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Supporting students with sex chromosome aneuploidies in educational settings: Results of a nationwide survey

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ABSTRACT

Children with sex chromosome aneuploidies (SCAs) are at an increased risk for neurocognitive and behavioral disorders that may interfere with academic success, including early developmental delays, learning disabilities, executive function problems, and social communication deficits. The present national survey aimed to update and extend our understanding of school supports and educational outcomes for students with these increasingly common genetic diagnoses. Parents of children with a diagnosed SCA, birth to 21 years, living in the United States ($N = 248$), responded to an electronic survey with questions focused on school support plans, academic accommodations, educational therapies, school completion, and perceptions of educator awareness of SCAs. Results revealed high rates of delayed kindergarten, grade retention in primary years, and educational support plans (IEPs = 71%; Section 504 Plans = 26%). A majority (73%) of respondents with children over age 18 years ($N = 41$) reported their children successfully completed high school, and nearly half (46%) pursued post-secondary education opportunities. Many parents reported their children's educators had little to no knowledge of SCA conditions, justifying a need to train teachers and policy makers in the unique educational needs of children and adolescents with SCAs. School psychologists should be aware of the frequent need for accommodations and individualized support plans in this population so they can support children and families by advocating for early and comprehensive evaluations and intervention plans.

1. Introduction

Sex chromosome aneuploidies (SCAs) are the most common chromosomal abnormalities with a collective prevalence rate of 1 in 500 live births (Jacobs et al., 1974). Previous research has described neurocognitive and behavioral phenotypes with developmental risk factors that may interfere with academic success, including early developmental delays, increased risk for learning disabilities, executive function problems, attention deficit hyperactivity disorder (ADHD), and social communication deficits (Cordeiro et al.,

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2012; Pennington et al., 1982; Tartaglia et al., 2012; Urbanus et al., 2020). Prior research on educational outcomes and school supports for students with SCAs in the U.S. is outdated, and current studies have been limited to Northern Europe. As educational systems vary greatly by country and over time, it is critical to update our understanding of U.S. supports for students with SCAs.

Understanding educational factors for students with SCAs is imperative as earlier diagnosis means many more children are recognized to have these conditions. Historically, <10% of those born with SCAs were diagnosed during childhood due to a lack of physical anomalies or other features that might trigger genetic testing (Abramsky & Chapple, 1997). Updated standard of care to offer noninvasive prenatal screening (NIPS) for all pregnancies is alerting more and more families to the presence of SCAs prior to birth or shortly after (Bevilacqua et al., 2018; Kornman et al., 2018; American College of Obstetricians and Gynecologists' Committee on Practice Bulletins, 2020). With a sharp increase in NIPS in routine clinical practice, schools will also see a dramatic rise in the number of students entering school with a prenatally identified SCA and either a known risk for future learning disabilities or an established history of developmental delays and deficits in pre-academic and/or cognitive skills. There is urgency and great need to improve our understanding of how learning disabilities manifest in this growing student population and to provide guidance to school systems and educators about how to support this unique, genetically-defined student population.

1.1. Sex chromosome aneuploidies

SCAs occur when a child is born with an atypical number of sex chromosomes, also known as X and Y chromosomes. Males typically have one X and one Y chromosome (46,XY), whereas females are typically born with two X chromosomes (46,XX). The most common SCAs are trisomy conditions, in which a male is born with an extra X or Y chromosome (47,XXY/Klinefelter syndrome [KS] and 47, XYY) or a female is born with an extra X chromosome (47,XXX/Trisomy X). However, there are also instances when a child is born with only one X chromosome (45,XO/Turner syndrome), or additional sex chromosomes (e.g., 48,XXYY, 48,XXXY, 49,XXXXY, etc.). Mosaic SCA occurs when cells are found to have multiple chromosome differences in an individual, and in cases of sex chromosomes, may result in a milder presentation of symptoms. Table 1 lists prevalence rates for some of the most common sex chromosome conditions (Boyd et al., 2011).

Previously published research has documented a variable range of physical and medical features associated with SCAs, with more significant features in children with tetra/pentasomy than trisomy SCAs. Trisomy SCAs commonly lack distinct physical or facial dysmorphology as seen in other chromosomal trisomies (e.g., Down syndrome). However, tall stature is common among most SCAs characterized by extra X or Y chromosomes, and children with SCAs may also present with widely spaced eyes (hypertelorism), epicanthal folds, a curved fifth finger (clinodactyly), low muscle tone (hypotonia), and hormone dysfunction (Davis et al., 2019; Wigby et al., 2016). Due to these subtle physical features, when learning disabilities and behavioral features are present, providers do not always consider the possibility of an underlying genetic diagnosis.

1.2. Implications for school

Previously published research in SCAs has documented several educationally significant learning problems; however, frequency and severity differ by genetic condition. Additionally, there is great individual variability in developmental and psychological outcomes for children with SCAs, reflecting the interplay of genetics and environment (Samango-Sprouse et al., 2013; van Rijn et al., 2018). Some children show no signs of developmental differences, whereas others will have significant impairments and require a great deal of educational support. Furthermore, methodological limitations in research (e.g., small sample sizes, ascertainment bias) have precluded researchers from understanding the exact relationships between the additional sex chromosomes and neurodevelopmental pathways. Nevertheless, there is a clear neurocognitive and behavioral phenotype for students with SCAs that is continuing to be defined.

Both historic and more recent natural history studies support that the majority of children born with an extra sex chromosome will experience developmental delays in early childhood, including early speech-language or motor delays (Bender et al., 1983; Robinson et al., 1982; Tartaglia et al., 2020). Reduced expressive language and speech sound disorders (e.g., apraxia, dyspraxia) are the most reported communication deficits, although receptive language may also be compromised for many young children with SCAs (Walzer et al., 1990, 1982). Low muscle tone and poor sensorimotor coordination can also present, leading to lagging motor skills, including delayed walking (Martin et al., 2019; Salbenblatt et al., 1987, 1989). A recent nationwide parental survey indicated that a significant portion of young children with SCAs living in the U.S. receive early intervention therapies and/or early childhood special education preschool services prior to entering kindergarten (Thompson et al., 2020).

Table 1
Common SCA conditions and prevalence rates.

Condition/Other Names	Prevalence
47,XXY, Klinefelter syndrome (KS)	1:600 live male births
47,XYY, Jacob syndrome	1:1000 live male births
47,XXX, Trisomy X syndrome	1:1000 live female births
48,XXYY	1:17,000 live male births
48,XXXY	1:18,000 live male births
49,XXXX	1:25,000 live female births
49,XXXXX	1:50,000 live female births

Older children and adolescents with SCAs are at an increased risk for several educationally significant problems and often require speech-language and motor therapies in addition to academic supports. Although general cognitive abilities tend to fall within the average range, language-based learning disabilities (e.g., dyslexia) are common across the SCA trisomies (Boada et al., 2009; Leggett et al., 2010; Simpson et al., 2014; Skakkebaek et al., 2014). Children with tetra/pentasomy SCAs often have more diminished cognitive ability, with 30%–50% having a diagnosis of mild intellectual disability (Tartaglia et al., 2011). The presence of an extra sex chromosome also greatly increases the risk of executive function disorders; for instance, children with SCAs have high rates of ADHD (Tartaglia et al., 2012). Additionally, social-emotional and behavioral problems are associated with the phenotype, including mental health problems (e.g., anxiety, depression), as well as increased rates of autism spectrum disorder (ASD; Tartaglia et al., 2017; Urbanus et al., 2020).

1.3. Educational outcomes

With a documented constellation of early delays and learning deficits, we would hypothesize that children with SCAs require significant educational supports. Epidemiological studies from northern Europe have shown significantly reduced educational achievement (lack of college degree) for adults with trisomy SCA (Berglund et al., 2020). Skakkebaek et al. (2014) documented high rates (> 50%) of self-reported history of dyslexia and special education in a sample of 73 Danish men with 47,XXY and educationally matched controls. Data from the United States are limited in this area, as studies on the educational needs and outcomes of U.S. students with SCAs were conducted in the 1990s and early 2000s, prior to increased rates of prenatal diagnosis (Chen et al., 2019) and before some major shifts in U.S. special education policies (e.g., early intervention and preschool special education, inclusion practices, SLD eligibility criteria, grade retention policies; Gargiulo & Bouck, 2020). The information we do have suggests a pattern of educational struggles and a need for individualized educational supports.

