CASE REPORTS

Two families with three young boys affected with fragile X syndrome are presented to convey an idea of their language and related characteristics. These boys were referred for language and behavioral evaluation by their teachers or physicians and were subsequently found to have fragile X syndrome as a result of comprehensive medical evaluations carried out in conjunction with these referrals. All were given standard speech and language evaluations, including an analysis of a free speech sample derived from a videotape of a 15-minute free play interaction.

Family 1

P_1. P_1 was 13:9 (yrs:mos) at the time of his evaluation, when he was referred by his classroom teacher for hyperactivity and behavioral problems. He lived at home with his younger brother (P_2), sister, and parents, and had been enrolled in special education since the age of 5.

Physical findings: P_1 had large ears. EEG was normal. He was going through puberty and refused to have his testes examined.

Nonverbal IQ: P_1 received an age equivalent score of 5:6 on the Leiter International Performance Scale (Arthur, 1952). His nonverbal IQ score was 40.

Speech and language: Language assessment results are given in Table 1. Receptive language age was commensurate with nonverbal developmental level. P_1's expressive language age score as measured by both DSS (Lee, 1974) and MLU (Brown, 1973) was significantly lower than mental or receptive language age. His low DSS score was related to a lack of complexity in his utterances, specifically in verb marking, sentence embedding, and conjoining. On the DSS, 16% of his utterances contained main verb forms scored at 4—*can, will, may,* or *do* auxiliaries—but none received higher scores for verb marking. Only 10% of his utterances contained secondary verbs. He used only one conjunction and two interrogative reversals out of five questions. Sentence points were earned by 86% of his utterances. Four of his utterances earned attempt marks.

On the Photo Articulation Test (Pendergast, Dickey, Selmar, & Soder, 1969), P_1's only error was consistent /r/ distortions on initial position and /r/-colored vowels in final position. On Shriberg and Kwiatkowski's Natural Process Analysis (1980), P_1 used Liquid Simplification consistently. None of the other processes was evident. In addition, 10% of P_1's utterances were unintelligible. He also showed some degree of disfluency—on 2% of the syllables—consisting of initial phoneme repetitions, but not at a level sufficient to consider P_1 a stutterer. Speech motor abilities were evaluated using the Screening Test for Developmental Apraxia of Speech (Blakely, 1980). P_1 needed tactile cues in order to lateralize his tongue and was unable to raise the tongue. Lip protrusion and retraction were normal. Diadochokinetic rates in reduplicative syllables (/pʌpʌpʌ . . ./) were compatible with mental age expectations. When asked to repeat rapidly a nonreduplicate series of syllables such as /pʌtʌkʌ/, P_1 reversed the order of syllables. He deleted unstressed syllables in polysyllabic words such as *linoleum.*

In conversation, P_1 answered questions appropriately and seemed to enjoy interacting with the examiner, although he frequently made statements such as "I'm so nervous" and "I have a headache." He seemed to enjoy ejaculating "Rats!" with great gusto at frequent intervals.

Behavior: P_1 was highly excitable and hyperactive. He showed hand flapping and facial grimacing and, unless deeply engaged in an activity, had difficulty remaining still. Play was somewhat rigid and repetitive. Most play with toys involved monologue, rather than interaction.

Cytogenetics: Marker X chromosome was observed in 26% of cells examined using a folate-deficient medium and a 96-hour culture time (Howard-Peebles & Stoddard, 1979).

P_2. P_2 was 10:6 at the time of the evaluation. He was referred by his mother in conjunction with his older brother's evaluation.

Physical findings: P_2 had large ears. EEG was normal. Testes were of normal size.

Nonverbal IQ: P_2 received an age-equivalent score of 5:3 and a nonverbal IQ of 50 on the Leiter International Performance Scale (Arthur, 1952).

Speech and language: As seen in Table 1, P_2's receptive language age score was commensurate with nonverbal mental age. Productive syntax was, again, at a much lower level than nonverbal or receptive scores.

P_2's DDS showed that 18% of his utterances earned only attempt marks in the main verb category, and 10% earned only attempts in the personal pronoun category. Only 14% of his utterances contained secondary verbs, and all of these were early-developing infinitives scored at 2 points. All but four of his

TABLE 1. Characteristics of three boys with fragile X syndrome.

	Subject		
	P_1	P_2	M_1
Chronological age	13:9	10:6	10:0
Cognitive performance			
Nonverbal IQ	40[a]	50[a]	70[b]
Nonverbal Mental Age	5:6	5:3	7:0
Speech-language performance			
Comprehension Age[c]	6:5	5:9	6:7
MLU (in morphemes)	3.5	3.8	3.2
MLU age-equivalent (Miller, 1981)	3:3	4:5	2:9
DSS[d]	5.5	4.4	4.4
DSS[d] age-equivalent	3:3	2:9	2:9
Sounds in error: isolated words	/r/	/r/	/l/, /r/, /s/
Phonological processes: connected speech	Liquid Simplification (/r/)	Final Consonant Deletion Liquid Simplification Stopping	Liquid Simplification (/r/, /l/)
% unintelligible utterances	10	18	14
% disfluent syllables	2	4	14
Genetic findings			
% cells showing marker X chromosome	26	21	28
Mother showed marker?	yes	yes	no
Siblings showing marker	sister	sister	brother, sister

[a]Leiter International Performance Scale (Arthur, 1952).
[b]WISC-R Performance Scale (Wechsler, 1974).
[c]Test for Auditory Comprehension of Language (Carrow, 1973).
[d]Developmental Sentence Score (Lee, 1974).

scorable main verbs were marked with simple third person singular or past tense morphemes without any auxiliary and earned scores of 2 or less. He used only one conjunction and no interrogative reversals out of four questions. Only 60% of his utterances earned sentence points for overall grammaticality. His low score, then, was due to both errors of verb and pronoun marking and to limited complexity.

