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Early Intervention Services for Young Boys With Fragile X Syndrome

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This study of boys with fragile X syndrome describes: service delivery; parents' satisfaction with services; early interventionists' perceptions of services needed; and the relationship between service intensity, developmental status, and demographic characteristics. Participants were 50 boys with fragile X syndrome, their parents, and teachers. Early intervention started on average at 21.6 months. There was a steady increase in the amount of early intervention across age periods. The intensity of speech-language and occupational therapies, however, remained constant. By the age of 60 months, the number of children receiving physical therapy and the intensity of physical therapy both decreased. Although parents reported satisfaction with services, most said they would have preferred more services. Early interventionists and teachers seemed more concerned about behavior than cognitive delays. No statistically significant patterns emerged regarding the relationship between developmental status, service intensity, and demographic characteristics.

Fundamental to the Individuals with Disabilities Education Act (IDEA) is the assumption that services for children with disabilities and their families should be individualized. Two primary factors drive this individualization: parent preferences and characteristics of the child receiving services. Much has been written about the importance of attending to parent preferences. The legislation requires that families must be able to participate as part of the intervention planning team, and that the plan reflects their goals and priorities. These requirements are consistent with the philosophy that families are consumers of a public service and should have choices about the nature and amount of services received.

With regard to characteristics of the child, individualization is emphasized in the requirements for conducting individualized assessments to determine needs and styles of learning and for writing individualized service plans. In designing individualized services,

early childhood special educators and other early intervention professionals must consider a wide range of child factors and work with families to establish goals that are developmentally appropriate and functional within the child's culture, home, and school.

In addition to the individualized assessments of children, practitioners also attend to the particular disorder that causes the disability, when that disorder is known. Known characteristics of disorders such as autism, Down syndrome, cerebral palsy, blindness, or deafness provide important information about learning styles and characteristics that can influence the provision of individualized services.

Beginning in 1991 and continuing throughout this decade, Hodapp and Dykens have challenged the field to consider the importance of etiology in helping families and practitioners determine appropriate treatments that promote optimal development (Dykens, 1995;

Hodapp, 1997; Hodapp & Dykens, 1991; 1994). For early intervention professionals, knowledge about both etiology and etiology-specific treatments can be a challenge for several reasons. With the advent of the Human Genome Project and other biomedical advances, the number of known causes of disabilities is increasing at a rapid rate. Many of these disabilities are rare, however, and often there is little if any research documenting specific treatments for children with that disorder. Disabilities such as Down syndrome, autism, or deafness have a long history of clinical interventions, in both their studied forms and in practice, and much is known about the characteristics of these disorders that can be synthesized and disseminated to practicing professionals (Calderon & Greenberg, 1997; Dawson & Osterling, 1997; Spiker & Hopmann, 1997). More recently discovered syndromes have typically received less attention in the research and clinical literature, and the focus in early investigations is usually on characterizing the disorder before the efficacy of various intervention models can be tested. For rare disorders, there might never be an adequate body of intervention research.

As new disorders are discovered, researchers and practitioners need to work together to determine the unique learning styles and characteristics of children who share that disorder. They also must characterize and understand the variability within the disorder to prevent the assertion of assumptions that turn out not to be true for a substantial number of children. Research is also needed to describe the services typically received by children with the disorder, and parent and professional perceptions of the appropriateness of those and other treatments. This paper provides initial information related to the services received and perceptions of services needed for young children with fragile X syndrome.

Fragile X syndrome is the leading known hereditary cause of mental retardation and results from a mutation on the long distal arm of the X chromosome. A DNA expansion of CGG (cytosine-guanine-guanine) repeats to 200 or more (normal genes typically have around 35 of these repeats) usually results in

decreased production of the FMR protein, which is believed to be necessary for normal brain development. After the identification of the FMR (fragile X mental retardation) gene in 1991, it became easier to diagnose fragile X syndrome through DNA testing. Although both boys and girls can be affected by fragile X syndrome, boys tend to be more severely affected because they have only one X chromosome.

Considerable variability in the early developmental trajectories of boys with fragile X syndrome is evident; however, most function in the mild to moderate range of mental retardation (Bailey, Hatton, & Skinner, 1998). Although developmental delay is a hallmark of fragile X syndrome, teachers' and parents' concerns often center around behavioral and communication issues (Hatton & Bailey, 1997). A series of studies by Bailey and colleagues (Bailey, Mesibov, et al., 1998; Bailey, Hatton, Mesibov, & Ament, 2000; Bailey, Hatton, Mesibov, & Ament, in press) reported that approximately 25% of a sample of young boys with fragile X syndrome exhibited autistic behaviors, which is consistent with findings of Cohen (1995) and Turk and Graham (1997). Boys with both fragile X syndrome and autism appear to have poorer developmental and adaptive outcome and more problem behaviors (Bailey et al., 2000; Cohen, 1995; Hatton & Bailey, in press).

Unlike previous studies of temperament in children with disabilities, Hatton, Bailey, Hargett, Skinner, and Clark (1999) found that boys with fragile X syndrome differed from the reference sample of the Behavioral Style Questionnaire (McDevitt & Carey, 1978) on five dimensions of temperament. Specifically, boys with fragile X syndrome were more active and less approachable, less adaptable, and less persistent. A recent study of sensory processing in boys with fragile X syndrome found that 40–50% of the variance in problem behaviors was related to sensory processing (Baranek, Hooper, Hatton, & Bailey, 2000).

Although the clinical literature in fragile X syndrome often lists recommendations for intervention and services, to date, there has only been one preliminary research report on this

subject (Hatton & Bailey, 1997). A recent study by York, von Fraunhofer, Turk, and Sedgwick (1999) found that special educators knew much less about fragile X syndrome than about autism or Down syndrome, and probably had inadequate knowledge to assure an appropriate program of special education services. These results supported those obtained in earlier studies by Wilson and Mazzocco (1993) and Madison, Mosher, and George (1986). Thus, additional knowledge about services and learning characteristics of children with fragile X syndrome is needed.

