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Mathematics Learning Disabilities in Girls With Fragile X or Turner Syndrome During Late Elementary School

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The present study focuses on math and related skills among 32 girls with fragile X (n = 14) or Turner (n = 18) syndrome during late elementary school. Performance in each syndrome group was assessed relative to Full Scale IQ–matched comparison groups of girls from the general population (n = 32 and n = 89 for fragile X syndrome and Turner syndrome, respectively). Differences between girls with fragile X and their comparison group emerged on untimed arithmetic calculations, mastery of counting skills, and arithmetic problem verification accuracy. Relative to girls in the comparison group, girls with Turner syndrome did not differ on untimed arithmetic calculations or problem verification accuracy, but they had limited mastery of counting skills and longer response times to complete the problem verification task. Girls with fragile X or Turner syndrome also differed from their respective comparison groups on math-related abilities, including visual–spatial, working memory, and reading skills, and the associations between math and those related skills. Together, these findings support the notion that difficulty with math and related skills among girls with fragile X or Turner syndrome continues into late elementary school and that the profile of math and related skill difficulty distinguishes the two syndrome groups from each other.

Keywords: *fragile X syndrome; Turner Syndrome; mathematics; mathematics learning disabilities; school-age children; genetics; dyscalculia; elementary age*

The prevalence of mathematics learning disabilities (MLD) in the general population, which is approximately 6% to 7% (Badian, 1983; Gross-Tsur, Manor, & Shalev, 1996), is comparable to that of reading disabilities, and yet less attention has been devoted to understanding MLD (Butterworth, 2005; Jordan & Montani, 1997; Mazzocco & Myers, 2003; Murphy, Mazzocco, Gerner, & Henry, 2006). Efforts to understand the underlying cognitive mechanisms leading to MLD may be enhanced by the study of genetic syndromes associated with poor math performance, such as fragile X and Turner syndrome (Mazzocco, 2001; see the appendix for syndrome details). The prevalence of MLD among girls with either of these common syndromes exceeds that in the general population during and beyond the primary school years (Mazzocco, 1998, 2001; Murphy et al., 2006; Rovet, 1993; Rovet, Szekely, & Hockenberry, 1994).

Despite evidence of persistent math difficulty in both syndromes, most studies of MLD in girls with fragile X or Turner syndrome have focused either on global mathematics achievement—rather than specific basic numeric skills—or on performance across individuals with a wide rather than narrow age range, or both. Thus, it is unclear how or whether the profile of relative difficulty associated with these syndromes changes over time. The present study focused on math and related skills among girls with fragile X or Turner syndrome in late elementary school.

Attention to late elementary school builds on two recent studies of primary school children with fragile X or Turner syndrome (Mazzocco, 2001; Murphy et al., 2006). From these studies, we know that math deficits are evident as early as kindergarten, even when girls with fragile X or Turner syndrome are compared to girls from an age- and IQ-matched sample (Mazzocco, 2001). Furthermore, this difficulty with math skills contrasts with performance on verbal tasks, such as reading words, where few differences emerge between the comparison group and either the fragile X (Mazzocco, 2001) or the Turner syndrome group (Mazzocco, 2001; Temple & Carney, 1996).

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We also know that math deficits are attributable to specific aspects of math performance rather than overall poor performance (Murphy et al., 2006). For example, despite age-appropriate mastery of rote counting skills, such as counting by ones, kindergarten girls with fragile X syndrome had more difficulty than their peers on aspects of applied counting, which included the ability to use one-to-one correspondence when counting and to identify the *n*th item in a set (Murphy et al., 2006). Difficulty with counting continues into third grade and distinguishes girls with fragile X syndrome from those with Turner syndrome and those in the general population (Mazzocco, Bhatia, & Lesniak-Karpiak, 2006).

In contrast, kindergarten girls with Turner syndrome demonstrated age-appropriate mastery of tasks dealing with quantity, including counting (Murphy et al., 2006), thus supporting a relative strength in number sense (Bruandet, Molko, Cohen, & Dehaene, 2004; Temple & Marriott, 1998). Although complex, *number sense* generally refers to the informal understanding of quantity (see Berch, 2005, for a review) and is distinguished from understanding of mathematical principles, which reflect more formal or learned rules. Despite the strength in number sense, by mid- to late elementary school, girls with Turner syndrome have difficulty relative to their peers on aspects of math, such as automaticity of fact retrieval, timed calculations, and procedural errors during problem solving (Rovet et al., 1994; Temple & Marriott, 1998).

Relative to what is known about poor math performance during primary school for girls with fragile X or Turner syndrome, less is known about the precise nature of math difficulty at later grades. As such, it is unclear whether the difficulties documented at early grades are also evident at later grades or represent a delay in acquiring the requisite procedure or skill. Also unclear is whether new difficulties emerge during the school-age years as more complex math concepts are introduced, such as place value. For example, in fragile X syndrome, limited mastery of conceptual knowledge, such as counting rules, may interfere with the strategies, such as finger counting, used to solve simple arithmetic problems. In turn, limited mastery of simple arithmetic problems may lead to difficulty with complex problems, such as multidigit calculation. In contrast, poor performance on multidigit calculation problems in Turner syndrome may reflect slow fact retrieval or poor procedural knowledge rather than basic number sense. The present study begins to address these questions by exploring math performance among girls with fragile X or Turner syndrome in late elementary school.

In the general population, knowledge of mathematical procedures and concepts as well as math performance can be influenced by competence in areas related to mathematics, such as working memory, visual–spatial ability, and reading (Geary, 2004). Based on this conceptualization, three subtypes of MLD are proposed (Geary, 1993, 2004), and each is associated with specific math-related skills. For example, the procedural and semantic memory subtypes of MLD are associated with working memory or executive function difficulty. Moreover, the semantic memory subtype can occur in conjunction with reading disability, highlighting the possible contribution of linguistic representation to math performance. In contrast, the visuospatial subtype of MLD is associated with visual–spatial deficits and their relation to poor math performance. As early as primary school, fragile X and Turner syndromes are associated with distinct cognitive characteristics in areas such as working memory (Kirk, Mazzocco, & Kover, 2005) and visual–spatial skills (Mazzocco et al., 2006) as well as difficulty with mathematics (Mazzocco, 2001). Efforts to understand these subtypes and the contribution of related skills to math performance may be strengthened by investigation into the nature of MLD in these syndromes. Toward that end, the present study examines math performance as well as visual–spatial, working memory, and reading-related skills during late elementary school among girls with fragile X or Turner syndrome.

Visual–spatial ability contributes to math performance in areas including the development of a mental number line, alignment of digits in calculation, and calculation skills, such as borrowing and carrying (Geary, 1994, p. 282). By third grade, specific rather than global visual–spatial deficits characterize girls with fragile X syndrome, whereas the visual–spatial deficits in Turner syndrome, though still selective, are more widespread (Mazzocco et al., 2006). Furthermore, an association between math and visual–spatial ability has been reported in fragile X (Mazzocco et al., 2006), but not in Turner syndrome (Mazzocco, 1998; Mazzocco et al., 2006; Rovet et al., 1994). However, in Turner syndrome, the introduction of concepts that rely on visual–spatial ability, such as multidigit calculations and place value, occur later during the school-age years and may enhance the association between math and visual–spatial ability (as reviewed by Murphy, Mazzocco, & McCloskey, in press). Alternatively, in both fragile X and Turner syndrome, an underlying skill common to both math and aspects of visual–spatial performance, such as working memory, may account for poor math performance, rather than visual–spatial ability per se (Buchanan, Pavlovic, & Rovet, 1998; Kwon et al., 2001; Mazzocco et al., 2006). Continued exploration of the association between math and visual–spatial ability in fragile X and Turner syndrome may inform the present understanding of the contributions of visual–spatial ability to MLD. Therefore, the present study focuses on specific aspects of math, including calculation, counting, and the association between math and visual–spatial ability.