P_2 substituted /w/ for /r/ in initial position on the Photo Articulation Test (Pendergast et al., 1969) and produced /r/-colored vowels in final position. On Natural Process Analysis (NPA), he used Liquid Simplification consistently and also occasionally used Stopping and Final Consonant Deletion. Disfluencies, consisting of initial consonant repetitions, were noted on 4% of his syllables, although, again, P_2 was not considered a stutterer. Eighteen percent of his utterances were unintelligible.

Oral motor examination, using the Screening Test for Developmental Apraxia of Speech (Blakely, 1980) showed slowness on tongue lateralization. P_2 was unable to protrude his lips without using his hands. Diadochokinetic rates in reduplicative syllables were compatible with mental age norms. When asked to repeat nonreduplicative syllable combinations rapidly, P_2 simplified the combination to two syllables, reversed the order of syllables, or repeated them at a reduced rate. He simplified phonemes in polysyllabic words, pronouncing *linoleum* as /mənomiʌm/, for example.

In conversation, P_2 answered questions appropriately, requested information, and used language to structure pretend play. He enjoyed repeating his favorite expression, "Pits!"

Behavior: Play was very similar to that of P_1. He became engaged in a pretend script with objects but was primarily involved in monologue, rather than interactive play.

P_2 was also hyperactive, although somewhat less so than his brother. P_2's affect was highly labile; he switched from laughter to crying within minutes without noticeable explanation. He also perseverated with vulgarisms.

Cytogenetics: Chromosome studies showed that 21% of P_2's cells displayed the marker X chromosome.

Family data: Pedigree studies showed no other intellectually impaired relatives. The P boys' mother and sister also exhibited marker X chromosomes, using the above culture methods. The sister was repeating her kindergarten year for "immaturity."

Family 2

M_1. M_1 was referred for evaluation by the family psychiatrist because of his perseverative, disfluent speech. He was 10:0 at the time of the referral, lived with his parents, younger brother, and sister, and had been in special education programs since the age of 3.

Physical findings: M_1 had large, slightly asymmetric ears and no enlargement of the testicles. EEG was normal.

Nonverbal IQ: M_1's nonverbal IQ was measured on the Performance Scale of the WISC-R (Wechsler, 1974) by his school psychologist. His Performance IQ was 70, with a mental age of 7:0.

Speech and language: As seen in Table 1, receptive language age was close to that expected for mental age level. Expressive syntax was below that expected for nonverbal mental and receptive language age.

On the DSS attempt marks were given on 20% of the main verbs. Like P_1, M_1 used main verbs marked with only the simplest inflections in all but one of the utterances that could be scored. Only 18% of his sentences contained secondary verbs, and all but three of these were the simplest early-developing infinitivals. He used only two conjunctions and one interrogative reversal out of seven questions. Sentence points were earned by 65% of his utterances.

On the Photo Articulation Test, M_1 showed distortion and substitution errors on /r/, /l/, and /s/. Natural Process Analysis showed Liquid Simplification on /l/ and /r/. Fourteen percent of M_1's utterances were unintelligible. A relatively large proportion—14%—of his syllables contained disfluencies involving initial phoneme repetitions, some of which involved the addition of the vowel /ʌ/ before a consonant, as in /ʌbʌ,ʌbʌ,ʌbʌ,bɪg/. M_1 also showed prolongations of 1–2 seconds (s) on a few initial consonants and occasional blocks of about 2 s in length on initial plosives. M_1 was considered by his school speech-language pathologist to have a stuttering problem, although the major focus of his therapy was on expressive language.

Speech-motor evaluation showed M_1 to be clumsy in his imitation of lip and tongue movements. He could lateralize the tongue, but movement to the left was easier for him and produced less overflow. Lip protrusion and retraction were normal. Diadochokinetic rates in reduplicative syllables were commensurate with mental age expectations. When asked to produce rapid repetitions of nonreduplicative sequences of syllables, M_1 simplified the vocalization to two syllables or produced the sequence only once. When asked to imitate polysyllabic words, he refused, saying they were "too hard."

M_1's conversation was appropriate but sparse and immature. Most of his spontaneous remarks consisted of asking his mother when he could go home. He seemed to enjoy using "jeepers" to punctuate his conversation and found saying the word so amusing that he giggled over it. He used language to request and comment, and when asked questions by his mother he could relate information about past and future events. Asked to tell a story, because his spontaneous utterances were somewhat infrequent, he chose "Goldilocks." He had difficulty sequencing and elaborating the events in the story, although he seemed to enjoy reciting the verbal formulas ("Too big, too small, just right").

Behavior: M_1's behavior was markedly immature. He tended to behave in a "silly" manner, often becoming uncontrollably giddy. He was somewhat hyperactive and had trouble persisting in tasks.

Cytogenetics: M_1's chromosome studies showed that 28% of the cells examined had the marker X chromosome.

Family data: M_1's brother, age 14 months, was found to be positive for fragile X syndrome with 18% of the cells examined showing the marker X. The younger boy had been perceived by the parents to be developing normally in all areas except for speech. He had no words and produced few vocalizations. M_1's sister, age 6, was also found to show the marker X chromosome in 14% of the cells examined. She was considered bright and was doing well in a regular classroom. M_1's mother did not show the marker X in any of the cells examined, despite her status as an obligate carrier.