Studying services for young children with disabilities poses a unique set of challenges to researchers. The highly individualized nature of early intervention and variability in programs and resources preclude simple descriptions of services and their efficacy (Bailey, Aytch, Odom, Symons, & Wolery, 1999; Guralnick, 1997; Harbin, McWilliam, & Gallagher, 2000; Kochanek & Buka, 1998; Shonkoff, Hauser-Cram, Krauss, & Upshur, 1992). The limited research that has been conducted suggests that multiple factors, such as child characteristics, family preferences, local resources, and state and local models, all interact to determine the amount and nature of services provided (Harbin et al., 2000; Kochanek & Buka, 1998).

Regarding intensity of services, Harbin and colleagues (1998) found that infants and toddlers received an average of 1.7 hours of services per week, whereas preschoolers received approximately 14 hours of services per week. Preschoolers served in noninclusive settings received approximately 18 hours of specialized services per week, compared to children in specialized segregated programs who received an average of 11 hours of specialized services per week (Harbin et al., 1998). Notably, Kochanek and Buka (1998) found that children from families in which the mother had a higher level of education received statistically significantly more intervention services. They also found that older children and children who had been enrolled in agencies for the longest period of time received statistically significantly more services. Researchers examining parental satisfaction with ser-

vices have reported that the majority of parents are satisfied with services. Most parents, however, would have preferred more services than they were receiving (Bailey et al., 1999; Harbin et al., 2000).

In examining early intervention, researchers have typically examined the relationship between developmental outcome or gain and intensity of services, age at which services were initiated, or parental satisfaction with services. Little attention has been focused on the perceptions of early interventionists regarding the strengths and needs of young children with disabilities or their recommendations for providing services. A more holistic description of early intervention would be achieved by examining service delivery over time and including perceptions of both parents and interventionists; one of the goals of this study. In the process, we hoped to gain insight into the issue of etiology-specific intervention. Literature on genetic syndromes often describes unique behaviors and characteristics. Whether or not parents and professionals perceive those characteristics as challenges in everyday life presents an interesting question in itself. If they do, recommendations for facilitating optimal development and enhancing intervention that would be useful to the field of early childhood special education, to clinicians in health-related fields, and to the families of these children could emerge.

As a first step in describing intervention services for young boys with fragile X syndrome, we used a longitudinal design, beginning in 1994, to collect both prospective and retrospective data to document services that children received during the first six years of life, parental satisfaction with services, and the perceptions of early interventionists who served these children. The purposes of this study are to a) describe the early intervention service delivery patterns for young boys with fragile X syndrome during the infant-toddler and preschool years; b) describe parents' satisfaction with intervention services and transitions; c) describe early interventionists' perceptions of the strengths and needs of young boys with fragile X syndrome, as well as their recommendations for serving these children;

Table 1.
Demographic Information of Boys With Fragile X Syndrome

Participants	
N	50
Mean Age at Entry (Months)	39.2
Mean Current Age (Months)	84.4
Ethnicity	
European American	42
African-American	6
Hispanic	1
Asian	1
Public Assistance	
Yes	20 (40%)
No	30 (60%)
State of Residence	
NC	21
VA	23
Other	6

and d) examine the relationship between intensity of services, developmental status, and demographic characteristics of the families in this study. The collection and analyses of descriptive data are the first step in the systematic study of the intervention needs of boys with fragile X syndrome, with the ultimate goal of providing recommendations that will enhance the provision of services to this group of children.

METHOD

Participants

The participants were 50 boys with fragile X syndrome and their parents and teachers in four states in the mid-Atlantic region of the United States. All children had been diagnosed with full mutation fragile X syndrome, using molecular tests prior to enrollment in the study. The average age of the children enrolled in the study was 39.2 months, with a range from 12 to 60 months. The mean age of diagnosis of fragile X syndrome for this sample was 27 months, with a range from birth (prenatal diagnosis) to 55 months. Recruitment into the study, which is ongoing, began in 1994. For this particular study, we included data regarding early intervention services starting at birth and data regarding chil-

Table 2.
Means and Standard Deviations for Developmental Status Across Age Intervals Using the Battelle Developmental Inventory, N = 50, Observations = 239

Chronological Age	Number of Assessments	Develop- mental Age, Mean in Months	Standard Deviation
12 months	2	9.5	7.1
18 months	5	14.5	2.1
24 months	10	17.0	3.7
30 months	17	18.1	3.9
36 months	27	19.9	4.0
48 months	29	25.3	5.0
60 months	41	30.5	7.3
72 months	30	36.2	9.9

dren's development collected from age of entry through age 6 years or kindergarten.

Children were recruited through genetics clinics, developmental evaluation centers, and intervention programs. Informed consent for participation was obtained from the parents or guardians of all participants. For each assessment period in which the child and family participated, families received a \$25 stipend and a brief summary of the developmental assessment and behavioral observations made by project evaluators. Demographic characteristics of the children are described in Table 1, and developmental status across time is shown in Table 2.

Parental participants were 45 mothers with the same ethnic background as their children. The mean age of the mothers who participated in the study was 35.5 years, with a range from 23 to 48 years. Only 1 mother did not have a high school degree. Although only 9 mothers (20%) held college degrees, 21 mothers (47%) reported some college education. The early interventionists who participated are described in the results section.