Deficits in executive function are well documented in both fragile X (Bennetto, Pennington, Porter, Taylor, & Hagerman, 2001; Cornish et al., 2004; Kirk et al., 2005; Mazzocco, Pennington, & Hagerman, 1993) and Turner syndrome (Buchanan et al., 1998; Kirk et al., 2005; Temple, Carney, & Mullarkey, 1996). Among girls with Turner syndrome, slowed response times, suggestive of processing speed deficits, are also evident (e.g., Buchanan et al., 1998; Mazzocco, 2001; Temple, 2002) and may contribute to the reported executive dysfunction (Kirk et al., 2005; Temple et al., 1996). In both syndromes, deficits in executive function may influence math performance by interfering with the ability to manipulate information in working memory. For example, multidigit calculations require remembering and tracking the procedures needed to solve the problem as well as the ability to perform the calculations (e.g., Swanson & Beebe-Frankenberger, 2004). Also, among girls with Turner syndrome, slow processing speed may contribute to difficulties with fact retrieval automaticity and timed calculations. Examining the profile of math and related skills in late elementary school may inform the extent to which the relationships between these skills continue through elementary school. Toward that end, the present study includes multiple measures of executive function, including working memory and processing speed.

Reading ability is also related to mathematics, as suggested by the proposed semantic subtype of MLD (Geary, 1993, 2004). Moreover, a relative strength in reading ability may mediate aspects of math performance, especially word problem solving, and provide information regarding possible routes for intervention (Fuchs & Fuchs, 2007; Jordan, Kaplan, & Hanich, 2002). At age 10 years, a relative strength in reading is reported among girls with Turner syndrome, even relative to IQ-matched peers (Temple & Carney, 1996). During kindergarten, specific deficits in reading are not reported for girls with Turner syndrome or fragile X syndrome relative to age- and IQ-matched peers on the *Woodcock-Johnson Psychoeducational Battery–Revised* (WJ-R; Woodcock & Johnson, 1990) Word Attack subtest (Mazzocco, 2001). However, lack of significant differences, especially among girls with fragile X syndrome, may be attributable to the young age range of the study's sample (kindergarten or first grade). WJ-R Word Attack is likely to be difficult for most children during early primary school; thus, differences may only emerge at later ages, when mastery of reading is expected. To assess the contribution of reading skills to math performance, the present study includes reading measures, such as phonological decoding and reading speed and accuracy.

In summary, we focus on the extent to which the characteristics of MLD observed in third-grade girls with fragile X or Turner syndrome persist through the late elementary school years. We hypothesize that girls with fragile X or Turner syndrome will continue to have difficulty in math performance, relative to Full Scale IQ (FSIQ)–matched peers, but in different domains. Specifically, the conceptual difficulties observed among girls with fragile X syndrome, especially with regard to counting principles, suggest less accuracy in understanding of mathematic principles despite ready retrieval of information learned by rote. Among girls with Turner syndrome, the relative strength in basic aspects of number sense suggests mastery of some fundamental number skills. However, slower processing speeds suggest that differences may emerge in late elementary school on tasks that require automaticity, such as timed math calculation (Rovet et al., 1994). Also, we predict that the patterns of relative strengths and weaknesses reported in areas of visual–spatial ability, working memory, and reading will persist into the late elementary school years.

Method

Participants

All participants were selected from an ongoing study of math development in children (see Mazzocco, 2001; Mazzocco et al., 2006; Murphy et al., 2006). Girls with fragile X (n = 14) or Turner syndrome (n = 18) were selected if they were approximately 10 to 11 years of age, and had a FSIQ score of 80 or higher. The majority of girls in both groups were in Grade 5 or 6 at the time of testing; however, 3 girls were in Grade 4. Table 1 presents a summary of participant characteristics.

The comparison groups consisted of girls participating in a longitudinal, normative study of math development, described elsewhere (Mazzocco & Myers, 2003; Mazzocco & Thompson, 2005). Participants in the normative study were English-speaking children recruited in kindergarten from seven schools within a single large metropolitan school district (Mazzocco & Myers, 2003). These schools reflected the diverse ethnic and socioeconomic levels within the school district and excluded schools at the extreme ends of the socioeconomic distribution to minimize the influence of socioeconomic status and high mobility, which were associated with one another (Mazzocco & Myers, 2003).

Table 1
Demographic Characteristics of Participants and Matched Comparison Peers

		Grade			Age		FSIQ	
Group	*n*	4	5	6	*M*	*SD*	*M*	*SD*
Fragile X syndrome[a]	14	4	8	2	11.09	0.75	94.86	10.12
Comparison	32	2	30	0	10.71	0.26	96.88	4.48
Turner syndrome[a]	17	2	13	2	11.33	0.59	108.12	10.91
Comparison	89	1	88	0	10.64	0.26	110.87	10.26

Note: FSIQ = Full Scale IQ score on the four subtests of the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999).
a. FSIQ for one participant in this group is based on the two-subtest WASI.

Separate comparison groups were formed for the fragile X and Turner syndrome groups following the procedure of Kirk et al. (2005). All girls from the normative group who had completed the measures on which we report here and whose FSIQ scores fell within the range of the corresponding syndrome group were included. An independent sample *t* test was performed to determine whether each syndrome group differed from its proposed comparison group on FSIQ. No difference in FSIQ was found between girls with Turner syndrome and their proposed comparison group, so the Turner syndrome comparison group included all of the eligible children from the proposed comparison group ($n = 89$).

A difference in FSIQ was found between girls from the normative group and those with fragile X syndrome. To identify an FSIQ-matched comparison group, participants from the normative group were rank-ordered according to FSIQ. They were then excluded one by one, beginning with the child with the highest FSIQ, until the group difference was not statistically significant ($p = .35$) and the mean FSIQ fell within one standard error of measurement of the syndrome group. Of the 63 initial girls in the normative group, 32 were included in the IQ-matched comparison group for the girls with fragile X syndrome. Most girls in were in Grade 5; 2 girls were in Grade 4 at time of testing.

Materials

Cognitive Ability

The *Wechsler Abbreviated Scale of Intelligence* (WASI; Wechsler, 1999) was used to assess cognitive ability. Children completed the WASI in Grade 3, with the exception of 2 girls with fragile X syndrome who completed it during Grade 5. Internal consistency reliability coefficients for the FSIQ are .95 or greater for children in the study range.

Mathematics Measures

The Calculations subtest of the *Woodcock-Johnson Psychoeducational Battery–Revised* (Woodcock & Johnson, 1990) was used to assess overall mathematics ability, including basic arithmetic operations involving whole numbers, decimals, and fractions. The internal consistency reliability is .89 for ages 9 and 13 years (Woodcock & Mather, 1990).

Select items from the second edition of the *Test of Early Mathematics Ability* (TEMA-2; Ginsburg & Baroody, 1990) were also administered to some participants. The TEMA-2 is a standardized measure of both formal and informal math skills and is normed for children through 8 years of age. During each year of the larger study (discussed previously), children received the TEMA-2. After age 9, children received only those test items that they did not pass the previous year. Although the possibility of missing any single item by chance may lead to error, the purpose of including these items was to assess the proportion of children in each group who, in Grade 5, continued to miss items on a test normed for younger children.

Furthermore, a nonstandardized problem verification task was administered (Mazzocco, Bhatia, & Early, 2005). Performance on this task is facilitated by fluent math fact retrieval. Following practice problems designed to exemplify that some problems may have incorrect solutions, children were presented with a series of 56 two-operand arithmetic problems involving basic arithmetic operations (e.g., $2 + 2$). An answer to each problem was also provided; however, the provided answer could be either correct (e.g., $2 + 2 = 4$) or incorrect (e.g., $2 \times 2 = 10$). For each problem, the participant was asked to “decide quickly whether the answer to the problem was right or wrong” without actually calculating the answer. Acceptable answers included “right,” “wrong” and “I don’t know” responses; however, the child was encouraged to respond “right” or “wrong” to as many problems as possible. Items consisted of both number facts (e.g., $4 + 5 = 9$) and number knowledge (e.g., $200 - 150 = 300$). Practice items ensured that the child understood the task. Each child was given 10 seconds before receiving a prompt from the examiner to "answer quickly" coupled with a finger snap. The total percentage correct and total time to complete the task were computed. The instructions for this item formed the basis for a verbal memory measure (discussed subsequently).

Data from a nonstandardized measure of counting skill (based on Geary, Bow-Thomas, & Yao, 1992; and Gelman & Meck, 1983) were also available for some participants. In this activity, the child watched the examiner count linear sequences of colored dots. For each sequence, she was asked to determine whether the examiner counted "the right number of dots." The 24 test trials were preceded by a warm-up where the child was taught that the examiner might count incorrectly. Correct counting could either follow conventional counting principles, such as counting items in order, or unconventional ones, such as counting items out of sequence. Incorrect counting items violated counting principles, for example, by counting some dots more than once.