ISSUES FOR SPEECH AND LANGUAGE RESEARCH

Reports of speech and language disorders in fragile X males are provocative, but as yet no specific features of communication disorders have been identified in this population. It is noteworthy that the language profiles of the three boys reported here are remarkably alike. Although one might expect the two brothers to resemble each other, even the unrelated boy presented a strikingly similar picture with respect to language, fluency, and speech intelligibility. These similarities include (a) mild to moderate mental retardation, (b) poorer performance on productive syntax than on receptive language or nonverbal tests, (c) poor intelligibility in connected speech despite good performance on single words in articulation tests and the use of a very limited number of phonological simplification processes, (d) poor oral and vocal imitation performance, and (e) disfluency. This constellation of deficits is consistent with descriptions of the speech characteristics of children of normal intelligence who are thought to display a developmental apraxia of speech. These characteristics include:

1. disfluencies consisting of prolongations and repetitions of sounds and syllables (Yoss & Darley, 1974);
2. adequate production of sounds and words in isolation, with connected speech less intelligible than would be predicted on the basis of articulation testing (Rosenbek & Wertz, 1972);
3. incorrect sequencing in tasks involving imitation of nonreduplicative syllables, such as /pʌtʌkʌ/ (Yoss & Darley, 1974);
4. greater difficulty in the production of polysyllabic than mono- or bisyllabic words (Morley, 1972);
5. concomitant deficits in expressive language, while receptive abilities are significantly superior (Jaffe, 1984).

These apraxic characteristics may reflect an underlying impairment in the capacity for formulating and executing speech that is expressed across all levels of linguistic encoding.

In order to substantiate such speculation, it will be necessary, first of all, to establish that there are, in fact, characteristic language deficits in persons with fragile X syndrome. Unfortunately, it cannot yet be demonstrated that the language disorders seen in this group are distinct from the general range of individual differences in language achievement observed in the retarded population. Comparisons of language performance in matched groups of fragile X and non-fragile X males with intellectual impairment are currently proceeding in our own center. It will be important, too, to know whether there are linguistic differences between males who show fragile X syndrome and those with other forms of X-linked mental retardation who do not. Such studies would help to determine whether fragile X syndrome is a truly distinct variety of X-linked retardation.

There is a danger in jumping too quickly to conclusions about the clinical features of fragile X syndrome, based on the small amount of information currently available. Even the apparent association of apraxic-like disorders with fragile X bears more careful investigation before it can be incorporated into the definition of the syndrome. Much more research on this population, using appropriate contrast groups, is needed in order to delineate features accurately. Still, clinicians encountering children with a constellation of physical, speech, and language findings similar to that reported here may wish to suggest a referral for genetic counseling and evaluation, particularly if there is more than one affected individual in a family. And despite the current uncertainties, the identification of a biological marker of a potentially specific cognitive-linguistic deficit is clear. This provides a unique opportunity for specialists in developmental disorders of speech and language to collaborate in investigation of the biological bases of the behaviors that we work toward understanding and ameliorating.

ACKNOWLEDGMENTS

We wish to thank Anthony Bashir and James Leckman for their critical comments. This research was supported by the NIMH Mental Health Clinical Research Center grant #MH30929, the Children's Clinical Research Center grant #RR00125, NICHD grant #03008, Mr. Leonard Berger, U.S. Public Health Service research grant HD-11624, and the Merck Foundation.

Rhea Paul
Yale Child Study Center, New Haven, CT

Donald J. Cohen
Yale Child Study Center, New Haven, CT
Department of Pediatrics, Yale School of Medicine

W. Roy Breg
Department of Pediatrics, Yale School of Medicine

Michael Watson
Department of Human Genetics, Yale School of Medicine

Steven Herman
Yale Child Study Center, New Haven, CT

REFERENCES

ALLAN, W., HERNDON, C., & DUDLEY, F. (1944). Some examples of the inheritance of mental deficiency: Apparently sex-linked idiocy and microcephaly. *American Journal of Mental Deficiency, 48,* 325–334.

ARTHUR, G. (1952). *The Arthur Adaptation of the Leiter International Performance Scale.* Washington, DC: Psychological Service Center Press.

BLAKELEY, R. (1980). *Screening Test for Developmental Apraxia of Speech.* Tigard, OR: C. C. Publications.

BROWN, R. (1973). *A first language.* Cambridge, MA: Harvard University Press.

BROWN, W., FRIEDMAN, E., JENKENS, E., BROOKS, J., WISNIEWSKI, K., RAGUTHU, S., & FRENCH, J. (1982). Association of fragile X syndrome with autism. *Lancet, 1,* 100. (Letter).

CARROW, E. (1973). *Test of Auditory Comprehension of Language.* Hingham, MA: Teaching Resources.

DAKER, M. G., CHIDIAC, P., FEAR, C. N., & BERRY, A. (1981). Fragile X in normal male. A cautionary tale. *Lancet, 1,* 780. (Letter).

DEROOVER, J., FRYNS, J. P., PARLOIR, C., & VANDENBERGHE, H. (1977). X-linked recessively inherited non-specific mental retardation. *Annales de Genetique, 20,* 263–268.

DUNN, H., RENPENNING, H., & GERRARD, J. (1963). Mental retardation as a sex-linked defect. *American Journal of Mental Deficiency, 67,* 827–843.