Results from prenatal and newborn screening studies from the 1970's and 1980's that prospectively followed children with SCAs throughout their childhood and adolescence have documented higher levels of academic need. Linden and Bender (2002) studied a cohort of 51 children and adolescents with a prenatal diagnosis of SCA. Results from parent questionnaires indicated that over half of the children enrolled in the study required academic interventions and that the majority had less academic proficiency than their siblings. Bender et al. (1993) examined the neuropsychological profiles of 42 adolescents diagnosed with SCAs at birth. They found a wide range of academic performance; although almost 20% of the sample required intensive special education services, another 20% of the sample graduated high school and attended college. However, regardless of educational outcome, the majority demonstrated impairment in one or more specific cognitive processes on standardized neurodevelopmental assessments. Rovet et al. (1996) followed 36 boys with 47,XXY/KS throughout their academic careers from age 6 to 20 years. Results from their longitudinal study indicated boys with 47,XXY/KS were significantly more likely to fail one or more grades in school and were also more likely to qualify for special education than same-aged controls.

Currently, studies that systematically document educational outcomes and supports for U.S. students with SCAs are missing from the literature. It can be challenging for schools and parents to develop effective educational plans for a child diagnosed with SCA when current data and guidance are not available. This study aimed to fill this gap with a national survey of parents regarding educational implications of SCAs. The purpose of this study was to quantify and characterize current educational outcomes and school supports in the U.S., as well as educator awareness of SCAs, as reported by parents. Results from the survey highlighted several specific areas of unmet need to target for future research and intervention trials to improve efficacy in educating children with SCAs.

2. Method

2.1. Participant recruitment and inclusion criteria

Recruitment of parents of children with SCAs took place through national advocacy organizations and local agencies, including the eXtraordinarY Kids Clinic at Children's Hospital Colorado, the Association for X & Y Chromosome Variations (AXYS), and social media platforms approved by these organizations. The protocol and plans for human subjects' protections were approved by the Colorado Multiple Institution Review Board (#19–0555) prior to recruitment of all study participants. All participants had the opportunity to review the consent form and study procedures and talk directly with investigators prior to agreeing to participate in the study. Participants were included in the broader survey if they were the parent/caregiver of a child with a diagnosis of SCA aged birth to 21 years. All participants were required to be able to read in English in order to access the survey content. The current analysis includes parents of children in the U.S., ages 5–21 years, who had entered and/or recently completed Grades Kindergarten through 12 (K−12) in the U.S. school system. Analyses were restricted to those living in the United States given the significant differences in educational policies and support systems between countries.

2.2. Data collection

The survey was designed in REDCap data management system by a team with expertise in SCAs, learning, and educational systems and piloted with a small subsample of professionals and parents to determine face validity and ease of comprehension. Branching logic assured questions were appropriate to the person responding to the survey (parent of a school-aged child or post-secondary aged child). Questions were a mixture of true/false, multiple choice, and short answer formats about clinical and educational diagnoses, qualification for individual education programs (i.e., IEPs/special education), Section 504 Plans (i.e., individualized accommodation plans

for students with disabilities, but without specialized instruction), grade retention, accommodations, service providers, service minutes, least restrictive environment (LRE; i.e., extent child is educated alongside peers without disabilities), and parent perceptions of their child's educational experiences.

Table 2
Study sample.

	N (%)
N	248
Age (years)	
Child	13.3, SD = 4.5, range = 5–21
Respondent	47.7, SD = 8.1, range = 28–69
Grade Level	
Elementary–Primary (Grades K–2)	37 (14.9)
Elementary–Intermediate (Grades 3–5)	43 (17.3)
Middle School (6–8)	53 (21.4)
High School (9–12)	74 (29.9)
Post-secondary	41 (16.5)
SCA Condition	
Trisomy SCA	197 (79.4)
47,XXX	35 (14.1)
47,XXY	133 (53.6)
47,XYY	29 (11.7)
Tetra/Pentasomy SCA	51 (20.6)
48,XXYY	33 (13.3)
48,XXXY	16 (6.5)
49,XXXX	1 (0.4)
49,XXXXX	1 (0.4)
Child Gender	
Male	210 (84.7)
Female	37 (14.9)
Other	1 (0.4)
Caregiver Respondent	
Mother	228 (91.9)
Father	20 (8.1)
Responding Parent's Education	
Some High school	3 (1.2)
High school diploma	13 (5.2)
Some college	29 (11.7)
Associates degree	24 (9.7)
Bachelor's degree	90 (36.3)
Advanced degree (Master's, PhD)	89 (35.9)
Timing of SCA Diagnosis	
Prenatal	96 (38.7)
Postnatal	152 (61.3)
Additional clinical diagnoses	
ADHD	137 (55.2)
Anxiety disorder	98 (39.5)
Autism spectrum disorder	59 (23.8)
Learning Disorder–Reading	90 (36.3)
Learning Disorder–Math	55 (22.2)
Learning Disorder–Other	41 (16.5)
Major Depression	40 (16.1)
Sensory Processing Disorder	52 (21)
Speech language disorder	100 (40.3)
Type of School[†]	
Public	149 (72)
Private	31 (15)
Homeschool	18 (8.7)
Other	9 (4.3)
US Region[‡]	
Northeast	48 (19.4)
Midwest	45 (18.1)
South	91 (36.7)
West	64 (25.8)

[†] Only includes school-aged children ($N = 207$).

[‡] US regions as designated by the US Census Bureau. Northeast: CT, MA, ME, NH, NJ, NY, PA, RI, VT; Midwest: IA, IL, IN, KS, MI, MN, MO, NE, ND, OH, SD, WI; South: AL, AR, DC, DE, FL, GA, KY, LA, MD, MS, NC, OK, SC, TN, TX, VA, WV; West: AK, AZ, CA, CO, HI, ID, MT, NM, NV, OR, UT, WA, WY.

Table 3
School supports and educational outcomes for students with SCAs.