The counting measure was initially administered to all children during second grade. Any children who missed more than two items received the counting task the following year, until no more than two items were missed. Based on this procedure, we were able to determine how many children in each group continued to receive this task in Grade 5, and what proportion of those children continued to have limited mastery of counting beyond Grade 5.

Girls in the fragile X or Turner syndrome groups also received the *KeyMath–Revised* Numeration, Geometry, Measurement, and Time and Money subtests (Connolly, 1998) to measure their understanding of basic math concepts (e.g., Numeration and Geometry), and applied math skills (Measurement, Time and Money). Internal consistency reliabilities for these subtests range from .61 to .85 for children in Grades 4 and 6 (Connolly, 1998). No reliabilities are reported for Grade 5. Data from these measures were not available for girls in the comparison group, so the performance of girls with fragile X or Turner syndrome was compared to the age-referenced norms provided in the manual (Connolly, 1998).

Visual Perceptual Measures

The *Beery-Butenica Developmental Test of Visual–Motor Integration* (VMI; Beery, 1997) was administered to assess visual–motor coordination. On this test, the child was asked to copy designs of varying complexity. The average internal consistency reliability of the VMI is .88 (Beery, 1997).

Furthermore, two motor reduced subtests of the second edition of the *Developmental Test of Visual Perception* (DTVP-2; Hammill, Pearson, & Voress, 1993) were administered: Position in Space and Figure Ground. Both subtests involve scanning and comparing each of several visual stimuli with a target stimulus of one (Position in Space) or many (Figure Ground) shapes. Standardized administration of these subtests is untimed, and testing stops once a child reaches a performance ceiling. We employed a modified administration that involved timing children on each of the two subtests (without their knowledge) and administering the last 14 or 11 items of each subtest, respectively, to all children, to have timed testing include the same number of test items for all children. Note that these modifications did not involve changing the standardized instructions given at the onset of each trial. The Position in Space and Figure Ground subtests are normed for use with children ages 4 through 10 years. The internal consistency reliability for the Position in Space and Figure Ground subtests is .84 and .83, respectively, for 10-year-olds.

Immediately following the Figure Ground subtest, a nonstandardized measure of incidental visual–spatial memory was administered (see Mazzocco et al., 2006). In this test, the child was given a drawing of an empty box and cutouts of the individual shapes that appeared in that box during the last 11 pages of the Figure Ground test. She was then asked to recall "where these shapes belong" in the box. This test is used to assess memory for spatial location.

An additional experimental task, the Paired Position in Space test (PPS), was administered to assess the potential role of visual scanning efficiency on the Position in Space subtest. The PPS test included all of the same stimuli that appear in the standardized Position in Space test. However, rather than present each target stimulus to the left of a horizontal array of choices (as is done during the standard version of the Position in Space subtest), the target figure appeared directly above each item in the forced choice arrays. For each of these pairs of drawings, the child was asked to mark whether the two drawings were the same or different by marking an *s* or *d* above each pair. The child was timed unobtrusively. The paired presentation of stimuli eliminated the need for distant scanning when comparing the target stimuli to their potentially matching stimuli. Therefore, although we anticipated that children's performance on the PPS task would be more accurate than their own performance on the standard version, we predicted a Group × Task version interaction, such that girls with Turner syndrome would show a greater increase in accuracy than girls without Turner syndrome. That is, if visual scanning during the Position in Space subtest is inefficient (as we predicted it would be for the group with Turner syndrome), the relative gains made in accuracy should be greater once scanning needs are diminished, relative to the gains observed for children whose scanning performance is already efficient. We used the difference in accuracy scores between standard and paired Position in Space tasks as an indicator of whether scanning inefficiency contributed to less accurate performance on the Position in Space subtest. Also, for children with versus without Turner syndrome, group differences in response time should be greater for the standard versions of the Position in Space task (for which there are greater scanning demands) than for the paired version.

Verbal Memory and Working Memory Measures

A nonstandardized measure of verbal memory was administered. This measure assessed recall of the main idea (or "gist") rather than verbatim recall and was derived from the problem verification task (discussed previously). Prior to administering the problem verification task, each child listened to instructions for the task. Following the presentation of the instructions (a standardized procedure that took approximately 2 min), each child was asked to recall the instructions: "Let's review what is going to happen in this activity. Carefully think about the instructions. Tell me everything you remember." Children were allowed 3 min to recall as many of the instructions as possible. Recall did not need to be verbatim, but needed to describe the central idea for each instruction. Children were prompted, "Anything else?" until they recalled all of the instructions, indicated that they did not remember any others, or 3 min had passed. There were 14 possible instructions to recall. The percentage of total items recalled was calculated for each child.

The Memory for Digits subtest of the fourth edition of the *Stanford-Binet Intelligence Scale* (SB-IV; Thorndike, Hagen, & Sattler, 1986) was included to assess verbatim recall. A total score was obtained for both forward and backward digit recall. The internal consistency reliabilities for this subtest are greater than .80 for the study age range.

Furthermore, the *Contingency Naming Test* (CNT; Anderson, Anderson, Northam, & Taylor, 2000; Taylor, Albo, Phebus, Sachs, & Bierl, 1987) was administered to assess executive function, including working memory and reactive flexibility, following the procedure described by Kirk et al. (2005). On this Stroop-like test, participants were overtly timed while naming a series of objects according to a set of increasingly difficult rules. The warm-up trials involved basic naming of the objects (i.e., by color and then by shape). On the experimental trials, the objects were named following a one- or two-attribute switching rule that dictated whether the child should name the color or the shape.

The response time to name the set of objects, number of self-corrections, and number of errors were recorded. An efficiency score was calculated to reflect the tradeoff between naming speed and accuracy (Anderson et al., 2000; Kirk et al., 2005) according to the following formula: 100 times the product of inverse of response time divided by the square root of the number of errors plus one.

Reading-Related Measures

The Word Attack subtest from the WJ-R was used to assess phonological decoding skills, or nonword reading. This measure is a standardized measure of academic achievement with an internal consistency reliability greater than .87 for ages 9 and 13 years (Woodcock & Mather, 1990).

The Colors, Numbers, and Letters subtests of the *Rapid Automatized Naming* task (RAN; Denckla & Rudel, 1974) was used to assess phonological retrieval automaticity. On each subtest, the child is asked to name sequences of colors, numbers, or letters as quickly as possible. Response times were obtained for each subtest. Furthermore, a nonstandardized measure of reading speed was administered. In this measure, the child read a paragraph consisting of 199 words. Although each child was allowed to finish reading the paragraph, the examiner noted the number of words read in 2 min. If the paragraph was completed in less than 2 min, a score of 200 words was recorded. Response time was obtained, as was the total number of errors made in 2 min; both were compared across groups. Response times were compared also between subtests, across groups.

Results

Normality of the data could not be determined due to the small sample size in both syndrome groups; therefore, nonparametric statistics were conducted. The Mann-Whitney test was used to examine differences in performance between girls with fragile X or Turner syndrome and their respective comparison groups. *Z* scores are reported for all analyses and are adjusted for tied rankings when required. Comparisons examining group differences in frequency were conducted using the chi-square test. In cases where the expected cell value was less than 5, a Fisher's exact test is indicated. All of the reported *p* values are two-tailed. Data are presented in Tables 2 and 3 for fragile X and Turner syndrome, respectively.