DUNN, L. M. (1965). *Peabody Picture Vocabulary Test.* Circle Pines, MI: American Guidance Service.

ESCALANTE, J. A., GRUNSPUN, H., & FROTA-PESSOA, O. (1971). Severe sex-linked mental retardation. *Journal de Genetique Humaine, 19,* 137–140.

GIRAUD, F., AYME, S., MATTEI, F., & MATTEI, M. G. (1976). Constitutional chromosomal breakage. *Human Genetics, 34,* 125–136.

GOLDMAN, R., & FRISTOE, M. (1969). *Test of Articulation.* Circle Pines, MI: American Guidance Service.

HARVEY, J., JUDGE, C., & WIENER, S. (1977). Familial X-linked mental retardation with an X-chromosome abnormality. *Journal of Medical Genetics, 14,* 46–50.

HERBST, D., DUNN, H., DILL, F., KALOUSEK, D., & KRYWANINK, L. (1981). Further delineation of X-linked mental retardation. *Human Genetics, 58,* 366–372.

HOWARD-PEEBLES, P. N., & STODDARD, G. R. (1979). X-linked mental retardation with macro-orchidism and marker X-chromosomes. *Human Genetics, 50,* 247–251.

HOWARD-PEEBLES, P. N., STODDARD, G. R., & MIMS, M. (1979). Familial X-linked mental retardation, verbal disability, and marker X chromosomes. *American Journal of Human Genetics, 31,* 214–222.

JACOBS, P. A., GLOVER, T. W., MEYER, M., FOX, D., GERRARD, J. W., DUNN, H. G., & HERBST, D. S. (1980). X-linked mental retardation: A study of 7 families. *American Journal of Medical Genetics, 7,* 171–189.

JAFFE, M. (1984). Neurological impairment of speech production: Assessment and treatment. In J. Costello (Ed.), *Speech disorders in children.* San Diego: CA: College Hill Press.

KIRK, S., MCCARTHY, J., & KIRK, W. (1968). *The Illinois Test of Psycholinguistic Abilities* (rev. ed.). Urbana, IL: University of Illinois Press.

LEE, L. (1974). *Developmental Sentence Analysis.* Evanston, IL: Northwestern University Press.

LEHRKE, R. (1974). *X-linked mental retardation and verbal disability.* National Foundation March of Dimes. New York: Intecontinental Medical Book Corporation.

LUBS, H. (1969). A marker X chromosome. *American Journal of Human Genetics, 21,* 231–244.

MARTIN, J., & BELL, J. (1943). A pedigree of mental defect showing sex linkage. *Journal of Neurological Psychiatry, 6,* 154–157.

MECHAM, M., REX, J., & JONES, J. (1969). *Utah Test of Language Development.* Salt Lake City: Communication Research Associated.

MERYASH, D. L., SZYMANSKI, L. S., & GERALD, P. S. (1982). Infantile autism associated with the Fragile X syndrome. *Journal of Autism and Developmental Disorders, 12,* 294–301.

MILLER, J. (1981). *Assessing language production in children: Experimental procedures.* Baltimore: University Park Press.

MORLEY, M. (1972). *Development and disorders of speech in childhood.* Edinburgh: Churchill Livingstone.

PENDERGAST, K., DICKEY, S., SELMAR, J., & SODER, A. (1969). *The Photo Articulation Test* (2nd. ed.). Danville, IL: Interstate.

RENPENNING, H., GERRARD, J., ZALESKI, W., & TABATA, T. (1962). Familial sex-linked mental retardation. *Canadian Medical Association Journal, 87,* 954–956.

ROSENBEK, J., & WERTZ, R. (1972). A review of fifty cases of developmental apraxia of speech. *Language, Speech, and Hearing Services in Schools, 3,* 23–33.

SHRIBERG, L., & KWIATKOWSKI, J. (1980). *Natural process analysis of continuous speech samples.* New York: Wiley.

SNYDER, R., & ROBINSON, A. (1969). Recessive sex-linked mental retardation in the absence of other recognizable abnormalities. *Clinical Pediatrics, 8,* 669–674.

SUTHERLAND, G. R. (1977). Fragile sites on human chromosomes: Demonstration of their dependence on the type of tissue culture medium. *Science, 197,* 265–266.

SUTHERLAND, G. R., & ASHFORTH, P. L. (1979). X-linked mental retardation with macroorchidism and the fragile site Xq27 or 28. *Human Genetics, 48,* 117–120.

TURNER, G., BROOKWELL, R., DANIEL, A., SELIKOWITZ, M., & ZILIBOWITZ, M. (1980). Heterozygous expression of X-linked mental retardation and X-chromosome marker fra(X) (q27). *New England Journal of Medicine, 303,* 662–664.

TURNER, G., EASTMAN, E., CASEY, J., MCLEAY, A., PROCOPIS, P., & TURNER, B. (1975). X-linked mental retardation associated with macro-orchidism. *Journal of Medical Genetics, 12,* 367–371.

TURNER, G., & FROST, D. (1980). X-linked mental retardation, macroorchidism, and the Xq27 fragile site. *Journal of Pediatrics, 96,* 837–841.

TURNER, G., & OPITZ, J. M. (1980). Editorial comment: X-linked mental retardation. *American Journal of Medical Genetics, 7,* 407–415.

TURNER, G., & TURNER, B. (1974). X-linked mental retardation. *Journal of Medical Genetics, 11,* 109–113.

WEBB, G. C., ROGERS, J. G., PITT, D. B., HALLIDAY, J., & THEOBALD, T. (1981). Transmission of fragile (X) (q27) site from a male. *Lancet, 2,* 1231–1232. (Letter).