	N (%)	Pre-natal	Post-natal	*p*	Trisomy	Tetra/Penta	*p*	XXX	XXY	XYY	*p*
Delayed Kindergarten	47 (19.0)	22 (22.9)	25 (16.4)	0.234	37 (18.8)	10 (19.6)	0.234	5 (14.3)	25 (18.8)	7 (24.1)	0.342
Repeated Grade(s)	42 (16.9)	16 (16.7)	26 (17.1)	0.956	29 (14.7)	13 (25.5)	0.039*	3 (8.6)	21 (15.8)	5 (17.2)	0.539
Any Individualized Support Plan	213 (85.9)	81 (84.4)	132 (86.8)	0.469	167 (84.8)	46 (90.2)	0.154	30 (85.7)	108 (81.2)	29 (100)	0.310
504 Plan	64 (25.8)	29 (30.2)	35 (23)	0.257	60 (30.5)	4 (7.8)	0.004*	13 (37.1)	38 (28.6)	9 (31.0)	0.670
IEP	176 (71)	63 (65.6)	113 (74.3)	0.115	130 (66)	46 (90.2)	0.001*	19 (54.3)	86 (64.7)	25 (86.2)	0.039**
Educational Dx†				0.031*			0.001*				0.216
Other Health Impaired	63 (35.8)	22 (34.9)	41 (36.3)	0.296	44 (33.8)	19 (41.3)	0.207	7 (36.8)	31 (36.0)	6 (24.0)	0.190
Specific Learning Disability	38 (21.6)	15 (23.8)	23 (20.4)	0.363	30 (23.1)	8 (17.4)	0.280	6 (31.6)	19 (22.1)	5 (20.0)	0.646
Autism	17 (9.7)	4 (6.3)	13 (11.5)	0.202	15 (11.5)	2 (4.3)	0.127	1 (5.3)	9 (10.5)	5 (20.0)	0.298
Speech Language Impairment	16 (9.1)	7 (11.1)	9 (8)	0.330	13 (10.0)	3 (6.5)	0.356	2 (10.5)	11 (12.8)	0 (0)	0.133
Intellectual Disability	10 (5.7)	1 (1.6)	9 (8)	0.072	4 (3.1)	6 (13.0)	0.021*	0 (0)	2 (2.3)	2 (8.0)	0.264
Multiple disabilities	9 (5.1)	5 (7.9)	4 (3.5)	0.180	7 (5.4)	2 (4.3)	0.568	0 (0)	5 (5.8)	2 (8.0)	0.618
Developmental delay	9 (5.1)	3 (4.8)	6 (5.3)	0.590	7 (5.4)	2 (4.3)	0.568	1 (5.3)	3 (3.5)	3 (12.0)	0.145
Hearing impaired	2 (1.1)	0 (0)	2 (1.8)	0.411	2 (1.5)	0 (0)	0.544	0 (0)	2 (2.3)	0 (0)	1.00
Other	7 (4)	1 (1.6)	6 (5.3)	0.215	3 (2.3)	4 (8.7)	0.077	0 (0)	3 (3.5)	0 (0)	1.00
Don't know	5 (2.8)	5 (7.9)	0 (0)	0.005*	5 (3.8)	0 (0)	0.215	2 (10.5)	1 (1.2)	2 (8.0)	0.057
Educational Services‡											
Academic supports	113 (77.9)	8 (76.0)	75 (78.9)	0.833	80 (76.9)	33 (80.5)	0.667	11 (73.3)	52 (76.5)	17 (81.0)	0.693
Speech therapy	87 (60.0)	23 (46.0)	64 (67.4)	0.020*	54 (51.9)	33 (80.5)	0.002*	5 (33.3)	37 (54.4)	12 (57.1)	0.309
Mental health/behavior	41 (28.3)	17 (34.0)	24 (25.3)	0.332	33 (31.7)	8 (19.5)	0.157	2 (13.3)	21 (30.9)	10 (47.6)	0.095
Social skills	53 (36.6)	20 (40.0)	33 (34.7)	0.588	41 (39.4)	12 (29.3)	0.339	6 (40.0)	28 (41.2)	7 (33.3)	0.799
Occupational Tx	49 (33.8)	10 (20.0)	39 (41.1)	0.016*	30 (28.8)	19 (46.3)	0.053	3 (20.0)	22 (32.4)	5 (23.8)	0.566
Physical Tx	14 (9.7)	3 (6.0)	11 (11.6)	0.381	10 (9.6)	4 (9.8)	1.00	0 (0)	9 (13.2)	1 (4.8)	0.227
IEP Inclusion‡				1.00			0.810				0.391
General education classroom with supports	121 (83.4)	42 (84.0)	79 (84.0)		86 (83.5)	35 (85.4)		14 (93.3)	56 (83.6)	16 (76.2)	
Self-contained classroom	23 (15.8)	8 (16.0)	15 (16.0)		17 (16.5)	6 (14.6)		1 (6.7)	11 (16.4)	5 (23.8)	
High School Completion§				0.164			0.056				0.321
HS Diploma	30 (73.2)	11 (68.8)	19 (76.0)		28 (77.8)	2 (40.0)		6 (100)	20 (76.9)	2 (50.0)	
GED	5 (12.2)	4 (25.0)	2 (8.0)		5 (13.9)	0 (0)		0 (0)	4 (15.4)	1 (25.0)	
HS Certificate	2 (4.9)	0 (0)	2 (8.0)		1 (2.8)	1 (20.0)		0 (0)	0 (0)	1 (25.0)	
Transition program	2 (4.9)	0 (0)	2 (8.0)		1 (2.8)	1 (20.0)		0 (0)	1 (3.8)	0 (0)	
Dropped out	0 (0)	0 (0)	0 (0)		0 (0)	0 (0)		0 (0)	0 (0)	0 (0)	
Other	2 (4.9)	1 (6.3)	1 (4.0)		1 (2.8)	1 (20.0)		0 (0)	1 (3.8)	0 (0)	

†These questions were only asked of respondents who reported their child had an IEP during their K–12 years ($n = 176$; for trisomy comparisons $n = 130$).
‡These questions were only asked of respondents with currently enrolled school-aged children (K–12) with IEPs ($n = 145$; for trisomy comparisons $n = 104$).
§These questions were only asked of respondents ages 18–21 years ($n = 41$; prenatal $n = 16$, postnatal $n = 25$; trisomy $n = 36$, tetra/pentasomy $n = 5$; XXX $n = 6$, XXY $n = 26$, XYY $n = 4$).
GED = General Education Development (high school equivalency).
*$p < .05$.
**Post hoc comparison results for group differences in IEPs by trisomy SCA condition: XXX versus XXY, $p = .501$; XXX versus XYY, $p = .011$*; XXY versus XYY, $p = .042$*

2.3. Data analysis

All data were downloaded from REDCap and imported into SPSS statistical software for analysis. Descriptive statistics (frequencies and percentages) were used to characterize educational outcomes and supports in students with SCAs. Logistic regression models were used to examine potential group differences for timing of diagnosis (prenatal vs. postnatal), and type of SCA (trisomy vs. tetra/penta SCA) for delayed kindergarten, grade retention, Section 504 plans, and IEPs. Analyses controlled for potential effects of parent education (college degree or no college degree) and child age. Exploratory sub-group analysis by sex (XXX vs. XXY + XYY) did not reveal any significant differences ($p > .05$ for all). Pearson's chi-square and Fisher's exact tests were used to examine group differences in the subset of students with IEPs regarding qualifying educational diagnoses and school-based interventions and to examine any significant ($p < .05$) differences in post hoc comparisons. No adjustments were made for multiple comparisons as the study was exploratory and analyses were meant to be descriptive and hypothesis generating. One-sample binomial tests were used to compare sample proportions to the general U.S. student population. Significance was set at $p < .05$. Survey branching logic was based on participant characteristics (e.g., child age group, types of supports) meant that not all participants were asked all questions, and therefore denominators for proportions differed: (a) delayed kindergarten, grade retention, Section 504 Plans, and IEPs included the entire sample ($N = 248$); (b) classroom accommodations included students who had any type of school support plan ($n = 213$); (c) school interventions included only those currently enrolled in the K–12 system with IEPs ($n = 145$); (d) educational outcomes included post-secondary only ($n = 41$); and (e) parent perceptions included students currently enrolled in the K–12 system ($n = 207$). All analyses were also conducted separately for the sample of postsecondary students ($n = 41$) to examine supports within a sample of students with SCAs who had completed their K–12 education. However, no differences were identified when compared with the larger sample, and thus all results reported here include the entire sample, ages 5–21 years.

3. Results

3.1. Sample

A total of 550 respondents consented to participate and 505 (92%) completed the survey. Of those who completed the survey, 248 survey respondents lived in the United States and were caregiver to a child with SCA between the ages of 5–21 years who was in, or had recently exited, Grades K–12 in the U.S. school system (Table 2). Results for children under the age of 5 years are reported elsewhere (Thompson et al., 2020). Additional clinical diagnoses were common, including particularly high rates of ADHD, anxiety, and learning disorders (LD), most notably in reading. Although 20% of parents reported that their child had a math LD, most had combined reading and math LD, and only 12 parents (5% of the total sample) endorsed math LD only. Participants were diverse in terms of geographic region, grade level of child, other clinical diagnoses, and timing of the SCA diagnosis (pre vs. postnatal). The sample that respondents were reporting on included a range of SCA conditions, with the majority having XXY (54%) and none with Turner syndrome. Children ranged in age from 5 to 21 years with a mean age of 13 years (SD = 4.5). Most respondents were mothers (92%) and the majority of children (72%) attended public schools. Respondents were significantly more likely to have a college degree (72%) than the general U.S. population (37.5%; $p < .01$; National Center for Education Statistics, 2020).