Procedures

Services. Children were seen once or twice yearly as participants in a larger longitudinal study investigating the development and educational needs of young boys with fragile X

syndrome. During each visit, parents were interviewed regarding the nature and intensity of services received. After conferring with the child's primary interventionist to confirm that these reported services were indeed being provided, data were entered into an Excel database by the child's age. In addition, records were requested from the agencies serving these children and then compared for accuracy with existing data. Discrepancies were clarified through phone interviews with parents and teachers.

Parents' perceptions. When children transitioned from infant-toddler into preschool programs and again when they transitioned into kindergarten, parents participated in a separate interview regarding their impressions of the extent to which services during either the intervention or preschool years met their expectations for content, intensity, and quality. This information was entered into an Excel database by transition time—either preschool or kindergarten. Data were coded, categorized, and summarized descriptively.

Early interventionists' perceptions. Surveys and interviews were used to collect detailed information regarding early interventionists' impressions of the strengths and needs of young boys with fragile X syndrome. A three-page survey was sent to each early interventionist serving children enrolled in this study during the 1996–1997 school year. Respondents were given a payment of \$15 as an incentive for completing and returning the survey.

Both surveys and interviews included open-ended questions to elicit unbiased responses from participants. Upon receipt of the survey, information was coded, checked for reliability, entered into Excel databases, and summarized descriptively. Several steps were used in the coding of the open-ended responses. Word-for-word responses were first recorded and then categories of responses were derived from this analysis. For example, "likes to be hugged," "affectionate," and "friendly" were collapsed into a sociable category. Two raters coded all responses and discrepancies were resolved by consensus. Results from the survey

were used as the basis for the interview protocol that was subsequently developed.

Project staff conducted phone interviews with the interventionists who worked with the children enrolled in this study during the 1997–1998 school year. Following the interview, data were coded, entered into an Excel database, and summarized descriptively.

Developmental status. The Battelle Developmental Inventory (BDI; Newborg, Stock, Wnek, Guidubaldi, & Svinicki, 1984) was used to measure severity of developmental disability. Because the BDI covers the age range from birth through 96 months, it provides a consistent measure of development during the infancy, preschool, and early elementary school years. The BDI yields an overall developmental age or developmental quotient in addition to scores for the subdomains of personal-social, adaptive, motor, communication, and cognitive development. It is particularly useful for monitoring growth over time. The BDI has a solid normative base with well-documented reliability and validity, drawn on a nationally representative sample of children. Additionally, the BDI has adaptations for children with disabilities, making it particularly well suited for longitudinal studies of development with those samples (Bailey, Hatton, et al., 1998; Bailey, Mesibov, et al., 1998; Hatton, Bailey, Burchinal, & Ferrell, 1997). Several independent studies have documented a high correlation between BDI scores and scores on measures of cognitive, adaptive, language, and social functioning with populations of both normally developing children and children with disabilities (Boyd, 1989; McClean, McCormick, Bruder, & Burdug, 1987; Sexton, McClean, Boyd, Thompson, & McCormick, 1988; Snyder, Lawson, Thompson, Strickland, & Sexton, 1993). The BDI was administered either once or twice yearly. A total of 239 BDI observations were completed for the 50 children enrolled in this study, with a mean of 4.8 observations and a range of 2 to 10 observations per child.

To determine if developmental status was related to intensity of services or to demographic characteristics, we calculated a devel-

opmental quotient for each child at each of the age periods of interest by dividing developmental age by chronological age and multiplying that number by 100. Developmental quotients were examined across time for stability for each child. In examining the relationship between developmental status and service intensity, we focused on age categories of 36 months and above, because developmental quotients obtained at ages under 30 months did not appear as stable as those at ages of 30 months and above did.

Hierarchical linear modeling (Bryk & Raudenbush, 1987; Burchinal & Appelbaum, 1991) was used to examine trends in intensity of services across time. This approach, sometimes referred to as mixed model analysis of variance, accounts for the dependence of observations across time through the estimation of random effects. These models can be conceptualized in two parts (Bryk & Raudenbush, 1987; Burchinal & Appelbaum, 1991). First, the random or within subjects effects of age can be modeled as a polynomial, which describes the intercept and slope of the trajectory of interest for each child and for the entire sample. In the second stage, the fixed effects are estimated, which explain differences in the shape of the original trajectory for different children. In this way, differences between the individual trajectories of the children can be explained by individual characteristics such as autistic behavior or family resources, or by group characteristics such as state of residence or type of intervention. Models of this type have been found to be robust under conditions of highly reliable measures regardless of the degree of individual differences across time (Burchinal, Bailey, & Snyder, 1994). Advantages of this approach include simultaneous estimation of the individual and population trajectories given that the individual curves are systematically distributed about the population curve. This approach can be used even when individuals have randomly missing data or when time-varying covariates are of interest.

In this analysis, the dependent variable was hours of services, and random effects were estimated for the intercept (mean level) and

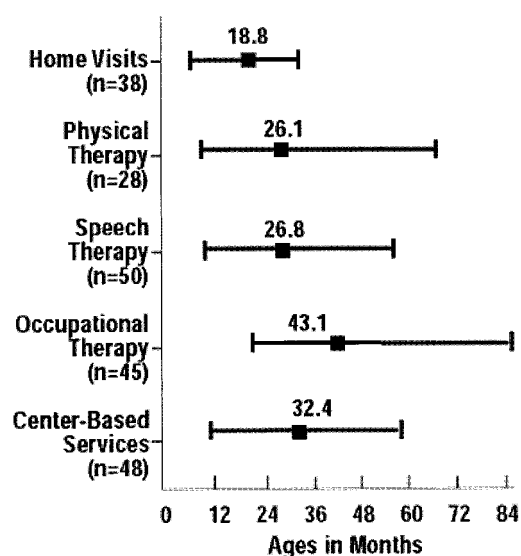


Figure 1. Age at which services were initiated. Mean age denoted by square with range in brackets.

slope (six categories of age at assessment). Fixed effects were estimated for age, mother's education level, state of residence, and developmental quotient.