WECHSLER, D. (1974). *Wechsler Intelligence Scale for Children—Revised Version.* New York: The Psychological Corporation.

YARBOROUGH, K. M., & HOWARD-PEEBLES, P. N. (1976). X-linked nonspecific mental retardation: Report of a large kindred. *Clinical Genetics, 9,* 125–130.

YOSS, K., & DARLEY, F. (1974). Therapy in developmental apraxia of speech. *Language, Speech, and Hearing Services in Schools, 1,* 23–31.

Received February 5, 1983
Accepted May 15, 1984

Postmortem examination: Shriberg and Kwiatkowski (1982)

Recently, Shriberg and Kwiatkowski (1982b) published the results of a series of three studies evaluating four structural management modes—Drill, Drill Play, Structured Play, and Play. The authors reported that "Drill and Drill Play modes are more effective and more efficient than Structural Play and Play modes" (p. 249, Studies A & B), and that clinicians preferred Drill Play whereas they perceived their clients to prefer Play, Structured Play, and Drill Play (Studies B & C). Also, "these data refute a position that stimulus-response paradigms (Drill) should be selected as the management structure of choice. . . . [A] certain element of play (Drill Play, Structured Play, or Play) appears to be not only defensible, but in some situations, preferable to Drill" (p. 251). Thus, choosing the structure of a management program seems as important as content selection. Shriberg and Kwiatkowski base these conclusions on statistically significant pair-wise comparisons of the four management modes, following nonparametric Wilcoxon Signed-Ranks tests on mean data. Their use of this nonparametric statistic and subsequent interpretation of obtained results warrant further comment and caution.

Both parametric and nonparametric models recognize a Type I error as rejection of the null hypothesis when it is in fact true. Researchers guard against Type I errors by careful selection of an appropriate significance level (α). Beyond this selected α value, the null hypothesis is rejected, and the probability of committing a Type I error is that significance value. There is a level of confidence that rejection of the null is truly false and not a chance occurrence. Selecting a critical value a priori is simple and appropriate when only one hypothesis with one comparison is tested. When multiple comparisons are made on the same data, as in the case of Shriberg and Kwiatkowski's comparisons, other conceptual units of error must be considered.

Shriberg and Kwiatkowski selected comparison-wise units of error rate; the probability that any comparison was significant was .05. By controlling comparison-wise error rate, the resultant experiment-wise error rate varies as a function of the number of comparisons made. That is, as the number of comparisons increases, the probability that one or more comparisons is spuriously significant also increases. Using data from Study B, six comparisons of four management modes for four questionnaire items, or 24 total comparisons, were calculated. The authors, themselves, seemed quite surprised at the number of significant differences: "Overall, clinicians' independent rankings were remarkably similar, yielding even for this small number of clinicians several statistically significant differences in the rank orderings" (p. 250). The authors have allowed themselves 24 chances, rather than one, to reject the null hypothesis. This procedure has often been called " 'hunting with a shot-gun' for significant differences" (Kish, 1959, p. 336). Through sheer perserverance in constructing multiple comparisons, "significant" random events are highly probable.

In addition to repeated comparisons, Shriberg and Kwiatkowski treated each management mode as a discrete independent variable when, in fact, they were continuous. The authors even defined this continuum:

> The goal in conceptualizing the four structural modes was to define the scope of possible intervention structures *ranging from "drill" to "play"* . . . [and] the notion is that these four arrangements of the basic elements of management define the *range of possibilities* [from "drill" to "play"] available to clinicians. (pp. 245–246, italics mine)

Each mode, then, was a level of a single independent variable (Hinkle, Wiersma, & Jurs, 1979), management structure. When correlated levels of a variable are statistically evaluated as discrete, the "true" variability (found in independent observation) may be masked, and significance level may be biased.

Furthermore, the authors graphically reported ceiling effects (Figure 4). Specifically, mean rank of clinicians' perceptions of the most effective management mode was 4.0, on a 4-point scale. Clinicians unanimously judged Drill Play most effective. There was no variance for this level of the independent variable. If an independent variable is not free to vary, it is not a "variable," but a constant (Hinkle et al., 1979). Given this descriptive information, further inferences from statistical comparisons are irrelevant.

While the comparison-wise error rate is easy to use (Ryan, 1959), the experiment-wise error rate is preferred when more than one comparison is made (Ryan, 1959, 1962). The experiment-wise error rate guards against a Type I error for *all* possible comparisons. Also, "if the total set of conclusions is considered as a pattern supporting some theoretical position in such a way that any erroneous statement would destroy the pattern, then the experiment-wise basis is clearly the one to use" (Ryan, 1959, p. 38). Shriberg and Kwiatkowski's purpose in evaluation of management modes for clinical use obviously parallels this recommendation.

To estimate an experiment-wise error rate, one calculates $\alpha_E' = c(\alpha)$, where c = the number of independent comparisons (Kirk, 1968). For instance, in Study B six pair-wise comparisons are reported at an error rate of .05 for each of two variables. Given the experiment-wise error estimate, Shriberg and Kwiatkowski are risking a Type I error for all comparisons at a probability level considerably greater than .05. Significant relationships that are not occurring by chance alone cannot be ensured at the reported 95% confidence level. The selected α value is inflated. Had the authors opted a priori for an experiment-wise error rate of .05, then a comparison-wise error rate of .008 would have been the appropriate critical value ($\alpha_c' = \frac{\alpha_E}{c}$) (Kirk, 1968).