3.2. Delayed kindergarten

Nearly 20% ($n = 47$) of respondents reported delaying their child's initiation of kindergarten by one or more years. There were no significant differences by timing of diagnosis, trisomy versus tetra/pentasomy SCA condition, or specific trisomy SCA karyotype

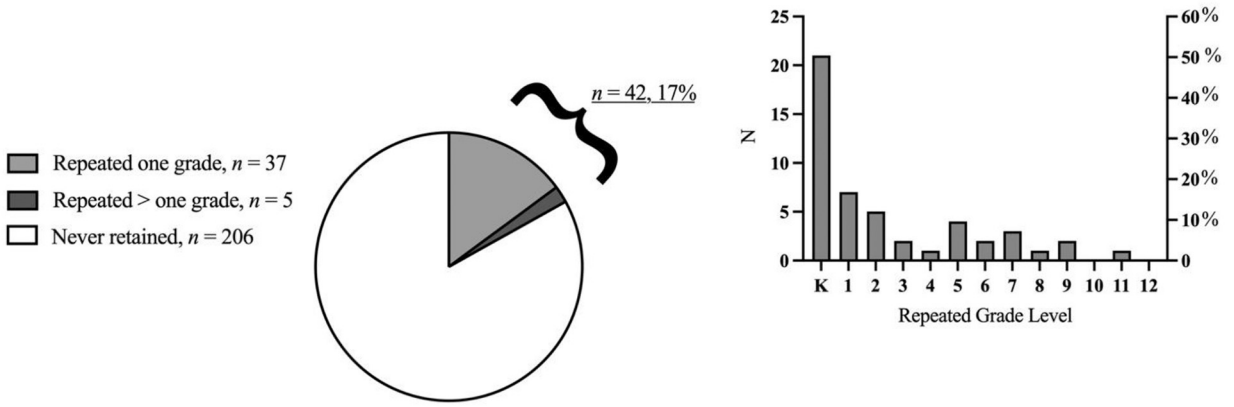

Fig. 1. Grade retention in students with SCA.
Note. Pie chart depicts number of students who repeated one or more grades during K–12 education ($n = 248$). Bar chart shows frequencies and percentages of each grade level repeated ($n = 42$).

(Table 3). A binomial test indicated our sample was significantly more likely to delay kindergarten than the general U.S. student population (U.S. rate = 6%; $p < .001$; Snyder & Dillow, 2013).

3.3. Grade retention

Approximately 17% ($n = 42$) of respondents reported that their child repeated one or more grades (Table 3). Although those with tetra or pentasomy SCA were significantly more likely to be retained (OR 2.3, 95% CI [1.04, 5.06], $p = .039$), there were no significant differences in retention by timing of diagnosis or specific trisomy SCA karyotype. Of those who were retained, the majority ($n = 40$) repeated grades in elementary school (K–5) with half ($n = 21$) specifically repeating kindergarten (Fig. 1). Only 14% of children repeated grades in middle school (Grades 6–8) and 7% repeated grades during their high school years (Grades 9–12). A binomial test indicated the sample was significantly more likely to repeat a grade than the general U.S. student population (U.S. rate = 2.2%; $p < .001$; U.S. Census Bureau, 2015).

3.4. School support plans

Overall, 213 (86%) respondents reported that their child had an individualized school support plan (Section 504 or IEP), with 27 respondents (11%) reporting their child had received both types of support plans at some point in their educational history (Table 3).

3.4.1. Section 504 plans

Over one quarter (26%) of respondents endorsed that their child had a Section 504 plan (Table 3). Children with trisomy SCA were significantly more likely to have a Section 504 Plan than those with tetra/pentasomy SCA (OR 0.21, 95% CI [0.07, 0.61], $p = .004$), but there were no significant differences by timing of diagnosis or among the trisomy SCA conditions. A binomial test indicated children of survey respondents were significantly more likely to have a Section 504 plan than the general U.S. student population (U.S. rate = 2.3%; $p < .001$; Zirkel & Huang, 2018).

3.4.2. Individualized education programs

A majority (71%) of survey respondents indicated their child had an IEP (Table 3), with no significant differences between ascertainment (prenatal vs. postnatal) groups. Children with tetra and pentasomy SCA conditions were significantly more likely to have an IEP than those with trisomy SCA (OR 6.61, 95% CI [2.23, 19.53], $p = .001$), and within the trisomy conditions, boys with 47, XYY were significantly more likely to have been reported as having an IEP (OR 0.30, 95% CI [0.10, 0.94], $p = .039$). A binomial test indicated the sample was significantly more likely to report having an IEP than the general U.S. student population (U.S. rate = 14%; $p < .001$; Hussar et al., 2020).

Parents reported a wide range of qualifying conditions for special education services (Table 3). The five most frequent eligible conditions included (a) Other Health Impaired (OHI; 36%), (b) Specific Learning Disability (SLD; 22%), (c) Autism (10%), (d) Speech Language Impairment (9%), and (e) Intellectual Disability (6%). There were no differences in qualifying educational diagnoses by timing of genetic diagnosis. Children with tetra/pentasomy SCAs were more likely to qualify for an IEP with an ID diagnosis than those with trisomy SCA ($p = .021$), although cell sizes were small and should be interpreted with caution. The vast majority (83%) of children

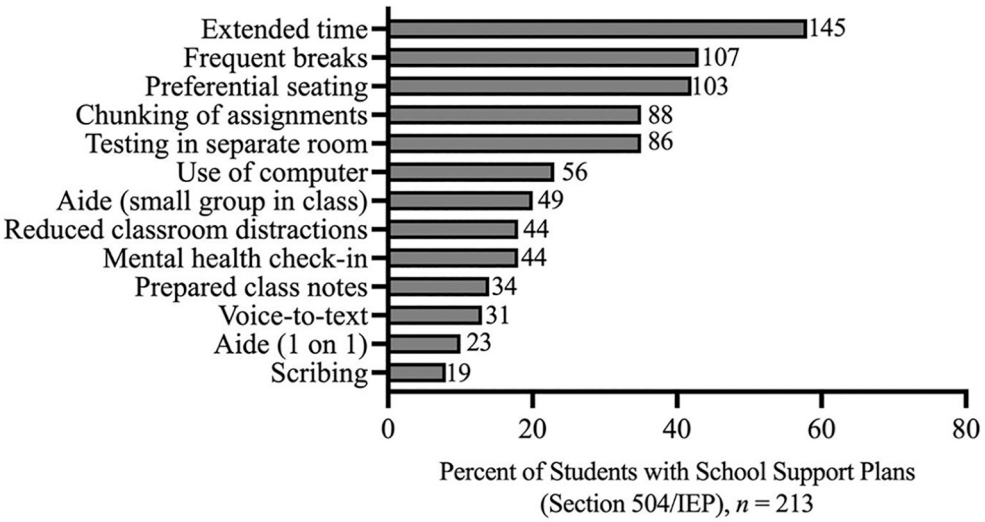

Fig. 2. Classroom accommodations for students with SCA.
Note. Frequencies of students with any individualized school support plan (Section 504 or IEP) receiving classroom accommodations ($n = 213$). Raw numbers are indicated at the end of each bar and percentages are indicated across the X axis.

with IEPs were receiving an inclusive education, with at least half of their school day spent in the general education classroom and no significant differences by timing of genetic diagnosis, trisomy versus tetra/pentasomy SCA condition, or among the trisomy conditions.

3.4.3. Classroom accommodations

Parents reported numerous classroom accommodations under the school support plans, with the five most frequently reported including (a) extended time for testing or assignments, (b) frequent breaks, (c) preferential seating, (d) chunking of larger assignments, and (e) testing in a separate room (Fig. 2).

3.4.4. School-based interventions

Parents reported their children received a range of school-based interventions through the IEPs, including academic interventions (78%), speech language therapy (60%), occupational therapy (34%), social skills instruction (37%), behavioral/mental health services (28%), and physical therapy (10%; Table 3). Fig. 3 shows school interventions for each SCA condition. Children with postnatal genetic diagnoses were reported to be significantly more likely to receive speech therapy ($p = .020$) and occupational therapy ($p = .016$) services through their IEP, whereas those with tetra/pentasomy SCAs were significantly more likely to receive speech therapy than those with trisomy SCAs ($p = .002$). There were no significant differences in school-based intervention services among the three trisomy conditions.