RESULTS

Early Intervention Services

The average age at which boys in this sample started receiving services was 21.6 months (range 6 to 51 months), approximately 3 weeks after they had been diagnosed as being developmentally delayed ($M = 20.7$ months, range 7 to 55 months). Most children actually started receiving early intervention an average of 6 months prior to the mean age of diagnosis of fragile X syndrome. By the age of 4.5 years, all of the boys were receiving early intervention services. Children under the age of 3 years typically received early intervention at home. Most children received other services as well. Figure 1 illustrates the average age, as well as the range, at which each type of service started for the boys in our sample.

Home visits started on average at 18.8 months whereas center-based services started at approximately 32.4 months. Physical and

Table 3.
Mean and Standard Deviations of Hours of Services Received Per Month at Each Age Interval

	12 to 23 Months	24 to 35 Months	36 to 47 Months	48 to 59 Months	60 to 71 Months
Early intervention or special education	9.9 (12.1) <i>n</i> = 26	30.2 (34.6) <i>n</i> = 42	69.9 (41.7) <i>n</i> = 42	78.8 (39.2) <i>n</i> = 45	94.7 (40.5) <i>n</i> = 39
Speech-language therapy	3.9 (1.5) <i>n</i> = 22	4.4 (2.9) <i>n</i> = 37	4.5 (2.7) <i>n</i> = 44	4.9 (3.4) <i>n</i> = 45	4.9 (2.7) <i>n</i> = 38
Occupational therapy	3.9 (1.9) <i>n</i> = 15	2.8 (1.9) <i>n</i> = 28	2.9 (1.7) <i>n</i> = 30	2.10 (1.6) <i>n</i> = 31	2.9 (1.5) <i>n</i> = 29
Physical therapy	3.4 (2.0) <i>n</i> = 12	2.5 (1.7) <i>n</i> = 18	2.3 (1.3) <i>n</i> = 17	2.3 (1.3) <i>n</i> = 20	1.3 (1.3) <i>n</i> = 12

Note. Numbers in parentheses are standard deviations.

speech-language therapy started, on average, at 26 months, whereas the mean age for the initiation of occupational therapy was 43 months.

Intensity of services is displayed in Table 3. As can be seen, the amount of early intervention increased dramatically at 24 to 35 months and again at 36 to 47 months, whereas the amount of therapies remained relatively stable across time. This information is also shown graphically in Figure 2. The stability of service hours across therapies contrasted sharply with the increase in early intervention across time. Statistical analysis using hierarchical linear modeling confirmed the increase in intensity (average hours per month across age categories) of early intervention as children grew older, $F = 109.34$, $df = 1$, $p < .001$.

Parent Interviews Regarding Transitions

Part H/C to Part B. Of the 24 parents who were interviewed during their child's transition from Part H/C to Part B services, 22 (92%) answered "yes" when asked if they were satisfied with the early intervention services they received. When asked if they would have changed anything about the services they received, however, 15 (63%) responded "yes." The most frequent change reported was an increase in therapies, noted by 7 parents. Only a third of the sample had comments about their child's transition from Part H/C services to Part B services. Of these 8 parents,

4 had positive experiences and 4 had stressful experiences. Positive comments included feeling involved and informed, whereas negative comments noted a lack of communication and fragmentation in the transition process, along with slowness and inconsistency of the process.

Transition to kindergarten. Of the 50 children enrolled in this study, 43 transitioned into kindergarten. Thirty-six parents (84%) reported satisfaction with preschool services, 3 (8%) reported dissatisfaction, and 3 (8%) noted that they were somewhat dissatisfied with services. When asked whether or not they would have changed anything about their child's services, 20 (47%) said "no" and 23 (53%) said "yes." When asked specifically about the changes they would make, the following responses were made: more services [usually more therapies] (9 or 21% of parents), different placements (6 or 14% of parents), different logistics [e.g., morning versus afternoon class, mode of therapy delivery] (3 or 7% of parents), and idiosyncratic changes (5 or 12% of parents). Regarding transition into kindergarten, 17 parents (40%) reported positive experiences, while 10 (25%) reported negative or somewhat negative experiences. The remaining parents seemed to have a neutral opinion of transition—that it was "all right", not particularly positive or negative.

Determining eligibility labels and class placement seemed to elicit anxiety on the part of some parents (7 or 16%), and those issues usually were related to negative experiences.