Given the facts that the authors (a) use an inflated comparison-wise value, (b) make multiple comparisons of overlapping and mean data, and (c) do not report exact significance levels or degrees of freedom, it is fair to ask whether reported significant relations can still be upheld with a more conservative a priori critical value of .008.

To hold *both* the comparison-wise and experiment-wise error rates equal to α, planned comparisons are most powerful. Planned orthogonal contrasts should be used when statistical hypotheses of interest to an experimenter can be determined a priori. For instance, Shriberg and Kwiatkowski spent considerable time developing a theoretical case for evaluation of management modes, including two related and supplementary articles (Shriberg & Kwiatkowski, 1982a, 1982c). Too, their review of literature pointed out that while "children did not like to 'drill' " (p. 245), their clinicians did not readily use an alternate structure, play. Given the lengthy rationale, it is unfortunate that the authors did not extend their reasoning to include planned rather than unplanned contrasts, such that only one form of play corresponded with each other mode (e.g., Structured Play vs. Drill, Structured Play vs. Drill Play, Structured Play vs. Play), resulting in use of nonoverlapping data and independent comparisons.

Finally, in lieu of an overly stringent a priori critical value or suggested planned orthogonal comparisons, it is proposed that "post-mortem" (McHugh & Ellis, 1955) or post hoc procedures be used to analyze these data and control for Type I error rate experiment-wise. Post hoc tests are designed to maintain error rate following rejection of the null hypothesis, with subsequent completion of a series of unplanned statistical comparisons. While well documented in most parametric texts (Hinkle et al., 1979; Kirk, 1968), post hoc and multiple comparison procedures for nonparametric statistics may not be as well known. However, they do exist (Marascuilo & McSweeney, 1977; McSweeney & Marascuilo, 1969).

Specifically, to evaluate effectiveness and efficiency of the four modes, it is recommended that a Friedman analysis of variance for k-related samples (Siegel, 1956, pp. 166–172) be used. The single null hypothesis suggested is: There is no significant difference between the four modes of management in terms of their efficiency (effectiveness). If the result of this analysis yields a significant difference, the null is rejected, and all six simple

comparisons (as well as any complex comparisons) may be analyzed using a nonparametric Scheffé procedure (Marascuilo & McSweeney, 1977). In this case, experiment-wise error rate is controlled, pair-wise comparisons are not made at extremely conservative a priori significance levels, and the confidence level avoiding a Type I error is maintained.

Multiple comparison procedures are recommended to evaluate clinicians' perceptions of the management modes. A test for main effects and interactions using a Friedman analysis of variance (Bradley, 1968; Marascuilo & McSweeney, 1977) is possible, with clinicians and management modes serving as the blocking variables,[1] and questionnaire items as the column variable. As above, error rate and Type I risk are controlled.

This critique serves as a challenge to Shriberg and Kwiatkowski to extend their data analysis, adopting the suggested conceptual unit of error and nonparametric post hoc/multiple comparison procedures. Borrowing from Skipper, Guenther, and Nass (1967; also *Publication Manual of the American Psychological Association*, 2nd ed., 1974), reporting the value of the test statistic, degrees of freedom, and exact probability level is recommended. We, the readers, can then serve as judges, determining whether "significant" results regarding Drill, Drill Play, Structured Play, and Play are plausible and constitute a basis for changing the structure of management programs.

Judith A. Gierut
Indiana University

[1]To minimize the size of error effects, Kirk (1968) suggested that subjects who are relatively homogeneous with respect to particular variables should be assigned to the same "block" or row. In this case, all those children assigned to the same management mode administered by the same clinician should be grouped in one block; hence, clinicians and management modes are "blocking variables."

REFERENCES

AMERICAN PSYCHOLOGICAL ASSOCIATION. (1974). *Publication manual of the American Psychological Association* (2nd ed.). Baltimore: Garamond/Pridemark.

BRADLEY, J. V. (1968). *Distribution-free statistical tests*. Englewood Cliffs, NJ: Prentice-Hall.

HINKLE, D. E., WIERSMA, W., & JURS, S. G. (1979). *Applied statistics for the behavioral sciences*. Boston: Houghton Mifflin.

KIRK, R. E. (1968). *Experimental design: Procedures for the behavioral sciences*. Belmont, CA: Brooks/Cole.

KISH, L. (1959). Some statistical problems in research design. *American Sociological Review, 24*, 328–338.

MARASCUILO, L. A., & MCSWEENEY, M. (1977). *Nonparametric and distribution-free methods for the social sciences*. Belmont, CA: Brooks/Cole.

MCHUGH, R. B., & ELLIS, D. S. (1955). The "post-mortem": Testing of experimental comparisons. *Psychological Bulletin, 52*, 425–428.

MCSWEENEY, M., & MARASCUILO, L. (1969). II. Nonparametric methods. *Review of Educational Research, 39*, 728–234.

RYAN, T. A. (1959). Multiple comparisons in psychological research. *Psychological Bulletin, 56*, 26–45.

RYAN, T. A. (1962). The experiment as the unit for computing rates of error. *Psychological Bulletin, 59*, 301–305.

SHRIBERG, L. D., & KWIATKOWSKI, J. (1982a). Phonological disorders I: A diagnostic classification system. *Journal of Speech and Hearing Disorders, 47*, 226–241.

SHRIBERG, L. D., & KWIATKOWSKI, J. (1982b). Phonological disorders II: A conceptual framework for management. *Journal of Speech and Hearing Disorders, 47*, 242–256.