3.5. Post-secondary outcomes

Forty-one respondents with children between the ages of 18–21 years reported on high school completion, with most children (73%) graduating with a diploma and an additional 12% having received a General Education Development (i.e., high school equivalency) diploma (see Table 3). Two (5%) respondents reported that their child received a high school completion certificate and another two (5%) respondents indicated that their child was still in the K–12 system through a special education transition program but had not yet officially completed high school. No survey respondents endorsed that their child had dropped out of high school prior to completion. There were no significant differences in high school completion status by timing of diagnosis or specific trisomy SCA karyotype, and sample size for post-secondary participants with tetrasomy SCA ($n = 5$) was too limited for meaningful comparisons. Parents reported on their child's post-secondary status (Fig. 4), with nearly half (46%) currently enrolled in college (i.e., community, 2-year, or 4-year college/university) and 34% working in their community. Less than 15% of respondents endorsed 'unemployed' as best describing their child's post-secondary status. Of the 19 parents who reported their child was enrolled in college, eight children (42%) majored in the fine arts (e.g., graphic design, musical theater, music, dance, photography) and 14 (74%) children were reported to have received educational supports (e.g., extended time, mentoring/tutoring supports, prepared class notes, testing in a separate room) in the post-secondary setting. Those who were employed reported a range of entry-level jobs across industries (e.g., retail, healthcare, food service, childcare). Supports in the work setting included job coaching, accommodations for time management, and reduced work hours.

3.6. Parent perceptions of educators and school experiences

A vast majority (84%) of parents with children currently in the K–12 system reported they had informed their child's teachers about the genetic diagnosis. However, most respondents reported their child's educators lacked knowledge of SCAs (Fig. 5). Despite frequent reports of uninformed teachers, almost half of the parent respondents reported they were *mostly satisfied* to *highly satisfied* with the school's current supports for their child and <20% reported that they were *unsatisfied*.

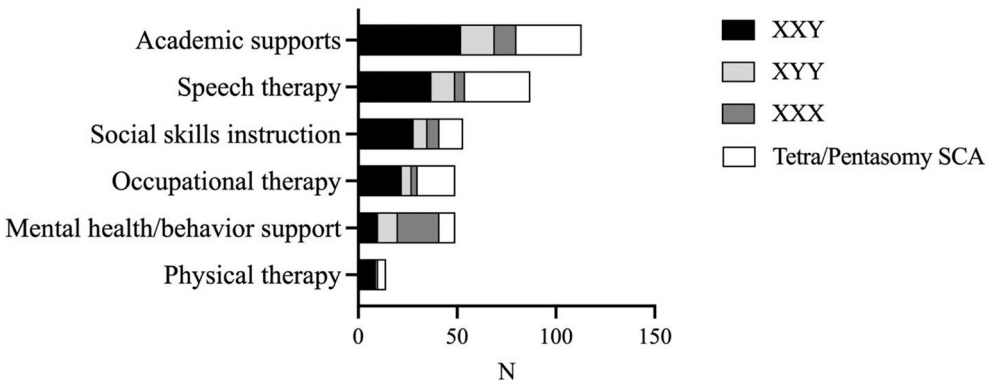

Fig. 3. School-based intervention services by condition.
Note. Frequencies of students, by condition, receiving school-based interventions from those currently enrolled in K–12 system who were endorsed as having an IEP ($N = 145$).

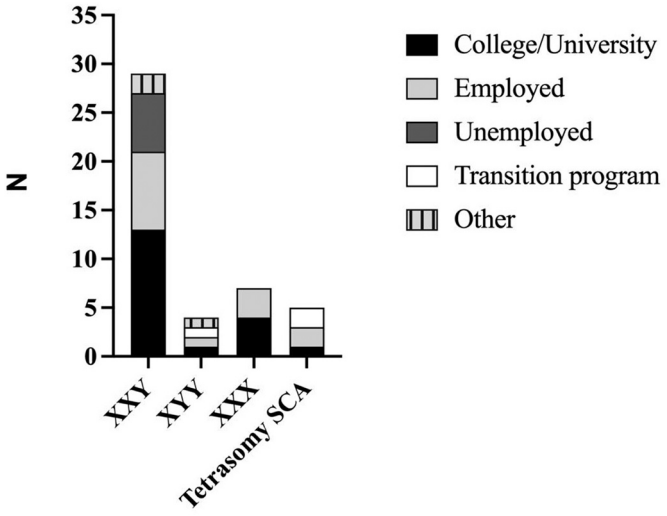

Fig. 4. Post-secondary status by condition.
Note. Frequencies of post-secondary status for students ages 18–21 years (n = 41; XXY = 26, XYY = 4, XXX = 6, tetra SCA = 5) by condition.

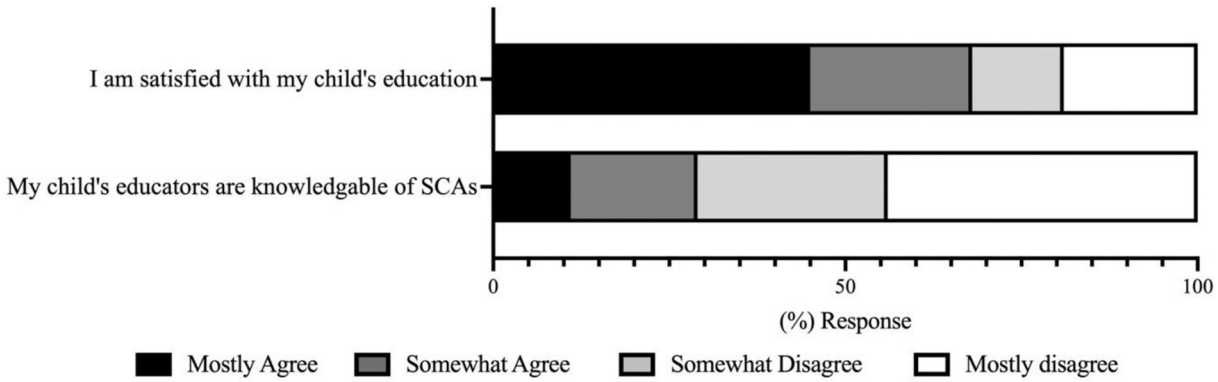

Fig. 5. Parent perceptions for students with SCAs.
Note. Parent perceptions on parent satisfaction with school supports and educators' knowledge of SCA conditions.

4. Discussion

This study was the first nationwide survey to document the school supports and educational outcomes for a relatively large sample of children with a known diagnosis of SCA in the United States. Our sample was significantly more likely to report receiving a variety of educational interventions and school support plans than the general population. As expected, based on more severe presentation in the phenotype, children in our sample with tetra/pentasomy SCA were significantly more likely to repeat a grade, receive special education services, qualify with an intellectual disability, and receive speech therapy than those with trisomy SCAs. Overall rates of individualized support plans did not differ by timing of genetic diagnosis (prenatal vs. postnatal diagnosis), but those with postnatal diagnoses were more likely to specifically receive speech and occupational therapies than prenatally diagnosed children. Prior research has documented improved outcomes in children with prenatal versus postnatally diagnosed SCAs, likely attributed to the fact that prenatally diagnosed individuals represent the broader spectrum of SCAs, including those with less pronounced symptoms and those who received proactive interventions earlier in life (Linden & Bender, 2002; Wigby et al., 2016). Most students were educated alongside typically developing peers in an inclusive classroom setting. Most parents disclosed their children's genetic diagnosis to the schools, yet most also felt their children's educators had little to no knowledge of the potential educational and behavioral implications of the diagnosis. Despite this lack of knowledge, many parents still felt at least somewhat satisfied with their children's educational experience.

Relatively high rates of delayed kindergarten entry in our sample (19%) suggest that some parents may have chosen to delay their child's school entry in response to a pattern of early developmental delays and/or recommendations from early interventionists or medical providers. Although delaying kindergarten entry is relatively rare in the United States (3%–7%), most parents make the decision based on concerns about development and a lack of school readiness (Fortner & Jenkins, 2017). Research in children

attending U.S. schools shows mixed results for academic outcomes in those who have delayed kindergarten entry. Children with speech-language impairment who delay kindergarten show mild academic advantages by Grade 3, whereas those with learning and cognitive disabilities who delay kindergarten show disadvantages in reading and mathematics as compared to those children who started kindergarten on time (Fortner & Jenkins, 2018). Similarly, grade retention was nearly four times more common for our sample (17%) than the public (4.3%; Child Trends Databank, 2018). Like delayed entry, research in the general population on grade retention has shown mixed results and is complicated by methodological challenges, such as the inability to control for external factors in students who are retained (Allen et al., 2009). Regardless, many studies have shown that grade retention in the early years (Grades 1–5) increases the risk of leaving high school without a diploma (Hughes et al., 2018). In our subset of children older than 18 years, all had a diploma equivalent or were still in secondary education, despite higher-than-average rates of delayed kindergarten and grade retention. Although this may suggest less harm from delayed kindergarten and grade retention for students with SCAs, it may also partially reflect an ascertainment bias in our sample, as those responding to a survey about educational outcomes may have been stronger advocates for school supports, or those feeling more negatively about their child's educational outcome may not have been enthusiastic about participation in the survey.