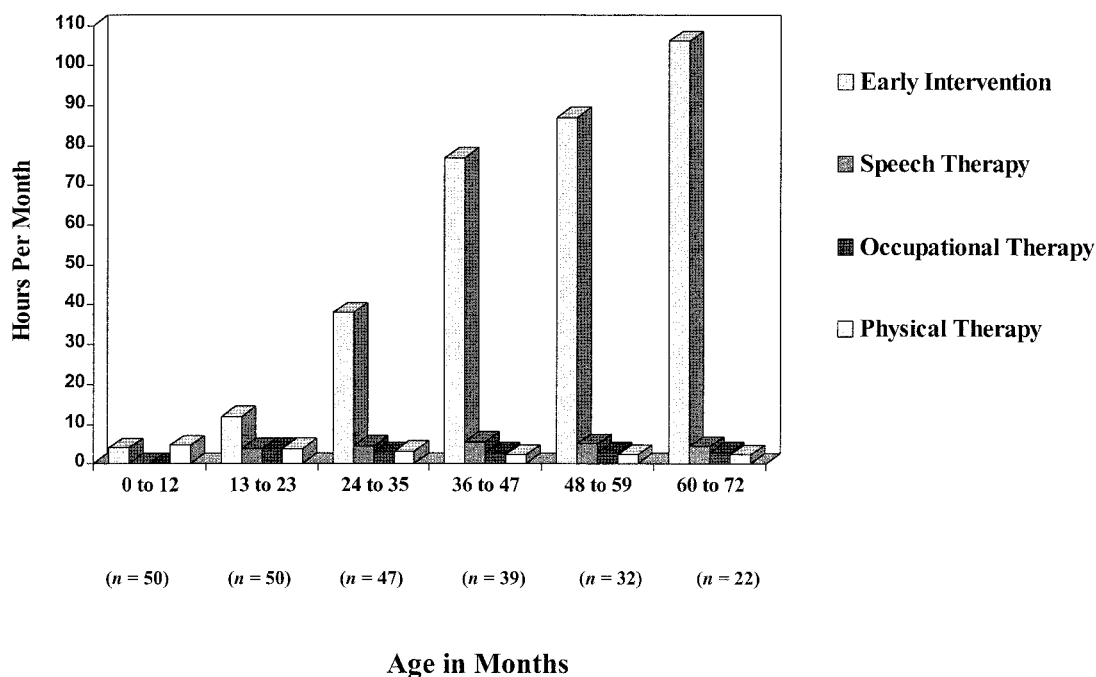


Figure 2.
Mean number of hours of services per month by age group.

For example, many parents had identified eligibility labels associated with access to more services in their school systems. Whether or not school personnel would agree with the label seemed to elicit anxiety before those meetings. In some cases (7 or 16%), this anxiety was justified, and parents had difficulty in securing the label they thought most appropriate. In other cases (28 or 65%), all parties concurred on both eligibility label and class placement.

Regarding the process of developing an Individualized Education Plan (IEP), 28 parents (65%) reported a positive experience in which their input was valued. Of the 7 parents (16%) who reported a negative experience, most reported that the IEP had been written before the meeting and that their input was not valued.

Early Interventionist Surveys

In an attempt to learn more about early interventionists' perceptions of boys with fragile X syndrome, we surveyed teachers and child

development specialists of the preschool boys enrolled in our study. Thirty-three surveys were returned (response rate of 80%). Most of the respondents (30 or 90%) provided intervention in self-contained classes for preschoolers with special needs, whereas two respondents taught in regular child care centers, and one provided intervention in the home. The majority of the respondents (49%) had a bachelor's degree, 38% had a master's degree, and 10% had an associate's degree. The mean number of years of experience for the group was 11.3 years with a range from 2 to 25 years. Only 28% of the respondents had previous experience working with children with fragile X syndrome.

Strengths. When asked, via open-ended questions, to describe their initial impressions and the strengths of young boys with fragile X syndrome, 33% of the early interventionists noted that the boys were sociable and enthusiastic with relatively good receptive and non-verbal communication skills, a good sense of

humor, good visual imitation skills, and a good response to structure and routine.

Concerns. In describing their concerns about boys with fragile X syndrome, again via open-ended questions, early interventionists noted the following: inability to focus on tasks or distractibility, short attention span (50% of respondents); hyperactivity or extreme activity (25%); high anxiety level (25%); hypervigilant or easily overstimulated (25%); aggression related to frustration or anxiety (18%); impulsivity (10%); and noncompliant behavior (6%).

Instructional strategies and suggestions. When asked what advice they would give another early interventionist who is about to work with a child with fragile X syndrome, the two most frequently mentioned suggestions were to develop a consistent routine with structured activities in a structured environment (54% of respondents) and to read as much as possible about children with fragile X syndrome (23% of respondents). Other suggestions are shown in Table 4.

Early interventionists were given a list of dimensions of classroom planning for young children and asked to rank the level of modification needed for their student with fragile X syndrome. Results are presented in Table 5 and suggest that major modifications were needed in adapting teaching strategies, dealing with inappropriate behaviors, group activities, and in reinforcing appropriate behavior to meet the needs of boys with fragile X syndrome. Few teachers felt that outdoor play, snack, or meal times had to be modified.

Environmental adaptations. The following adaptations were employed by the 17 early interventionists (52%) who reported using environmental modifications: using techniques that provide structure, positioning the child away from peers to avoid overstimulation and distraction, limiting distractions—particularly loud noises and excessive visual stimuli (e.g., covering computer when not in use), providing consistency and routine, providing an area for the child to go to regroup, using music to calm the child, using an adaptive chair with a tray to help contain the child and allow him to focus on activities, and using a picture

schedule and objects to help the child transition from one activity to another.

Learning styles. Approximately 22 of the respondents (67%) considered boys with fragile X syndrome primarily visual learners who require hands-on, structured activities. Eight respondents mentioned the need for tactile and kinesthetic experiences, lots of repetition, one-on-one instruction, and consistency. Verbal praise seemed the most effective type of reinforcement used by teachers, but food, stickers, hugs, and special activities also seemed to work with some boys. Many of the boys were described as enjoying music, outdoor playground activities, and books. They reportedly disliked fine-motor activities and seatwork.

When asked if boys with fragile X syndrome seemed to be more easily stimulated by sensory information than other children were, 21 of the 33 early interventionists (64%) said “yes.” Tactile defensiveness and sensitivity to loud sounds and noisy environments seemed to be the greatest concerns, followed by sensitivity to visual stimulation (children closed eyes when they seemed overwhelmed). Also mentioned under hyper- or hypo-sensitivity were difficulty with transitions and oral motor difficulties (stuffing mouth with too much food).