Table 2
Summary of Results for Girls With Fragile X Syndrome and Their Comparison Group

Variable	Fragile X Syndrome		Comparison		
	M	*SD*	*M*	*SD*	Cohen's *d*
Math measures					
WJ-R Calculations (SS)	89.43**	14.34	100.09	9.88	0.87
TEMA-2 (*n* items given)	13.85**	8.00	7.78	4.58	0.96
Problem verification task					
Accuracy (%)	60.16*	0.14	70.59	0.09	0.92
RT	212.92	98.44	245.13	75.37	0.39
KM-R (SS)[a]					
Numeration	8.64	3.32	—	—	
Geometry	8.14	3.23	—	—	
Measurement	8.00	3.70	—	—	
Time and Money	7.86	2.38	—	—	
Visual perceptual measures					
VMI (SS)	84.07	6.87	86.28	9.17	0.26
DTVP-2					
Figure Ground					
SS	10.27	2.80	9.88	2.35	0.16
RT	180.73	53.12	158.56	29.43	0.59
Figure Ground Memory					
n correct (of 10)	4.82**	3.03	7.59	2.01	1.07
RT	68.82	27.05	65.58	34.55	0.10
Position in Space					
SS	7.73	3.85	9.50	2.30	0.62
RT	133.73	37.37	118.48	24.12	0.54
PPS					
n correct (paired–standard)	2.45	2.12	2.00	1.69	0.25
RT	300.64	76.14	286.75	70.87	0.19
Verbal and working memory measures					
Verbal memory (*n* of 14)	2.69**	1.25	4.19	1.82	0.83
SB-IV Memory for Digits					
Forward recall (total)	5.36	1.55	5.78	1.18	0.32
Backward recall (total)	4.64	1.69	5.06	1.37	0.29
CNT rule efficiency					
One attribute	1.47	0.92	1.82	0.52	0.53
Two attribute	0.50**	0.36	1.08	0.51	1.09
Reading-related measures					
WJ-R Word Attack (SS)	91.64**	11.08	103.50	15.05	0.80
RAN (RT)					
Colors	45.00**	9.00	36.06	5.61	1.13
Numbers	28.07	8.83	25.44	4.98	0.41
Letters	27.71	8.11	25.47	5.00	0.37
Colors–Numbers	16.93	10.83	10.63	5.19	0.81
Informal reading					
Accuracy	8.79	5.06	7.50	2.94	0.35
RT	89.64	20.40	88.84	16.78	0.05

Note: WJ-R = *Woodcock-Johnson Psychoeducational Battery–Revised* (Woodcock & Johnson, 1990); TEMA-2 = *Test of Early Mathematics Ability* (2nd ed.; Ginsburg & Baroody, 1990); SS = standard score; RT = response time in seconds; KM-R = *KeyMath–Revised* (Connolly, 1998); VMI = *Beery-Butenica Developmental Test of Visual–Motor Integration* (Beery, 1997); DTVP-2 = *Developmental Test of Visual Perception* (2nd ed.; Hammill, Pearson, & Voress, 1993); PPS = Paired Position in Space subtest; SB-IV = *Stanford-Binet Intelligence Scale* (4th ed.; Thorndike, Hagen, & Sattler, 1986); CNT = *Contingency Naming Test* (Anderson, Anderson, Northam, & Taylor, 2000); RAN = *Rapid Automatized Naming* (Denckla & Rudel, 1974). Dashes indicate that data were not available.
a. An average score is based on a mean of 10; scores between 7 and 13 are considered to be in the average range (Connolly, 1998).
$^{*}p \le .05$. $^{**}p \le .01$. $^{***}p \le .001$.

Table 3
Summary of Results for Girls With Turner Syndrome and Their Comparison Group

Variable	Turner Syndrome		Comparison		
	M	*SD*	*M*	*SD*	Cohen's *d*
Math measures					
WJ-R Calculations (SS)	107.82	21.09	110.76	15.04	0.18
TEMA-2 (*n* items given)	7.47*	5.50	4.36	3.67	0.75
Problem verification task					
Accuracy (%)	80.0	0.12	77.0	0.10	0.30
RT	275.00	85.01	225.51	60.76	0.74
KM-R (SS)[a]					
Numeration	11.80	2.86	—	—	
Geometry	11.93	2.99	—	—	
Measurement	9.93	3.62	—	—	
Time and Money	10.13	3.25	—	—	
Visual perceptual measures					
VMI (SS)	86.40*	13.72	92.97	11.83	0.54
DTVP-2					
Figure Ground					
SS	8.50**	1.79	10.16	2.22	0.74
RT	191.64***	32.82	153.16	26.45	1.27
Figure Ground Memory					
n correct (of 10)	4.71***	2.53	7.67	1.95	1.31
RT	80.18	34.20	67.85	31.54	0.39
Position in Space					
SS	10.17	1.85	10.09	1.94	0.04
RT	132.92	47.66	117.09	31.53	0.47
PPS					
n correct (paired–standard)	1.50	1.68	1.55	1.55	0.03
RT	308.46	88.28	266.48	59.80	0.64
Verbal and working memory measures					
Verbal memory (*n* of 14)	4.14	1.88	4.26	1.53	0.08
SB-IV Memory for Digits					
Forward recall (total)	6.06	1.82	6.61	1.68	0.32
Backward recall (total)	4.06***	1.09	5.71	1.61	1.00
CNT Rule efficiency					
One attribute	1.80	0.62	2.02	0.55	0.39
Two attribute	1.14	0.55	1.28	0.51	0.27
Reading-related measures					
WJ-R Word Attack (SS)	101.12*	14.61	110.54	14.79	0.62
RAN (RT)					
Colors	39.47*	6.41	35.31	7.23	0.57
Numbers	23.71	3.98	24.48	4.51	0.17
Letters	23.24	3.46	24.52	4.40	0.30
Colors–Numbers	15.76**	7.01	10.83	6.01	0.77
Informal reading					
Accuracy	6.29	3.37	6.19	3.26	0.12
RT	74.94	17.37	78.04	16.19	0.19

Note: WJ-R = *Woodcock-Johnson Psychoeducational Battery–Revised* (Woodcock & Johnson, 1990); TEMA-2 = *Test of Early Mathematics Ability* (2nd ed.; Ginsburg & Baroody, 1990); SS = standard score; RT = response time in seconds; KM-R = *KeyMath–Revised* (Connolly, 1998); VMI = *Beery-Butenica Developmental Test of Visual–Motor Integration* (Beery, 1997); DTVP-2 = *Developmental Test of Visual Perception* (2nd ed.; Hammill, Pearson, & Voress, 1993); PPS = Paired Position in Space subtest; SB-IV = *Stanford-Binet Intelligence Scale* (4th ed.; Thorndike, Hagen, & Sattler, 1986); CNT = *Contingency Naming Test* (Anderson, Anderson, Northam, & Taylor, 2000); RAN = *Rapid Automatized Naming* (Denckla & Rudel, 1974). Dashes indicate that data were not available.
a. An average score is based on a mean of 10; scores between 7 and 13 are considered to be in the average range (Connolly, 1998).
$*p \leq .05$. $**p \leq .01$. $***p \leq .001$.

Fragile X Syndrome Versus Matched Comparison

Math Measures

Girls with fragile X syndrome had lower scores on the WJ-R Calculations subtest than girls in the comparison group, $Z = -2.69$, $p = .007$. Even in Grade 5, all children in both groups required additional TEMA-2 items. Girls with fragile X syndrome needed 14 items, which was almost twice the number of items required by girls in the comparison group (8 items), $Z = -2.60$, $p = .009$. On the problem verification task, girls with fragile X syndrome were less accurate at judging arithmetic solutions than were girls in the comparison group, $Z = -2.35$, $p = .019$, but the two groups did not differ on the time required to complete the task ($p = .11$).

Prior to the Grade 5 assessment, the majority of girls in the fragile X syndrome comparison group (84%) achieved mastery on the counting task, relative to 33% of the girls with fragile X, $\chi^2(1, N = 44) = 10.92$, Fisher's exact $p = .002$. Among the 8 girls with fragile X who had not demonstrated mastery, 6 girls (75%) achieved mastery by obtaining a score of 22 or better (out of 24) on the task at the time of assessment. Although this difference was not significantly different (Fisher's exact $p = .10$) from the subset of the comparison group who had not yet met the criterion for mastery prior to Grade 5, only 1 (20%) of the 5 children in the comparison group obtained a score of 22 or more at their Grade 5 assessment.

Performance on the *KeyMath–Revised* among girls with fragile X syndrome was compared to the age-based norms provided by the test designers. An average score is based on a mean of 10; scores between 7 and 13 are considered to be in the average range. Based on these norms, the overall performance of girls with fragile X on the Numeration, Geometry, Measurement, and Time and Money subtests was in the average range for their age.

Visual Perceptual Measures

No differences were found between girls with fragile X syndrome and their comparison group on the VMI, DTVP-2 Position in Space, or Figure Ground standard scores (*p*s = .20 to .68). Also, response times to complete the Position in Space and Figure Ground subtests did not differ between the two groups (*p*s > .10). Relative to the comparison group, girls with fragile X syndrome took comparable amounts of time to complete the Figure Ground Memory task ($p = .42$) but correctly recalled fewer locations, $Z = -2.66$, $p = .008$. Moreover, all of the girls in the comparison group arranged the shapes according to the 2 × 5 grid in the standardized Figure Ground task, but only about half of the girls with fragile X syndrome created grid-like arrangements, $\chi^2(1, N = 43) = 16.46$, Fisher's exact $p < .001$. The remaining 46% of girls with fragile X syndrome did not align the shapes according to a grid, and instead created a scattered array of shapes. On the PPS task, no differences in response time to complete the task were found between girls with fragile X syndrome and their comparison group ($p = .42$), nor did the difference in performance accuracy on the paired versus standard task distinguish the two groups ($p = .54$).