High rates of Section 504 plans and special education IEPs were confirmed as anticipated, as prior research has consistently shown that learning and academic challenges are common for students with SCAs, even though cognitive abilities are typically within the average range (Bender et al., 1993). Parents reported a range of qualifying special education disability categories. These mixed results reflect the previously described variable phenotype in children with SCAs, with their disability category likely reflecting their greatest area of educational need. Furthermore, students may have more than one area of need that may not be reflected in their disability category. For example, a child with dyslexia with an IEP under the SLD category may also have an expressive language impairment. For this reason, using primary and secondary disability codes may be helpful in the SCA student population. In our sample, families reported IEPs with a wide variety of intervention services, including academic, language, behavioral, social-emotional, and motor supports. An individualized approach is, therefore, recommended for students with SCAs.

Only one third of the sample was qualified under the OHI category, despite the fact that many areas of cognitive and academic deficit are likely predisposed from their genetic condition. OHI is currently the third most common IEP disability category nationwide, and encompasses students with limited strength, vitality, or alertness due to chronic or acute health problems (e.g., ADHD, diabetes, asthma; Hussar et al., 2020). There are high rates of ADHD diagnoses within the SCA conditions, ranging from 36% for boys with 47, XXY, 52% for girls with 47,XXX, 76% for boys with 47,XYY, to 72% for boys with 48,XXYY (Tartaglia et al., 2012). Inattentive type ADHD is the most common presentation in SCAs; however, signs of hyperactivity and impulsivity can also be present, especially in boys with an extra Y chromosome. Educators should routinely consider the Other Health Impairment special education category for children with SCAs to reflect common neurocognitive consequences of their medical disorder (e.g., problems with mental stamina, inhibition, mental flexibility, initiation of tasks, visual working memory; van Rijn & Swaab, 2015) and to understand that children may have ongoing needs related to their genetic condition.

Most respondents (78%) indicated their children received academic interventions through their IEP, with over 20% qualifying for special education due to an educational diagnosis of SLD. These results are consistent with prior research documenting deficits in reading accuracy, comprehension, and spelling for children with SCAs. In the early birth cohort natural history studies, up to 75% of participants met criteria for a diagnosis of language-based learning disability/dyslexia (Bender et al., 1986; Pennington et al., 1982). Several key cognitive processes have been identified across the lifespan as underlying skill deficits for individuals with SCAs, including weaknesses in phonemic discrimination, auditory processing, verbal working memory, comprehension, and fluency (Urbanus et al., 2020). Our findings emphasize that these previously identified skill deficits commonly interrupt learning in the school setting and are often being addressed through formal educational service plans for students with SCAs. Furthermore, although language-based learning disabilities were most common, our findings expand previous literature to highlight that many students with SCAs meet criteria for math learning disabilities or a combination of learning disabilities, similar to what is seen in the general student population where dyslexia and dyscalculia are highly comorbid and brain activity is similar between learning disabilities (Peters et al., 2018; Willcutt et al., 2019).

Approximately 10% of parents with a child who had an IEP reported that their child qualified for special education services under the Autism disability category and nearly half of all children with IEPs were reported to have received behavioral and/or social skills support services in the school setting. Prior SCA research has shown an increased risk for social skill deficits, with a particularly high risk for autism spectrum disorders in children with Y chromosome aneuploidy who are up to 20 times more likely to meet diagnostic criteria (Tartaglia et al., 2017). Supernumerary X chromosomes are also associated with an increased risk for ASD, and prior research shows that students with SCAs who do not meet the cutoff criteria for an ASD diagnosis may still demonstrate deficits in social cognition and communication skills (Cordeiro et al., 2012; Van Rijn et al., 2008).

High rates of reported behavioral health and social skill supports in the schools aligns with the documented risk profiles for mental health problems for youth with SCAs. Several studies have shown increased risk of developing internalizing problems throughout the lifespan, such as anxiety, depression, poor self-esteem, and social withdrawal (Freilinger et al., 2018; Turriff et al., 2011; Wigby et al., 2016). Externalizing challenges may also be present, including hyperactivity, impulsivity, or behavioral dysregulation in some children, which can present additional challenges and require further academic accommodations. School psychologists should be aware of the mental health risks in SCAs, as students who do not qualify for an IEP may still benefit from behavioral health consultation or supports within the school setting.

In terms of educational outcomes, the post-secondary group ($n = 41$) showed higher rates of attending college than an earlier prospective study of 42 individuals with SCAs (Bender et al., 1993). In their sample, Bender and colleagues documented that eight students (~20%) attended college after high school, whereas 19 students (~46%) from our sample reported attending college.

Although this may be a factor of bias in our sample, it may also reflect the impact of increased early intervention for young children with SCAs (Thompson et al., 2020) as well as improvements to transition planning and disability supports programs in U.S. colleges (Keenan et al., 2019; Newman et al., 2016). Many of the responding parents reported that their child continued to receive classroom accommodations at the college level, similar to those reported in the school-age group. Relatively high rates of students in our sample majored in fine arts (42%), which is an interesting and sensible finding considering the typical cognitive phenotype demonstrating strengths in visual-spatial processing in children with SCAs. This topic has not yet been explored in the literature and merits further investigation.

4.1. Limitations

Our study is limited by the survey design, as we were unable to verify the accuracy of parent reported details of Section 504 plans and IEPs (e.g., educational diagnoses, school-based service delivery, classroom accommodations). Furthermore, the cross-sectional nature of the survey meant that our denominators for key educational outcomes included students across all grade levels, including younger students who may not yet have experienced retention or school support plans, and therefore our proportions may be an underestimate. Although we report on a relatively large sample, our recruitment methods (open URL) precluded us from determining the response rate, and our sample size limited some statistical comparisons, particularly for subgroup analyses where cell sizes were often small. Therefore, these results should be interpreted with caution and are not generalizable to each SCA condition. However, as with any rare and understudied conditions, descriptive data can still be beneficial and add to our understanding of SCAs. In asking parents to rate the school's knowledge of SCAs, we were unable to specify which educators they were rating. Therefore, we were unable to determine if parents were describing the knowledge of classroom teachers, special educators, school psychologists, or other school-based therapists. Our sample was disproportionately college educated as compared with the U.S. population, and because children's educational outcomes are strongly associated with parent education attainment (Ludeke et al., 2021), our results may be skewed (although we controlled for parent education in all analyses to address this limitation). Respondents were required to read and respond to survey questions in English, potentially excluding non-English speaking participants who may experience additional educational barriers. Furthermore, our sample may be biased by those who chose to complete a survey titled *School Supports and Educational Outcomes for Children with X & Y Variations*. For example, our recruitment covered clinical populations, including many with postnatal diagnoses who may have more significant educational needs (although timing of diagnosis was rarely a significant covariate in our analyses). Additionally, families of children who dropped out of school may have chosen not to participate in a survey about school outcomes. Finally, we did not collect data on interventions that could potentially impact educational outcomes such as ADHD and anxiety medications or hormone treatment, as documenting the essential details of such treatments (e.g., dosage, timing, pubertal onset) was beyond the scope of this project. Despite these limitations, these are important data on educational practices and outcomes from a contemporary, nationwide sample.

4.2. Practical implications and future directions

Educators working with students with SCA conditions can use the results from this study to inform educational programming. Early childhood education teams should consider the high frequency of IEPs in school-aged children with SCAs when transitioning a child from Part C (early intervention) into Part B services (preschool special education). This is especially important considering parents tend to report a pattern of subclinical developmental concerns that narrowly miss the qualification cutoff for special education services in early childhood (Thompson et al., 2020). As such, it is important for teachers to closely follow the progress of children with SCAs as students who did not qualify for services in preschool may qualify in the early elementary grades or beyond as academic demands increase. Preschool and primary grade teachers should also be aware of an increased tendency for students with SCA to delay kindergarten and/or repeat early grade levels and be prepared to partner with school support teams and families to carefully consider potential harm and benefits of these practices. Acknowledging the possibility of ascertainment bias in our sample, our results support that some of the traditional arguments against grade retention being associated with lower rates of achieving a diploma may not be applicable to the SCA population.