Specialized needs. Twenty-one of the 33 respondents (64%) felt that boys with fragile X syndrome had specialized needs different from those of other children with disabilities whom they had worked with in the past. Differences noted by teachers were: need for behavior management plan, lack of consistent response to reinforcers, aggression, high anxiety level, hypervigilant behavior, easily overstimulated, need for constant redirection, high distractibility, more intense than other children, impulsivity, tactile defensiveness, sensory integration problems, and poor communication skills. Other characteristics included below-average self-control and ability to attend. When asked to describe factors that might interfere with placement in an inclusive setting, many teachers mentioned these same factors. A majority of the teachers felt that boys with fragile X syndrome had average or

Table 4.
Recommendations of Early Interventionists Working With Boys With Fragile X Syndrome

Strategies and Suggestions	% ^a	S ^b	I ^c
Behavior management	54–58%	X	X
Have a behavior management plan in place that addresses aggression toward self or others		X	X
Use consistent behavior management			X
Provide guidance in controlling impulsivity		X	
Use consistent reinforcers		X	
Use stern voice or affect to convey displeasure; be firm			X
Don't let frustration escalate; allow break for regrouping and calming			X
Be patient, persistent, and loving		X	X
Read child's cues			X
Motivators	50–60%	X	X
Use hands-on activities involving a variety of senses		X	
Alternate quiet and active activities—be flexible in demands for sitting still, use frequent movement breaks		X	X
Use music and movement to motivate and teach concepts		X	
Use favorite toys, activities, praise, and music as motivators and to ease transitions			X
Let the child hold things			X
Build on the strengths and interests of the individual child		X	
Environmental modification	50–60%	X	X
Have a consistent routine with structured activities and a structured environment		X	X
Provide a personal work space and an area for the child to go to regroup		X	
Reduce distractions			X
Provide small teacher-student ratio or one-on-one instruction		X	X
Be flexible and give the child choices		X	
Use visual cues, modeling, picture schedules			X
Instructional strategies	60–70%	X	X
Work up to desired skill through successive approximations		X	
Consider sensory integration and occupational therapy needs; use sensory diet, work with occupational therapist		X	X
Consider similarities to autism and incorporate appropriate strategies			X
Anticipate and schedule for needs, prepare for transitions and new experiences			X
Break tasks down into manageable steps			X
Don't ask for or require direct responses			X
Work on attention, basics			X
Focus on functional and social skills			X
Teach the child how to interact and play			X
General	5–10%	X	X
Learn as much as possible about children with fragile X syndrome		X	X
Look at the unique characteristics of each child—great variability in boys with fragile X		X	X
Work with families		X	
Respect the need for medication			X

^a Proportion of respondents who identified strategies or suggestions in these categories.

^b Survey response.

^c Interview response.

Table 5.*Percentage of Teachers' Need for Modification in Dimensions of Classroom Planning*

Dimension	Level of Modification			
	Substantial	Moderate	Minimal	None
Dealing with inappropriate behavior	44	35	18	3
Reinforcing appropriate behavior	30	42	17	11
Teaching strategies	41	44	6	9
Transition strategies	27	32	35	6
Center activities	27	38	26	9
Group activities	24	44	20	12
Schedule of activities	20	20	30	30
Physical activities	15	9	32	44
Meals or snacks	6	9	44	41
Outdoor play	6	9	44	41

above average computer and gross motor skills.

Inclusion. Of the 18 early interventionists who responded to this item, 10 (56%) thought it would be difficult for their children with fragile X syndrome to function in an inclusive setting whereas 8 (44%) thought their children could possibly function in such a setting with adequate supports. Behaviors noted as inhibiting successful inclusion were: hyperactivity, anxiety, tendency to become overly stimulated, distractibility, tantrums, lack of interest in peers, poor communication skills, noncompliant behavior, need for one-on-one instruction, aggression, and immaturity. Early interventionists thought that the following strategies would enhance the children's ability to function in an inclusive class: consistent reinforcers, adequate support staff, small class size, provision of one-on-one instruction, a regular teacher who facilitates interaction between children, provision of an area for regrouping, and use of routine. They noted that the sociability of boys with fragile X syndrome and their visual imitation skills would serve them well in an inclusive classroom. A few respondents noted the value of peer models and appropriate language models available in an inclusive class.

Early Interventionist Interviews

Thirty-five teachers and child development specialists were interviewed regarding their perceptions of young boys with fragile X syn-

drome. Most of the respondents (81%) provided intervention in self-contained classes for preschoolers with special needs, whereas 3 respondents taught in regular child care centers, and 1 provided intervention in the home. The majority of the respondents (65%) had a master's degree, whereas 25% had a bachelor's degree, and 10% had an associate's degree. The mean number of years of experience for the group was 8.7 years, with a range from 2 to 22 years. Only 29% of the respondents had previous experience working with children with fragile X syndrome.

When asked to identify strengths of the children with fragile X syndrome whom they served, 15 of the 35 professionals noted sociability and enjoyment of interactions with others, whereas helpfulness (6 responses) and affection (5 responses) were also noted. Twenty-three (66%) of the respondents who were interviewed identified behavior as the area of greatest concern. Other concerns were developmental issues (12 or 20%), family issues (5 or 9%), and other individual issues (3 or 5%).

The major instructional or intervention goals described by early interventionists for children with fragile X syndrome are shown in Table 6. Primary goals addressed behavior, language development, social development, self-help skills, or academic readiness.

Because many children with fragile X syndrome are described as being hyperaroused (Cohen, 1995; Roberts, 1998), early interventionists were asked whether or not they ob-

Table 6.
Primary Instructional Goals for Children With Fragile X Syndrome

Domain	Specific Goals
Behavior	Increase attention
	Increase compliance
	Reduce stereotypic behaviors, normalize behavior
	Reduce sensory defensiveness
	Increase ability to wait for turn
Social	Increase interactions with other children
	Increase inclusion
	Improve social skills
	Increase pretend play
Language	Use words instead of aggression
	Increase expressive language
	Answer questions appropriately
	Follow three-step directions

served atypical sensory responses in the children with whom they worked. Approximately 89% of the early interventionists responded affirmatively. Approximate percentages of respondents noting hypersensitivity to particular sensory stimuli included: auditory, 42%; tactile, 23%; oral, 16%; visual, 13%; and other, 6%. Finally, early interventionists were asked to provide recommendations for other professionals who might serve young children with fragile X syndrome. Their responses are summarized in Table 4.