Verbal and Working Memory Measures

Girls with fragile X syndrome recalled fewer items on our measure of verbal memory than girls in the comparison group, $Z = -2.62$, $p = .009$. However, no differences were found between girls with fragile X syndrome and the comparison group on either the forward or backward digit span tasks (*p*s > .50). On the CNT, girls with fragile X syndrome had significantly lower efficiency scores than girls in the comparison group in applying the two-attribute rule, $Z = -3.15$, $p = .002$, but not the one-attribute rule ($p = .08$). Follow-up analyses on the two-attribute rule revealed that lower efficiency among the girls with fragile X syndrome relative to the comparison group was attributable to more errors made by girls with fragile X syndrome, $Z = -3.30$, $p = .001$, rather than longer response times ($p = .09$). Thus, on the most challenging rule, girls with fragile X syndrome traded accuracy for speed relative to girls in the comparison group.

Reading-Related Measures

On the Word Attack measure, girls with fragile X syndrome had lower standard scores than girls in the comparison group, $Z = -2.65$, $p = .008$; however, scores for both groups were in the average range based on age-referenced norms. Girls with fragile X syndrome had longer response times on the RAN Colors subtest than did girls in the comparison group, $Z = -3.09$, $p = .002$. Group differences in response time were not found on RAN Numbers or Letters (*p*s > .40) or on response time discrepancy (RAN Colors–RAN Numbers; $p = .07$). Also, no differences were found on the informal measure of reading fluency (*p*s > .80).

Summary

Relative to their peers, girls with fragile X syndrome had lower scores on WJ-R Calculations, were less accurate on the problem verification task, were less likely to have achieved mastery of counting principles, and required almost twice the number of TEMA-2 items to be administered due to prior failure on these items. Also, girls with fragile X syndrome had selective deficits on the visual perceptual measures, verbal memory, and working memory. Although girls with fragile X syndrome had longer RAN Colors response times, their overall reading skills did not distinguish them from their comparison group.

Turner Syndrome Versus Matched Comparison

Math Measures

In contrast to girls with fragile X syndrome, girls with Turner syndrome did not differ from girls in their comparison group on the WJ-R Calculations subtest ($p = .48$). The majority of girls with Turner syndrome and their comparison group required additional TEMA-2 items. Girls with Turner syndrome needed an average of seven items, nearly twice the number of items required by girls in their comparison group, who needed four items on average, $Z = –2.42$, $p = .015$. On the problem verification task, accuracy across trials did not differ between girls with Turner syndrome and their comparison group ($p = .21$). However, girls with Turner syndrome took about 50 s longer to complete the task (see Table 3). Although this response time difference was not statistically significant ($p = .06$), the effect size was moderate (Cohen's $d = 0.74$).

Prior to the Grade 5 assessment, the majority of girls in the Turner syndrome comparison group (94%) had demonstrated mastery on the counting task, relative to 65% of the girls with Turner syndrome, $\chi^2(1, N = 106) = 13.52$, Fisher's exact $p = .002$. All 6 of the girls with Turner syndrome who had not met criteria for mastery of counting prior to Grade 5 demonstrated mastery at the present assessment by obtaining a score of 22 or better (out of 24) on the task. Although this difference was not significantly different (Fisher's exact $p = .06$) from the comparison group, only 2 (40%) of the 5 children in the comparison group obtained a score of 22 or more.

Based on age-referenced norms from the *KeyMath–Revised*, the overall performance of girls with Turner syndrome was in the average range on the Numeration, Geometry, Measurement, and Time and Money subtests.

Visual Perceptual Measures

Girls with Turner syndrome had lower scores than their comparison group on the VMI, $Z = –2.49$, $p = .013$, and the DTVP-2 Figure Ground subtests, $Z = –2.83$, $p = .005$. Girls with Turner syndrome also took longer than the comparison group to complete the Figure Ground subtest, $Z = –3.75$, $p < .001$. No differences in standard score or response time were found between the two groups on the Position in Space subtest ($ps > .20$). Relative to the comparison group, girls with Turner syndrome took comparable amounts of time to complete the Figure Ground Memory task ($p = .19$) but correctly recalled fewer locations, $Z = –3.91$, $p < .001$. In contrast to girls with fragile X syndrome, no difference was found for grid-like versus non-grid-like shape arrangements (Fisher's exact $p = 1.0$). All of the girls with Turner syndrome and all but one of the girls in the comparison group arranged the shapes according to the 2 × 5 grid appearing in the standardized Figure Ground task. On the PPS task, no group differences emerged for either the response time to complete the task ($p = .09$) or for the difference in performance accuracy on the paired versus standard task ($p = .89$).

Verbal and Working Memory Measures

No difference was found in the number of items recalled on the verbal memory measure ($p = .89$). However, girls with Turner syndrome had lower scores than their comparison group on backward digit recall, $Z = –3.91$, $p < .001$, but not forward recall ($p = .24$). Contrary to expectations, no differences were found on the CNT between groups on either the one- or two-attribute switching rules ($ps > .28$). These results are inconsistent with other reports of CNT performance in girls with Turner syndrome (Kirk et al., 2005) and may reflect the effect of limiting the participants in the present sample to those with a higher range of IQ scores.

Reading-Related Measures

On the Word Attack task, girls with Turner syndrome had lower standard scores than girls in the comparison group, $Z = –2.05$, $p = .040$; however, the mean score among girls with Turner syndrome was within the average range ($M = 101.12$, range = 79 to 128). Girls with Turner syndrome had longer response times on the RAN Colors subtest than girls in the comparison group, $Z = –2.49$, $p = .013$. No differences in response time were found on RAN Numbers or Letters subtests ($ps > .35$), although the difference in response time between the RAN Colors and Numbers subtests was greater among girls with Turner syndrome than girls in the comparison group, $Z = –3.10$, $p = .002$. No difference was found for reading speed or accuracy ($ps > .49$).

Summary

Relative to their peers, girls with Turner syndrome did not differ on WJ-R Calculations; they took longer to complete the problem verification task, but they were just as accurate; they were less likely to have achieved mastery of counting principles and required almost twice the number of TEMA-2 items. Deficits on visual perceptual measures

Table 4
Spearman Rank Correlations Between WJ-R Calculation Scores and Related Visual Perceptual, Memory, and Reading Measures

	Fragile X				Turner			
	Syndrome[a]		Comparison[b]		Syndrome[c]		Comparison[d]	
Measure	ρ	*p*	ρ	*p*	ρ	*p*	ρ	*p*
Visual perceptual measures								
VMI (SS)	.32	.27	.10	.58	.29	.29	.30	.004**
DTVP-2								
Position in Space (SS)	.65	.03*	.03	.88	.33	.30	.13	.220
Figure Ground (SS)	–.04	.91	–.11	.57	.11	.71	–.06	.570
Figure Ground Memory (*n* correct)	.31	.35	.17	.37	.48	.08	–.02	.890
Verbal and working memory measures								
SB-IV Memory for Digits								
Forward recall (total)	.34	.24	–.20	.28	–.06	.82	.13	.250
Backward recall (total)	.48	.08	.09	.61	.33	.19	.38	< .001***
CNT rule efficiency								
One attribute	.74	.009**	–.11	.54	–.21	.43	.15	.160
Two attribute	.73	.01*	–.04	.83	.22	.41	.10	.360
Reading-related measures								
WJ-R Word Attack (SS)	.76	.002**	.15	.43	.22	.39	.38	< .001***
RAN (RT)								
Colors	–.57	.03*	.18	.32	–.25	.34	–.27	.010**
Numbers	–.57	.03*	.09	.62	.05	.86	–.32	.002**
Letters	–.63	.02*	.10	.59	–.36	.16	–.28	.009**
Informal reading (RT)	–.54	.045*	–.08	.68	–.29	.26	–.48	< .001***

Note: SS = standard score; RT = response time in seconds; VMI = *Beery-Butenica Developmental Test of Visual–Motor Integration* (Beery, 1997); DTVP-2 = *Developmental Test of Visual Perception* (2nd ed.; Hammill, Pearson, & Voress, 1993); SB-IV = *Stanford-Binet Intelligence Scale* (4th ed.; Thorndike, Hagen, & Sattler, 1986); CNT = *Contingency Naming Test* (Anderson, Anderson, Northam, & Taylor, 2000); WJ-R = *Woodcock-Johnson Psychoeducational Battery–Revised* (Woodcock & Johnson, 1990); RAN = *Rapid Automatized Naming* (Denckla & Rudel, 1974).
a. $n = 14$.
b. $n = 32$.
c. $n = 17$.
d. $n = 89$.
$*p \leq .05$. $**p \leq .01$. $***p \leq .001$.

and selective working memory measures were also found. The RAN Colors–Numbers difference in response time was greater among girls with Turner syndrome relative to their comparison group, suggesting selective processing deficits in Turner syndrome.