SHRIBERG, L. D., & KWIATKOWSKI, J. (1982c). Phonological disorders III: A procedure assessing severity of involvement. *Journal of Speech and Hearing Disorders, 47*, 256–270.

SIEGEL, S. (1956). *Nonparametric statistics for the behavioral sciences*. New York: McGraw-Hill.

SKIPPER, J. K., JR., GUENTHER, A. L., & NASS, G. (1967). The sacredness of .05: A note concerning the uses of statistical levels of significance in social science. *American Sociologist, 2*, 16–18.

Received February 7, 1983
Accepted August 16, 1983

Corpus Delicti: Response to Gierut

Gierut's letter apparently has two objectives: to suggest a different conceptual and hence statistical approach to our data, and to provide a tutorial on materials generally covered in a second-level statistics course. Unfortunately, many of Gierut's factual assertions and suppositions towards both objectives are debatable, inconsistent, or incorrect. With regard to the attempted statistical tutorial, our own quantitative consultant indicated to us that there are numerous inaccuracies in Gierut's exposition (Joel R. Levin, personal communication; Marascuilo & Levin, 1983). In this letter, however, we will respond only to those assertions that directly concern our method and the reliability of our findings.

In the first paragraph, Gierut asserts that we base our conclusions about management modes on the results of inferential statistical analyses. In fact, the statistical data provide only one source of input for our conclusions. As developed in Shriberg and Kwiatkowski (1982, pp. 249–251): (1) Study A results were presented as group trends over management sessions without inferential statistics (Figure 2), (2) results of a "Drillers" versus "Players" analysis were described without using inferential statistics, and (3) results of a content analysis of student clinicians' anecdotal perspectives on relevant differences among the four management modes were described without using inferential statistics. Gierut's assertion and apparent motivation for a review of standard statistical options in the second paragraph is difficult to reconcile with the following three summative excerpts (p. 251):

> Overall, clinicians' anecdotal impressions were that three factors dictate choice of management mode: (a) a general knowledge of the child's personality, (b) the intended target response, and (c) the stage of management.
>
> Clinicians enjoyed working in whatever mode seemed appropriate for the child, the target behavior, and the stage of management. That is, even Drill was acceptable if the clinician truly felt that it yielded the most effective and efficient learning for a particular child on a target behavior and at a specific stage in management.
>
> Data from the three studies suggest that the structure of a management program for young children with delayed speech is as important as the choice of program content. Keeping in mind the size and scope of these studies, these data refute a position that stimulus response paradigms (Drill) should be selected as the management structure of choice. For preschool children in particular, a certain element of play (Drill Play, Structured Play, or Play) appears to be not only defensible, but in some situations, preferable to Drill. Clear-cut guidelines for the selection of an appropriate management structure for individual children have not emerged from these studies, however. The choice of mode appears to require sound clinician judgment from a person acquainted with the child's personality.

Our clinical and research experience with speech-delayed children since these conclusions continues to support the view that individual differences among children, target behaviors, and management stages comprise the relevant considerations for choice of management mode.

In the third paragraph, Gierut incorrectly infers that we were "quite surprised" at the "number of statistical differences" that occurred in Study B, likening our statistical approach to a "shotgun" procedure that yields significant differences only through "sheer perseverance." Our surprise, as stated, was in response to the finding that "clinicians' independent rankings were remarkably similar" (p. 250). Although we have deliberately embarked on "fishing expeditions" in other aspects of our work, the pairwise comparisons here were straightforward extensions of the basic question, Does any one of the four modes differ from another?

Paragraphs four and five, by our reading and close inspection by our statistical consultant (JRL), include particularly obtuse assertions about "correlated levels of a variable." Confining our response to issues that directly affect our findings, however, Gierut's discussion evidently is based on our use of the term *range*. Her claim that the four modes should be arranged on the underlying continuum of "management structure" conceivably follows from our discussion, but what are the relevant correlates of structure? Multiple, interactive continua could be posited as underlying the salient differences among the modes, including such diverse variables as activity level of the clinician, saliency of knowledge of results, reinforcement schedules, amount of eye contact, and so forth. Each such variable would suggest an arbitrary ordering of the four modes along a different continuum. Because the three studies were completed in real time, we preferred to view modes simply as discrete, operationally defined management conditions.

The sixth paragraph asserts the following logic: If all respondents give the same rank to a level of a variable, there is no variability; no variability is tantamount to a "constant" and, in the presence of a constant, "further infererences from statistical comparisons are irrelevant." Aside from the fact that Gierut is incorrect on the appropriate source-of-variance estimate for nonparametric data, it is the case that ceiling effects pose a validity threat. Ceiling effects occur, however, when an experimenter has failed to construct a measure that has sufficient sensitivity to the upper end of potential performance. The limitation in variability that results is a function of the scale, not, as Gierut suggests, of the individuals using the scale. In any case, Gierut's discussion is unfounded because clinicians did not rate the modes; they rank-ordered them on each of the four questionnaire items.

TABLE 1. Wilcoxon Matched-Pairs Signed-Ranks Tests for effectiveness and efficiency of the four modes. The number of nontied pairs ranged from 8 to 10 for each contrast.