Developmental status. In Figure 3, the mean developmental ages of the boys in this study are plotted across time. Table 2 provides additional information, including standard deviations. A total of 239 observations were completed for the 50 subjects in this study. Not all boys had assessments at all ages because they entered the study at varying ages. These data served as the basis for calculating developmental quotient that was used as a

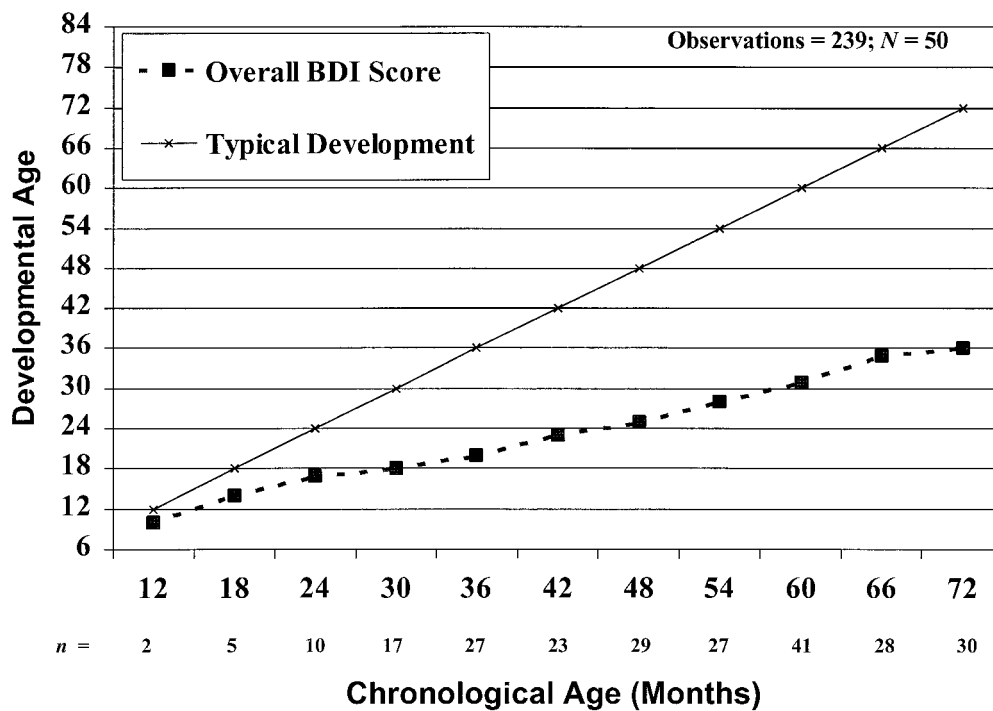


Figure 3.
Mean Battelle developmental age scores for boys with fragile X syndrome as compared to typically developing children.

fixed effects variable for the hierarchical linear modeling analysis. As described in the method section, developmental quotients were calculated for each child at each assessment and compared across time for stability.

The results of hierarchical linear modeling revealed that intensity of early intervention increased as children grew older. Developmental status was related to intensity of services for some children. Specifically, children with higher developmental quotients received less early intervention, $F = 4.23$, $df = 167$, $p < .05$.

Discussion

This study of early intervention services for young boys with fragile X syndrome paints a beginning picture of how the field perceives and is responding to the needs of these children. The use of both qualitative and quantitative research methodologies in the design of surveys and interviews enriched our findings and enabled parents and teachers to share information that might not have been elicited otherwise. Combined with quantitative descriptions of services across time and longitudinal developmental data, these data represent a description of services provided to a sample of young boys with fragile X syndrome during the mid 1990s.

We found considerable variation in the service patterns and amount of services provided. The developmental status of the children in the study also showed tremendous variability, as demonstrated in Table 2. Although early interventionists consistently described behavior as their primary concern, they offered a variety of strategies to deal with that behavior (Table 4). Thus, we conclude that early interventionists and parents should consider this variability and note that a specific set of recommendations for intervention must be based on individual needs and circumstances.

Because this study was planned as a descriptive study, limitations should be considered. Interview and survey data were collected at one point in time and were analyzed qualitatively. Thus, results might not be generalizable, and quantitative researchers might question the reliability of these data.

Services

For this sample of boys with fragile X syndrome, the average age of entry into early intervention services was 21.6 months. For most children, home visits started first, followed by the addition of physical and speech-language therapy. Occupational therapy began, on average, at 43 months. The amount of early intervention increased over time. The intensity of speech-language and occupational therapy, however, remained constant over time. Physical therapy decreased at the 5-year age level in both intensity and the number of children receiving physical therapy. Two factors probably influenced this pattern of services. First, as children acquired motor milestones, there was less need, or a perception of less need, for these services. Second, an increase in occupational therapy coincided with the decrease in physical therapy. Limited resources and overlap between physical and occupational therapies at young ages probably precluded the provision of both therapies for many children.

Considering early intervention alone, the children in this sample appeared to receive more services than would have been expected from previous research (Harbin et al., 2000; Kochanek & Buka, 1998; Shonkoff et al., 1992). During the 12- to 23-month age range, the children received an average of 10 hours of services per month, or 2.5 hours per week. This increased to 30 hours per month at the 24- to 35-month age range, and again to 70 hours at the 36- to 47-month age range.