Teachers may find classroom accommodations, such as extended time, frequent breaks, preferential seating, testing in a separate room, and prepared notes particularly useful for students with SCAs. As students with SCAs progress through school, they may need more interventions as difficulties related to attention and executive functioning deficits may become more apparent in the upper grades as demands for more complex and independent work increase. Therefore, updated testing and modifications to the IEP and consideration of moving from Section 504 Plans to IEPs are essential. Moreover, educational teams must be aware of the variability within SCA conditions and the need to individualize support plans to each unique student's pattern of strengths and needs. This may include consideration of how a student qualifies for special education (e.g., under which special education eligibility category), which educational interventions would likely benefit the student, and what supports may be indicated as a student transitions into their post-secondary years (Firth & Thompson, 2020). Furthermore, educators can capitalize on patterns of strengths that parents have identified in their children with SCAs, such as extraordinary kindness and an eagerness to please classmates and teachers, creative thinking, a love of learning, hardworking persistence, and relative aptitudes in science and math (Thompson et al., 2021).

Ample opportunities exist for further investigation of the educational experiences of more diverse samples of students with SCAs (e.g., non-English speaking, parents without college degrees), including higher education choices and workplace supports. Prospective studies could assess potential benefits of kindergarten delay, grade retention, special education services, and classroom accommodations on educational outcomes, as well as reasons why parents and schools make specific educational decisions. Finally, previous

research on learning profiles for children with SCAs provides theoretical support for implementation of evidence-based reading interventions (Pennington et al., 1982; Pennington & Bishop, 2009). However, more research is needed on specific educational interventions for areas of known risk in SCA populations (e.g., executive function, language impairment, reading disabilities, social skills, mood, behavior) so that recommendations for evidence-based practice can be shared with educators and therapists.

5. Conclusion

In summary, results suggested high rates of educational support services for students with SCA conditions in the U.S. educational system. As prenatal genetic testing practices, such as NIPS, are identifying more and more children with SCAs prenatally (Kornman et al., 2018), we anticipate a subsequent rise in the number of students with SCAs entering the school system. Educators, therapists, and medical providers should be aware of the frequent need for accommodations and individualized support plans in this population so that appropriate evaluations can be planned and interventions implemented in a timely manner to effectively support the student's highest level of success. Findings indicate that, with appropriate ecological supports, many students with SCA can successfully complete high school and pursue post-secondary education opportunities if desired. Finally, parent perceptions that educators lack knowledge of SCA conditions justifies a need to develop resources to train teachers and policy makers in the unique educational needs of children and adolescents with SCAs.

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Declaration of Competing Interest

None.

References

Abramsky, L., & Chapple, J. (1997). 47,XXY (Klinefelter syndrome) and 47,XYY: Estimated rates of and indication for postnatal diagnosis with implications for prenatal counselling. *Prenatal Diagnosis, 17*(4), 363–368. http://www.ncbi.nlm.nih.gov/pubmed/9160389.

Allen, C. S., Chen, Q., Willson, V. L., & Hughes, J. N. (2009). Quality of research design moderates effects of grade retention on achievement: A meta-analytic, multilevel analysis. *Educational Evaluation and Policy Analysis, 31*(4), 480–499.

American College of Obstetricians and Gynecologists' Committee on Practice Bulletins. (2020). Screening for Fetal Chromosomal Abnormalities: ACOG Practice Bulletin, Number 226. *Obstetrics and Gynecology, 136*(4), e48–e69. https://doi.org/10.1097/AOG.0000000000004084

Bender, B., Fry, E., Pennington, B., Puck, M., Salbenblatt, J., & Robinson, A. (1983). Speech and language development in 41 children with sex chromosome anomalies. *Pediatrics, 71*(2), 262–267. http://www.ncbi.nlm.nih.gov/entrez/query.fcgi?cmd=Retrieve&db=PubMed&dopt=Citation&list_uids=6823432.

Bender, B. G., Linden, M. G., & Robinson, A. (1993). Neuropsychological impairment in 42 adolescents with sex chromosome abnormalities. *American Journal of Medical Genetics, 48*(3), 169–173. https://doi.org/10.1002/ajmg.1320480312?sid=nlm%3Apubmed.

Bender, B. G., Puck, M. H., Salbenblatt, J. A., & Robinson, A. (1986). Dyslexia in 47,XXY boys identified at birth. *Behavior Genetics, 16*(3), 343–354.

Berglund, A., Stochholm, K., & Gravholt, C. H. (2020). The epidemiology of sex chromosome abnormalities. *American Journal of Medical Genetics, Part C Seminars in Medical Genetics, 184*(2), 202–215. https://doi.org/10.1002/ajmg.c.31805.

Bevilacqua, E., Ordonez, E., Hurtado, I., Rueda, L., Mazzone, E., Cirigliano, V., & Jani, J. C. (2018). Screening for sex chromosome aneuploidy by cell-free DNA testing: Patient choice and performance. *Fetal Diagnosis and Therapy, 44*(2), 98–104.

Boada, R., Janusz, J., Hutaff-Lee, C., & Tartaglia, N. (2009). The cognitive phenotype in Klinefelter syndrome: A review of the literature including genetic and hormonal factors. *Developmental Disabilities Research Reviews, 15*(4), 284–294. https://doi.org/10.1002/ddrr.83.

Boyd, P. A., Loane, M., Garne, E., Khoshnood, B., Dolk, H., & Group, E. W. (2011). Sex chromosome trisomies in Europe: Prevalence, prenatal detection and outcome of pregnancy. *European Journal of Human Genetics, 19*(2), 231–234. https://doi.org/10.1038/ejhg.2010.148.

Chen, K. M., White, K., Shabbeer, J., & Schmid, M. (2019). Maternal age trends support uptake of non-invasive prenatal testing (NIPT) in the low-risk population. *The Journal of Maternal-Fetal & Neonatal Medicine, 32*(23), 4039–4042. https://doi.org/10.1080/14767058.2018.1481033.

Child Trends Databank. (2018). Children who repeated a grade. https://www.childtrends.org/?indicators=children-who-repeated-a-grade.

Cordeiro, L., Tartaglia, N., Roeltgen, D., & Ross, J. (2012). Social deficits in male children and adolescents with sex chromosome aneuploidy: A comparison of XXY, XYY, and XXYY syndromes. *Research in Developmental Disabilities, 33*(4), 1254–1263. http://www.ncbi.nlm.nih.gov/entrez/query.fcgi?cmd=Retrieve&db=PubMed&dopt=Citation&list_uids=22502852.

Davis, S., Kowal, K., Tahsin, A., & Ross, J. (2019). *Testicular function in boys with 47,XYY. E-PAS2019:2155.4*.

Firth, E., & Thompson, T. (2020). Educational guidelines, IEPs, and school services for children with X & Y chromosome variations. https://genetic.org/wp-content/uploads/2020/12/AXYS-Consensus-Document-Educational-Guidelines-IEPs-School-Services-updated-12-14-20.pdf.

Fortner, C. K., & Jenkins, J. M. (2017). Kindergarten redshirting: Motivations and spillovers using census-level data. *Early Childhood Research Quarterly, 38*, 44–56.

Fortner, C. K., & Jenkins, J. M. (2018). Is delayed school entry harmful for children with disabilities? *Early Childhood Research Quarterly, 44*, 170–180.

Freilinger, P., Kliegel, D., Hanig, S., Oehl-Jaschkowitz, B., Henn, W., & Meyer, J. (2018). Behavioral and psychological features in girls and women with triple-X syndrome. *American Journal of Medical Genetics, Part A, 176*(11), 2284–2291. https://doi.org/10.1002/ajmg.a.40477.

Gargiulo, R. M., & Bouck, E. C. (2020). *Special education in contemporary society: An introduction to exceptionality*. SAGE Publications.

Hughes, J. N., West, S. G., Kim, H., & Bauer, S. S. (2018). Effect of early grade retention on school completion: A prospective study. *Journal of Educational Psychology, 110*(7), 974–991.