Correlations Between Math and Related Measures

Spearman rank correlations were conducted between girls with fragile X or Turner syndrome and their respective comparison groups to assess the relationship between math performance, as measured by the WJ-R Calculations subtest, and each related area (see Table 4).

Fragile X Versus Matched Comparison

Many significant correlations were observed between the WJ-R Calculations subtest and each of the related skill areas among girls with fragile X syndrome. Specifically, the WJ-R Calculations standard score was positively correlated with CNT efficiency on the one- and two-attribute rules. Similarly, all of the measures of reading skills were significantly related to WJ-R Calculations scores (ρs = –.54 to .76, *p*s = .045 to .002). Of the visual–spatial measures, only the DTVP-2 Position in Space standard score was associated with WJ-R Calculations performance. The remaining measures—the DTVP-2 Figure Ground subtest, the number correct on Figure Ground Memory, and VMI—were not significantly associated with WJ-R Calculations scores (*p*s > .25). In contrast to the fragile X syndrome group, no significant correlations were observed between the WJ-R Calculations score and any of the related areas in the comparison group (*p*s > .25).

Turner Syndrome Versus Matched Comparison

In contrast to fragile X syndrome, no significant correlations were observed between the WJ-R Calculations score and any of the related areas among girls with Turner syndrome (*p*s > .08). However, several correlations were observed between the WJ-R Calculations subtest and each of the related skill areas among girls in the comparison group. In the comparison group, WJ-R Calculations standard score was positively related to Memory for Digits backward recall but not to forward recall or the CNT (*p*s > .10). All measures of reading skills were significantly related to WJ-R Calculations scores (ρs = –.27 to .38, *p*s = .01 to < .001). Of the visual–spatial measures, only the VMI standard score was associated with the WJ-R Calculations score. The remaining DTVP-2 measures were not significantly associated with WJ-R Calculations scores (*p*s > .20).

Summary

The pattern of correlations between math and related skills distinguished girls in each syndrome group from each other and from their comparison group, suggesting that different pathways contribute to math performance across groups.

Discussion

The present study examined math and related skills performance among 9- to 12-year-old girls with fragile X or Turner syndrome to assess whether the characteristics of MLD present in primary school continue into late elementary school. Although this was not a longitudinal study, more than half of the participants had also been included in our previous studies of Grade 3 (Kirk et al., 2005; Mazzocco et al., 2006). We predicted that girls with fragile X syndrome would have difficulty understanding and applying mathematical principles, and girls with Turner syndrome would continue to manifest slower processing speeds and difficulty on tasks requiring automaticity, such as timed math calculations. Furthermore, we predicted that the within-group profiles of visual–spatial, working memory, and reading skills observed during primary school would also persist into late elementary school. Consistent with our predictions, the results suggest that girls with fragile X syndrome have incomplete mastery of calculation and difficulty with number sense, including counting, relative to peers matched on cognitive ability. In contrast, girls with Turner syndrome are distinguished from their cognitive ability–matched peers primarily on tasks measuring speed of processing. Although there are some similarities in the overall profile of math and related skills between the two syndromes, distinct profiles are evident for both syndromes relative to their respective comparison groups.

Math Skills of Girls With Fragile X Syndrome

In the present study, girls with fragile X syndrome showed poorer performance than their comparison group on math calculations. These results contrast with our earlier findings documenting poorer performance on calculations among third-grade girls with fragile X relative to the general population before, but not after, matching the samples based on FSIQ (Mazzocco et al., 2006). Girls with fragile X syndrome were also less accurate at judging arithmetic solutions, despite taking the same amount of time to complete the task. Although not all items on this measure were number facts, performance on this task is facilitated by fluency of math fact retrieval. Consistent with our predictions, these results suggest that the time needed to process numerical information does not distinguish girls with fragile X syndrome from their comparison group; rather, their limited knowledge of counting principles and number facts may contribute to poor math performance, including calculation skills, at least in late elementary school.

Additional investigation is needed to determine the causes of poor calculation performance, as knowledge of mathematical principles or concepts not measured in the present study—such as recognizing that numbers can be added in any order (commutativity)—may also contribute to poor math performance. However, as with counting skills, it is possible that difficulty with calculation reflects specific rather than global deficits. Rivera et al. reported that accuracy at verifying basic math facts involving two operands does not differ from age-matched peers among female individuals with fragile X syndrome between 10 and 23 years of age (Rivera, Menon, White, Glaser, & Reiss, 2002; S. Rivera, personal communication, July 20, 2006). Thus, one explanation is that rote calculation skills, such as recalling basic number facts, may be a strength for girls with fragile X syndrome relative to skills that reflect understanding of number concepts (e.g., place value and overall number sense). Indeed, girls in Rivera et al.'s study were less accurate than their peers at judging the accuracy of problems with three operands. Such a profile would be consistent with the observed strength on rote, but not applied counting, as reported during primary school (Murphy et al., 2006), and with the lack of group differences on our verbal memory tasks (as reported earlier).

Alternatively, difficulty with calculation and accuracy at judging arithmetic solutions may reflect continued difficulty with formal and informal math skills, including understanding counting principles. Evidence in support of this notion includes the number of girls from both syndrome groups who had failed to master developmentally appropriate math tasks by 10 years of age. Although all of the girls in both the fragile X and comparison group had failed to pass some items on the TEMA-2 prior to Grade 5, the number of items failed was higher among girls with fragile X syndrome than girls in the comparison group. Because the TEMA-2 is normed for children through age 8, failing fewer items suggests earlier, age-appropriate mastery of the skills measured by the TEMA-2 by girls in the comparison group, and a lack of age-appropriate performance by girls with fragile X syndrome. Consistent with the TEMA-2 findings, the number of girls who had not mastered performance on the counting principles task was considerably larger among girls with fragile X syndrome (67%) than in the comparison group (16%). Taken together, these findings are consistent with the notion that difficulty with aspects of number sense, including counting, and more complex math skills, such as calculation, persist even into late elementary school among girls with fragile X syndrome. Continued difficulty in these areas suggest that by Grade 5, girls with fragile X syndrome fall further behind their peers in acquiring the basic math skills needed to acquire the more complex mathematical concepts introduced in later grades.

Math Skills of Girls With Turner Syndrome

In contrast to fragile X syndrome, girls with Turner syndrome did not differ from their comparison group on math calculations. At first glance, this finding is inconsistent with the notion of poor math performance in Turner syndrome (Mazzocco, 2001; Murphy et al., 2006; Rovet, 1993). However, the girls with Turner syndrome continued to receive more items on the TEMA-2 than their comparison group, suggesting later mastery of the skills measured by the TEMA-2. More girls with Turner syndrome than in the comparison group also continued to demonstrate incomplete mastery of counting skills in Grade 5. Similar to fragile X syndrome, difficulty with counting skills is common for girls with Turner syndrome relative to their comparison group, who represent a minority of the general population. Moreover, the WJ-R Calculations subtest is not overtly timed. Thus, the nonsignificant findings are consistent with those of Rovet et al. (1994) who reported poorer performance among girls with Turner syndrome relative to age- and grade-matched peers on timed but not untimed math tests. Indeed, girls with Turner syndrome were just as accurate as girls in the comparison group at detecting correct and incorrect problems on our problem verification task but took much longer to complete the task. Although the response time difference was not statistically different, the moderate effect size (Cohen's $d = 0.74$) is noteworthy. Thus, our results support the notion that certain math skills, such as calculation, may be influenced by time demands in Turner syndrome.