Parents' Perceptions of Services

As has been found in other studies of parents' perceptions of services, the parents of children in this study were generally satisfied with the services their children received. Increased amounts of speech-language and occupational therapy and improved communication with teachers and therapists were listed as desirable improvements in the services. These results are consistent with those described by Harbin et al. (1998) and McWilliam, Tocci, Harbin, and Siders (1997).

Regarding transition from preschool into kindergarten, negative experiences reported

by parents were related to dissatisfaction with eligibility labels and class placements. Positive experiences were related to feeling included and informed regarding their children's needs. Most parents felt that the IEP process was positive. The few who did not feel that way noted that the IEP was written before the meeting and that their input was neither sought nor valued.

Early Interventionists' Perceptions

The early interventionists of boys with fragile X syndrome who were surveyed and interviewed appeared more concerned about the behavioral characteristics of the boys than about their developmental or cognitive needs. Although many expressed concerns about expressive language skills, the majority of responses dealt with behavioral or sensory characteristics and the accommodations required for the children to function within the classroom. The need for a structured routine and classroom environment and a consistent behavior management program were mentioned repeatedly in both surveys and interviews, as was the need for environmental adaptations. In addition, the goals developed for the children with whom they were working reflected those same concerns.

The information provided by the early interventionists in this study was consistent with the quantitative data we collected on temperament, autistic behavior, sensory processing, and developmental outcome of the children in our study and with qualitative data describing parental perceptions. Nonetheless, we see this information as a first step in describing the perceptions of these individuals. Our findings need to be replicated by others, but they should also prove useful in generating hypotheses for more rigorous quantitative studies.

Developmental Status and Services

In comparing developmental status to demographic variables, no consistent patterns emerged. This finding is consistent with the findings of Harbin et al. (2000). Intensity of early intervention was related to developmental status. Children with the highest develop-

mental quotients received less intense early intervention. These children might not have needed intense early intervention because they were not as delayed as the group as a whole. The small number of children with relatively high developmental quotients in this sample might, however, limit the generalizability of this finding. Another cautionary note is the use of developmental quotient scores as markers for developmental status. Because the Battelle Developmental Inventory does not provide standard scores below 65, we used developmental quotients. This measure of developmental status might not be sufficiently sensitive to detect associations that might be present.

Etiology-Specific Needs

The information in Table 4, provided by early interventionists, serves as a summary of recommendations from this study. Because developmental delay is a hallmark feature of fragile X syndrome, we expected a number of recommendations for enhancing cognitive development. This is, however, not the case. Instead, a number of strategies that might be similar to recommendations for children with behavioral or emotional disorders emerged: having a behavior management plan that addresses aggression to self or others, consistency in behavior management, controlling impulsivity, using consistent reinforcers, preventing escalation of frustration, and providing a consistent routine and structured environment. Some of these suggestions are also appropriate for children with attention deficit hyperactivity disorder, as are these additional recommendations: flexibility in demands for sitting still, providing a personal work space, reducing distractions, facilitating attention to tasks, and respecting the need for medication. Again, features from the attention deficit hyperactivity disorder list also apply to children with autism or sensory dysfunction, as do these additional items: preparing for transitions, using visual cues and picture schedules, including sensory integration occupational therapy, incorporating strategies for children with autism, anticipating schedule changes, and preparing children for new ex-

periences. The atypical sensory characteristics of the children in this sample included hypersensitivity to the following types of stimuli: auditory (42%), tactile (23%), oral (16%), visual (13%), and other (6%), and might also be seen in a sample of children with autism or sensory dysfunction. Although research describing atypical sensory reactions of children with fragile X syndrome has been reported (Baranek et al., 1999; Miller et al., 1999), we should note that no one has published research supporting the use of sensory integration with these children. Reviews of research studies examining the efficacy of sensory integration training with children with other types of disabilities have not substantiated its value. Small samples and poor research design have complicated research on this subject.

These findings suggest a unique combination of needs related to behavior management, attention deficit hyperactivity disorder, and autistic characteristics that have not been reported in other studies of young children with developmental delay. These characteristics are distinct from descriptions of children with Down syndrome, another genetic disorder characterized by mental retardation.

Considering that early interventionists in both surveys and interviews cited behavior management and consistency often as concerns, the need for close collaboration with parents seems obvious. To enhance the development of young boys with fragile X syndrome and support and sustain families, parents and professionals must work together to develop and implement consistent behavior management and to identify and implement methods and techniques for teaching skills and concepts across different environments. By working with parents as partners in instruction, early interventionists can address another important goal of early intervention: providing support and information to the families of young boys with fragile X syndrome.

Although parents of boys with fragile X syndrome often serve as an information source to teachers and other professionals (Bailey et al., 1999), our results indicate that early interventionists are also seeking information about fragile X syndrome on their

own. The recommendation of early interventionists to "learn as much about fragile X syndrome as possible" supports the idea of etiology-specific needs for these young children. Apparently, the early interventionists in our sample secured literature or resources on fragile X syndrome, and they must have found this information useful. We hope that the findings from this study add to that knowledge base, especially since the recommendations of the teachers themselves are seen as its most valuable contribution.

Even though our results suggest that boys with fragile X syndrome might have etiology-specific needs, the considerable variability demonstrated in their developmental status across time, as well as the variation in services provided, indicate that each child must be considered individually. The tremendous variability documented in this study and in our studies of developmental status (Bailey, Hatton, et al., 1998), autistic behavior (Bailey et al., 2000; Bailey, Mesibov, et al., 1998; Bailey et al., in press; Hatton & Bailey, in press), and temperament (Hatton et al., 1999) demonstrates that there might be as much variability within this etiology as across some disorders. Thus, although we can profit from descriptive studies such as this one, we must consider the uniqueness of the individual child, family, and teacher, as well as the context within which they function to design appropriate intervention plans.

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