Contrast	$p < .05$	$p < .01$	$p < .005$
Effectiveness			
Drill/Drill Play	*	*	
Drill/Structured Play			
Drill/Play	*		
Drill Play/Structured Play	*	*	*
Drill Play/Play	*	*	*
Structured Play/Play	*	*	
Efficiency			
Drill/Drill Play			
Drill/Structured Play	*	*	*
Drill/Play	*	*	*
Drill Play/Structured Play	*	*	*
Drill Play/Play	*	*	*
Structured Play/Play	*		

Turning to the final series of paragraphs on statistical alternatives, it is important to consider that the studies reported were conducted over a period of 3 years. As described above, only after the results from the three studies were available were we in the position to allow the type of a priori perspectives that would allow for planned orthogonal comparisons. If one were heavily invested in the results of inferential statistics, which we were not, standard planned comparisons would be useful. Our descriptive approach intended to focus on clinically significant findings, those that would pass "eyeball" tests of significance, rather than statistically significant findings that might not replicate.

Based on the published data, Gierut is correctly concerned that some of the pairwise contrasts in Study B may not have reached significance at the .05 alpha level because more than one contrast was performed. However, in contrast to her statement (third from last paragraph) that a planned comparison approach would be "extremely conservative" relative to an alternative "postmortem" or post hoc approach based on the same familywise alpha, for the present data it can be shown that exactly the reverse is true. For the four-treatment problem with $C = 6$ pairwise comparisons, the needed large-sample critical value ($\alpha = .05$, familywise) associated with Gierut's recommended approach is about 2.80, in contrast to a critical value of about 2.64 for the alternative planned approach presented below (JRL, personal communication).

Both the original statistics and those rerun for the present purpose were done on a Hewlett Packard 9810A programmable calculator using the Nonparametric Stat Pac card for the Wilcoxon Matched-Pairs Signed-Ranks Test. This package included a manual with tabled values for Wilcoxon tests excerpted from McCornack (1965). We elected to use the slightly more conserva-

TABLE 2. Wilcoxon Matched-Pairs Signed-Ranks Tests for clinicians' perceptions of the four modes. Each pairwise contrast involved six matched pairs.

Contrast	$p < .05$	$p < .01$	$p < .005$
"Most effective"			
Drill/Drill Play	*	*	*
Drill/Structured Play	*		
Drill/Play	*	*	*
Drill Play/Structured Play	*	*	*
Drill Play/Play	*	*	*
Structured Play/Play	*	*	*
"Most efficient"			
Drill/Drill Play			
Drill/Structured Play	*		
Drill/Play	*	*	*
Drill Play/Structured Play	*	*	*
Drill Play/Play	*	*	*
Structured Play/Play	*	*	*
"Personally prefer"			
Drill/Drill Play	*	*	*
Drill/Structured Play			
Drill/Play			
Drill Play/Structured Play	*		
Drill Play/Play	*		
Structured Play/Play			
"Children prefer"			
Drill/Drill Play	*	*	*
Drill/Structured Play	*	*	*
Drill/Play	*		
Drill Play/Structured Play			
Drill Play/Play			
Structured Play/Play			

tive tabled entries for the Wilcoxon tests in Siegel (1956) in Shriberg and Kwiatkowski (1982). Because Siegel's table for two-tailed tests extends only to the .05 alpha level, however, we used the McCornack *T* values for the present purposes. McCornack's table includes entries that extend only to the .005 alpha level. The choice between using an experimentwise error rate for all comparisons and a familywise error rate for each dependent variable is by no means as straightforward as Gierut asserts. Note that earlier in her discussion of "experiment-wise" error rates (paragraph 3), Gierut actually means familywise error rates, or the error rate appropriate for all contrasts on one dependent variable. In the tables to follow we simply provide the lowest possible alpha levels for pairwise *n*s available in McCornack's tables. The .005 level provided is slightly more stringent than the .008 level appropriate for the smallest familywise comparisons (.05/6).

As shown in Table 1, six pairwise contrasts were significant at both the .05 and .005 alpha levels. Two of the four contrasts that did not reach significance at the .005 alpha level were significant at .01 (and the third at .025). As seen in Table 2, 12 pairwise contrasts were significant at both the .05 and .005 alpha levels. In McCornack's table, the lower sum of ranks must be 0 to reach the .005 level of significance. Moreover, the five contrasts that did not reach significance at .005 were only 1 to 3 *T* units greater than the required 0 ($p < .025$).

Given our clinical, descriptive approach, as emphasized earlier in this letter, the number of significant contrasts affirms that modes are associated with statistically significant differences on relevant dependent variables. Moreover, the pattern of significant findings supports our original claim that alternatives to Drill mode are clinically defensible.

We appreciate the opportunity to provide these data in response to Gierut and we endorse the general sense of her call to consider statistical options in the design of studies in our discipline.

Lawrence D. Shriberg
Joan Kwiatkowski
University of Wisconsin, Madison

REFERENCES

MARASCUILO, L. A., & LEVIN, J. R. (1983). *Multivariate statistics in the social sciences: A researcher's guide.* Monterey, CA: Brooks/Cole.

MCCORNACK, R. L. (1965). Extended tables of the Wilcoxon Matched Pair Signed Rank statistic. *Journal of the American Statistical Association, 60,* 864–871.

SHRIBERG, L. D., & KWIATKOWSKI, J. (1982). Phonological disorders II: A conceptual framework for management. *Journal of Speech and Hearing Disorders, 47,* 242–256.

SIEGEL, S. (1956). *Nonparametric statistics for the behavioral sciences.* New York: McGraw-Hill.

Received January 2, 1984
Accepted June 1, 1984

Corpus Delicti: Response to Gierut

Lawrence D. Shriberg, and Joan Kwiatkowski
J Speech Hear Disord 1984;49;334-336