At present, the mechanisms underlying a possible deleterious effect of overt timing on math performance in Turner syndrome are unclear. One explanation is that individuals with Turner syndrome rely more heavily than their peers on alternative strategies, such as finger counting, rather than on retrieval to solve calculation problems (Bruandet et al., 2004). Continued reliance on immature problem-solving strategies may slow response times and result in more time needed to complete a given task, but not necessarily in less accurate performance. Alternatively, slow processing speed, which has been documented among female individuals with the syndrome (e.g., Bruandet et al., 2004; Temple & Marriott, 1998), may account for longer response times and poor performance on timed tasks. For example, slower processing speed may result in girls with Turner syndrome completing fewer items during the same fixed amount of time relative to peers. If this is the case, scores based on the total number of items correct should be lower among girls with Turner syndrome relative to peers than scores based on the number correct out of the number of items completed. Empirically evaluating causal mechanisms was beyond the scope of the present study; however, the notion of slow processing speed in Turner syndrome has received some support from our measures of cognitive ability (discussed subsequently) and underscores the utility of considering the cognitive correlates of MLD in addition to basic math skills.

Math-Related Skills

Math-related skills, such as visual–spatial, working memory, and reading ability, may contribute to math performance and toward establishing subtypes of MLD (Geary, 1993, 2004). Thus, the present study examined these related skills in the two syndrome groups relative to their respective comparison groups.

Visual–Spatial Ability

Overall, the present results are consistent with our previous findings suggesting specific rather than global visual–spatial deficits in fragile X syndrome and selective, albeit more widespread, deficits in Turner syndrome (Mazzocco et al., 2006). For example, relative to their peers, girls with fragile X syndrome had difficulty recalling location and spatial arrangement on our nonstandardized Figure Ground Memory task. In contrast, girls with Turner syndrome had lower performance relative to peers on the VMI, took longer to complete the standardized Figure Ground subtest, and had lower scores on that subtest (albeit still within the average range). Relative to their peers, girls with Turner syndrome also recalled fewer locations than their comparison group on the Figure Ground Memory measure, despite a longer exposure to the array during the standardized Figure Ground subtest.

For both syndrome groups, the results of the Figure Ground Memory task are consistent with those from Grade 3 (Mazzocco et al., 2006), despite a higher IQ range and older sample age in the present study. Specifically, 10-year-old girls with fragile X syndrome continue to be distinguished from their comparison group based on their production of non–grid-like arrangements. In contrast, girls with Turner syndrome continue to take more time on the standardized Figure Ground task, thereby gaining more exposure to the shape arrangement, but ultimately recall fewer locations. Girls with fragile X syndrome also recalled fewer locations than their comparison group in the present study. Although this difference was not statistically significant in Grade 3, the performance improvement in the present study relative to Grade 3 was small among girls with fragile X (M = 4.82 vs. 4.20 for Grades 5 and 3, respectively), especially relative to the comparison groups (M = 7.59 vs. 5.94 for Grades 5 and 3, respectively), suggesting few age-appropriate gains in visual–spatial memory. Thus, although visual–spatial deficits appear to persist into late elementary school for both girls with fragile X and Turner syndrome, these findings are consistent with the notion that the nature of the deficits distinguishes the two syndromes. Although conclusions regarding the persistence of deficits must be considered preliminary because of the different samples included in the two studies, the majority of the girls in the fragile X (11 of 14 participants) and Turner syndrome (16 of 17 participants) groups were included in the previous study.

We had further hypothesized that one potential source of difficulty between the two syndrome groups was inefficient visual scanning in girls with Turner syndrome. This hypothesis was tested by comparing performance on the standardized Position in Space subtest and the PPS task. Overall, the data failed to support this hypothesis. If lack of scanning efficiency affected performance on the standardized Position in Space task, performance accuracy should have improved to a greater degree for girls with Turner syndrome. However, reducing the visual scanning demands of the standardized Position in Space task improved performance across all groups and to similar degrees. A confound to this assessment was the ceiling effect present for both the standard and paired Position in Space tasks for all groups. This ceiling effect was evident in the comparison group, which was expected, and in the Turner syndrome group, which was not anticipated, because of the difficulty with the task noted in Grade 3 (Mazzocco et al., 2006). Regardless, even when scanning demands were minimized using the PPS task, girls with Turner syndrome still took longer to complete the task than did their peers. Although this response time difference was not statistically significant, the effect size was moderate (Cohen's d = 0.64).

Furthermore, it must be noted that the mean response time was comparable for the two syndrome groups, despite the higher FSIQ scores for girls with Turner syndrome relative to girls with fragile X syndrome. Response times for the two comparison groups differed more than did the response times for the syndrome groups, but this difference could not be evaluated statistically because the two comparison samples were not mutually exclusive. When the comparison group was limited to girls who were IQ-matched with the Turner syndrome group (see Table 3), response times were 20 s faster than when the comparison group was limited to girls matched with the fragile X group (see Table 2)—perhaps because the former group had a higher mean IQ score than did the latter. Whereas higher IQ scores were associated with faster response times on the PPS for the comparison group, the response times were comparable for the two syndrome groups (even slightly slower for the Turner syndrome group) despite the higher IQ scores in the Turner syndrome group. Thus, the girls in this group did not demonstrate the IQ advantage evident for the comparison group.

In our earlier work, we found it important to consider both whether girls from either syndrome group differed from age- and FSIQ-matched peers and whether their performance characteristics were consistent with children with MLD who did not have any known syndrome (see Murphy et al., 2006). For the PPS task, the lack of differences between the two syndrome groups and their respective comparison groups distinguishes these syndromes from children with MLD from the general population (unpublished data). In contrast to girls with fragile X or Turner syndrome, we have found that diminishing the visual scanning demands distinguishes children with versus without MLD. Specifically, the improvement in accuracy was slightly greater for the children with MLD relative to those without MLD. The significantly low score on the standardized Position in Space test for fifth graders with versus without MLD (unpublished data) drove this effect—a finding that failed to emerge for either syndrome group in the present study. Also, response times for children with MLD vary with IQ, just as is the case for the general population (unpublished data).

Verbal Memory and Working Memory

The findings in the present study are consistent with previous findings suggesting difficulty with aspects of executive function among girls with fragile X syndrome (Bennetto et al., 2001; Kirk et al., 2005; Mazzocco et al., 1993) and Turner syndrome (Buchanan et al., 1998; Kirk et al., 2005; Temple et al., 1996) and support the notion that such deficits continue into late elementary school. Although executive function difficulties were found in both syndrome groups, the profile of difficulty across the executive function–working memory tasks distinguished the two groups. Girls with fragile X syndrome were characterized by lower efficiency than their matched comparison group on the most demanding subtest of the CNT but did not differ from their comparison group on verbal memory or digit recall. In contrast, girls with Turner syndrome had more difficulty than their comparison group on backward digit recall, suggesting some working memory impairment, but no differences were found on the CNT, forward digit recall, or verbal memory measure.

Although these findings are partially consistent with results from earlier studies, the profiles of CNT scores differed from those reported for third graders with fragile X or Turner syndrome (Kirk et al., 2005). Relative to their peers, girls in Grade 5 with Turner syndrome had less difficulty on the CNT at Grade 5 than at Grade 3; whereas girls with fragile X syndrome had difficulty at both grades. Despite reported correlates of IQ on executive functions, the discrepant set of findings between Grades 3 and 5 is not likely to solely reflect differences in the FSIQ range of participants between the two separate studies. In the present study, the mean FSIQ was slightly higher than that in our previous study, for both the Turner syndrome (mean IQ = 108 vs. 104, respectively) and fragile X syndrome groups (mean IQ = 95 vs. 88, respectively). For both groups, the higher mean FSIQ resulted from a higher threshold at the lower end of the IQ score range (80 vs. 73 in the fragile X group, and 95 vs. 84 in the Turner syndrome group, present vs. past study). Nevertheless, among girls with fragile X syndrome, a significant difference on the CNT efficiency emerged in Grade 5, but not in Grade 3, despite the higher FSIQ range in the present sample. This is consistent with the notion that FSIQ alone is insufficient to account for the deficits in executive function and working memory observed on the CNT (Kirk et al., 2005).

For girls with Turner syndrome, differences apparent at Grade 3 were not evident at Grade 5, which may implicate the role of higher IQ scores in the Grade 5 sample. However, when we reanalyzed the third-grade CNT data including only those 15 girls with Turner syndrome who had also participated in the Grade 3 study, the results continued to demonstrate significantly lower efficiency on the two-attribute task among girls in Grade 3 with Turner syndrome relative to their peers. Thus, the higher FSIQ alone does not account for the lack of group differences during Grade 5.

Instead, we believe that the current and previous (Kirk et al., 2005) findings from the CNT have implications for the development of working memory in fragile X and Turner syndrome. Kirk et al. (2005) reported no difference in efficiency between Grade 3 girls with fragile X and an FSIQ-matched comparison group for either the one- or two-attribute rules; but as working memory demands increased, girls with fragile X syndrome made more errors than their comparison group. By Grade 5, performance efficiency is significantly lower in girls with fragile X but only on the more difficult, two-attribute task. This difference may emerge at Grade 5 because the age-appropriate improvement on this task (Mazzocco & Kover, 2007) was observed only in the comparison group, and not among the fragile X group. Specifically, among typically developing girls in Grade 5, performance efficiency on the two-attribute task typically parallels Grade 3 performance on the one-attribute task. Yet girls with fragile X syndrome were less efficient on the two-attribute task at Grade 5 (M = 0.50, SD = 0.36) relative to their performance on the one-attribute task in Grade 3 (M = 0.90, SD = 0.70). Of note is that at both Grades 3 and 5, less accurate performance rather than slower processing speed characterized the performance of girls with fragile X syndrome overall.

Meanwhile, at Grade 3, girls with Turner syndrome were less efficient than an FSIQ-matched comparison group at both the one- and two-attribute rules and traded accuracy for speed when applying the more complex rule. Among girls with Turner syndrome, efficiency on the two-attribute task at Grade 5 (M = 1.14, SD = 0.55) was fairly consistent with efficiency on the one-attribute task in Grade 3 (M = 1.08, SD = 0.60); this finding parallels the normative data for CNT test performance (Mazzocco & Kover, 2007). Thus, diminished group differences by late elementary school may suggest that girls with Turner syndrome achieve the minimum level of proficiency of their peers on both components of the task and reach the ceiling effects anticipated for their age. It remains to be seen whether group differences would have emerged if additional working memory demands had been added to the CNT, which we did not explore in the present study, and whether rates of continued improvement in efficiency over time will continue to parallel what is observed in the general population.

Reading-Related Skills

Consistent with previous reports (e.g., Mazzocco, 2001; Temple & Carney, 1996), reading appears to be a relative strength compared to math performance among girls with fragile X or Turner syndrome. Although differences were found between both syndrome groups and their respective comparison groups on decoding skills, it is important to note that both syndrome groups had standard scores in the average range for their age ($M = 91.64$ and $M = 101.12$ for fragile X and Turner syndrome, respectively). Moreover, no differences were found for reading fluency, further supporting the notion that reading is a relative strength for both syndromes. Differences were observed for both syndrome groups on RAN Colors but not Numbers or Letters.

In the general population, by third grade, the processing demands associated with naming numbers may be less than those associated with color naming (Stringer, Toplak, & Stanovich, 2004). We examined the difference between RAN Colors and Numbers to assess the extent to which processing demands contribute to performance response time at Grade 5, when reading should be automatic for both syndromes. Thus, if reading is automatic, we would expect a shorter response time on the Numbers than on the Colors task. However, if performance is characterized by global processing speed deficits, no discrepancy should emerge between the Colors and Numbers tasks. Alternatively, as we predict may be the case in Turner syndrome, if processing difficulty were selective, then we would predict a greater disparity between Colors and Numbers tasks than observed in the general population. Indeed, the Colors–Numbers difference was greater relative to their comparison group among girls with Turner syndrome, but not among girls with fragile X syndrome. This pattern of results suggests that response time increases as processing demands increase among girls with Turner syndrome. Although this finding supports the notion of processing speed deficits in Turner syndrome, our findings from the CNT indicate that not all aspects of processing are slowed. Thus, additional support is provided for the notion of selective rather than global processing deficits in Turner syndrome (Temple, 2002).

Correlations Between Math and Related Skills

In addition to performance profiles, correlations may inform us about potential pathways underlying cognitive deficits. For this reason, we examined correlations between WJ-R Calculations performance and select measures within each domain of related skills. Previous studies with younger children (Mazzocco, 1998; Mazzocco et al., 2006) have consistently demonstrated a larger number of such correlations among girls with fragile X relative to girls with Turner syndrome. Our findings are consistent with these earlier reports. For girls with Turner syndrome, no correlations were significant, whereas multiple significant correlations were observed for girls with fragile X syndrome (see Table 4).

Among girls with fragile X syndrome, correlations were significant with both parts of our executive function measure, only one measure of spatial reasoning, and all reading-related skills that we examined. These findings could not be associated with the lower FSIQ range for fragile X versus Turner syndrome groups, because no significant correlations emerged for the fragile X comparison group. However, significant correlations emerged for girls in the Turner syndrome comparison group, including some that had also been observed in the fragile X group (e.g., all of the reading measures and verbal memory) but not others (e.g., VMI). Our objective in examining models of MLD—specifically, syndrome groups with associated difficulties in mathematics—is to determine the different cognitive pathways leading to poor math achievement. Although these group differences in correlation patterns do not provide definitive proof of what those potential pathways may be, they support the notion that different cognitive correlates serve as potential underpinnings of poor mathematics performance and achievement.

Limitations and Future Directions

The present findings suggest that poor math performance among girls with fragile X or Turner syndrome persists into the late elementary school years. Although the conclusions of the present study are limited by the cross-sectional nature of the study design, the majority of the study participants with fragile X (64% to 79%) or Turner syndrome (82% to 94%) had been included in our earlier reports on this cohort at Grade 3 (Kirk et al., 2005; Mazzocco et al., 2006), and about half in each group were included in the initial report from kindergarten (Mazzocco, 2001). Future studies that assess performance longitudinally will contribute toward assessing the trajectory of math and related skill development.

Furthermore, the variation in the IQ range between girls with fragile X and Turner syndrome precluded directly matching the two syndrome groups. Rather, we evaluated the performance of each group relative to an IQ-matched comparison group, as we have done in our earlier reports (Kirk et al., 2005; Mazzocco, 2001; Mazzocco et al., 2006; Murphy et al., 2006). Such an approach allowed us to address the questions of interest while controlling for differences in overall cognitive ability. However, future studies aimed at addressing the question of whether girls with fragile X or Turner syndrome are distinguished from each other on the measures assessed will further contribute to the elucidation of the syndrome specificity of the observed deficits.

Many contextual factors may further contribute to mathematics performance outcome, including school curriculum, quality of instruction, and exposure to special education. In the present study, most of the children in the syndrome groups attended schools in different school districts across North America, which likely led to variability in these factors across individuals. It was beyond the scope of the present study to address the contribution of these factors to math performance. However, it is noteworthy that despite these sources of variability, differences did emerge between both syndrome groups. Future studies including these contextual factors may contribute toward understanding the influences on math performance among individuals with these syndromes and the degree to which such factors mitigate performance outcomes in children with MLD.

Conclusions

Regardless of its limitations, the present study contributes to the existing literature by documenting the difficulties of girls with fragile X or Turner syndrome in mathematics performance during late elementary school. Moreover, the findings suggest that the nature of poor math performance and the profile of math-related skills distinguish these two syndromes from each other and from children in the general population, as does the association between math and related skills. Together, these findings demonstrate how different cognitive profiles may lead to poor mathematics performance and further serve to delineate potential variation in the cognitive profiles of children with MLD.

Appendix

The prevalence of fragile X and Turner syndromes is approximately 1 in 4,000 to 1 in 9,000 live births, and 1 in 2,000 to 1 in 5,000 live births, respectively (see Hagerman, 2002, and Rovet, 2004, for additional information about these syndromes). Although the range of cognitive impairments in fragile X is broader than in Turner syndrome and includes mental retardation, about 50% of female individuals with fragile X syndrome may present with learning disabilities or be unaffected by the syndrome. The lower incidence of mental retardation among female individuals with fragile X syndrome is in contrast to male individuals, almost all of whom meet criteria for mental retardation (Bailey, Hatton, & Skinner, 1998). The present study focuses on individuals without mental retardation, which allows the exploration of the subtle phenotypic characteristics associated with the syndrome.

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