Hussar, B., Zhang, J., Hein, S., Wang, K., Roberts, A., Cui, J., Smith, M., Bullock Mann, F., Barmer, A., & Dilig, R. (2020). *The condition of education 2020*.

Jacobs, P. A., Melville, M., Ratcliffe, S., Keay, A. J., & Syme, J. (1974). A cytogenetic survey of 11,680 newborn infants. *Annals of Human Genetics, 37*(4), 359–376. http://www.ncbi.nlm.nih.gov/entrez/query.fcgi?cmd=Retrieve&db=PubMed&dopt=Citation&list_uids=4277977.

Keenan, W. R., Madaus, J. W., Lombardi, A. R., & Dukes, L. L., III (2019). Impact of the Americans with disabilities act amendments act on documentation for students with disabilities in transition to college: Implications for practitioners. *Career Development and Transition for Exceptional Individuals, 42*(1), 56–63.

Kornman, L., Palma-Dias, R., Nisbet, D., Scott, F., Menezes, M., da Silva Costa, F., & McLennan, A. (2018). Non-invasive prenatal testing for sex chromosome aneuploidy in routine clinical practice. *Fetal Diagnosis and Therapies, 44*(2), 85–90. https://doi.org/10.1159/000479460.

Leggett, V., Jacobs, P., Nation, K., Scerif, G., & Bishop, D. V. (2010). Neurocognitive outcomes of individuals with a sex chromosome trisomy: XXX, XYY, or XXY: A systematic review. *Developmental Medicine and Child Neurology, 52*(2), 119–129. https://doi.org/10.1111/j.1469-8749.2009.03545.x.

Linden, M. G., & Bender, B. G. (2002). Fifty-one prenatally diagnosed children and adolescents with sex chromosome abnormalities. *American Journal of Medicap Genetics, 110*(1), 11–18. https://doi.org/10.1002/ajmg.10394.

Ludeke, S. G., Gensowski, M., Junge, S. Y., Kirkpatrick, R. M., John, O. P., & Andersen, S. C. (2021). Does parental education influence child educational outcomes? A developmental analysis in a full-population sample and adoptee design. *Journal of Personality and Social Psychology, 120*(4), 1074.

Martin, S., Cordeiro, L., Richardson, P., Davis, S., & Tartaglia, N. (2019). The association of motor skills and adaptive functioning in XXY/Klinefelter and XXYY syndromes. *Physical & Occupational Therapy in Pediatrics, 39*(4), 446–459. https://doi.org/10.1080/01942638.2018.1541040.

National Center for Education Statistics. (2020). Educational attainment of young adults. In *The condition of education 2020 (NCES 2020–144)*.

Newman, L. A., Madaus, J. W., & Javitz, H. S. (2016). Effect of transition planning on postsecondary support receipt by students with disabilities. *Exceptional Children, 82*(4), 497–514.

Pennington, B. F., Bender, B., Puck, M., Salbenblatt, J., & Robinson, A. (1982). Learning disabilities in children with sex chromosome anomalies. *Child Development, 53*(5), 1182–1192. http://www.ncbi.nlm.nih.gov/entrez/query.fcgi?cmd=Retrieve&db=PubMed&dopt=Citation&list_uids=7140426.

Pennington, B. F., & Bishop, D. V. (2009). Relations among speech, language, and reading disorders. *Annual Review of Psychology, 60*, 283–306. http://www.ncbi.nlm.nih.gov/entrez/query.fcgi?cmd=Retrieve&db=PubMed&dopt=Citation&list_uids=18652545.

Peters, L., Bulthé, J., Daniels, N., Op de Beeck, H., & De Smedt, B. (2018). Dyscalculia and dyslexia: Different behavioral, yet similar brain activity profiles during arithmetic. *NeuroImage Clinical, 18*, 663–674. https://doi.org/10.1016/j.nicl.2018.03.003.

van Rijn, S., Barneveld, P., Descheemaeker, M. J., Giltay, J., & Swaab, H. (2018). The effect of early life stress on the cognitive phenotype of children with an extra X chromosome (47,XXY/47,XXX). *Child Neuropsychology, 24*(2), 277–286. https://doi.org/10.1080/09297049.2016.1252320.

van Rijn, S., & Swaab, H. (2015). Executive dysfunction and the relation with behavioral problems in children with 47,XXY and 47,XXX. *Genes, Brain, and Behavior, 14*(2), 200–208. https://doi.org/10.1111/gbb.12203.

Robinson, A., Bender, B., Borelli, J., Puck, M., Salbenblatt, J., & Webber, M. L. (1982). Sex chromosomal abnormalities (SCA): A prospective and longitudinal study of newborns identified in an unbiased manner. *Birth Defects Original Article Series, 18*(4), 7–39. http://www.ncbi.nlm.nih.gov/entrez/query.fcgi?cmd=Retrieve&db=PubMed&dopt=Citation&list_uids=7159724.

Rovet, J., Netley, C., Keenan, M., Bailey, J., & Stewart, D. (1996). The psychoeducational profile of boys with Klinefelter syndrome. *Journal of Learning Disabilities, 29*(2), 180–196. http://www.ncbi.nlm.nih.gov/pubmed/8820203.

Salbenblatt, J., Meyers, D. C., Bender, B., Linden, M. G., & Robinson, A. (1987). Gross and fine motor development in 47,XXY and 47,XYY males. *Pediatrics, 80*, 240–244.

Salbenblatt, J. A., Meyers, D. C., Bender, B. G., Linden, M. G., & Robinson, A. (1989). Gross and fine motor development in 45,X and 47,XXX girls. *Pediatrics, 84*(4), 678–682. http://www.ncbi.nlm.nih.gov/entrez/query.fcgi?cmd=Retrieve&db=PubMed&dopt=Citation&list_uids=2780130.

Samango-Sprouse, C. A., Stapleton, E., Sadeghin, T., & Gropman, A. L. (2013). Is it all the X: Familial learning dysfunction and the impact of behavioral aspects of the phenotypic presentation of XXY? *American Journal of Medical Genetics, Part C Seminars in Medical Genetics, 163C*(1), 27–34. http://www.ncbi.nlm.nih.gov/entrez/query.fcgi?cmd=Retrieve&db=PubMed&dopt=Citation&list_uids=23359595.

Simpson, N. H., Addis, L., Brandler, W. M., Slonims, V., Clark, A., Watson, J., Scerri, T. S., Hennessy, E. R., Bolton, P. F., & Conti-Ramsden, G. (2014). Increased prevalence of sex chromosome aneuploidies in specific language impairment and dyslexia. *Developmental Medicine and Child Neurology, 56*(4), 346–353.

Skakkebaek, A., Bojesen, A., Kristensen, M. K., Cohen, A., Hougaard, D. M., Hertz, J. M., Fedder, J., Laurberg, P., Wallentin, M., Østergaard, J. R., Pedersen, A. D., & Gravholt, C. H. (2014). Neuropsychology and brain morphology in Klinefelter syndrome - the impact of genetics. *Andrology, 2*(4), 632–640. https://doi.org/10.1111/j.2047-2927.2014.00229.x.

Snyder, T. D., & Dillow, S. A. (2013). *Digest of education statistics, 2012. NCES 2014–015*. National Center for Education Statistics.

Tartaglia, N., Ayari, N., Howell, S., D'Epagnier, C., & Zeitler, P. (2011). 48,XXYY, 48,XXXY and 49,XXXXY syndromes: not just variants of Klinefelter syndrome. *Acta Paediatrica, 100*(6), 851–860. https://doi.org/10.1111/j.1651-2227.2011.02235.x.

Tartaglia, N., Howell, S., Davis, S., Kowal, K., Tanda, T., Brown, M., & Thompson, T. (2020). Early neurodevelopmental and medical profile in children with sex chromosome trisomies: Background for the prospective eXtraordinarY babies study to identify early risk factors and targets for intervention. *American Journal of Medical Genetics Part C: Seminars in Medical Genetics, 184*(2), 428–443. https://doi.org/10.1002/ajmg.c.31807

Tartaglia, N. R., Ayari, N., Hutaff-Lee, C., & Boada, R. (2012). Attention-deficit hyperactivity disorder symptoms in children and adolescents with sex chromosome aneuploidy: XXY, XXX, XYY, and XXYY. *Journal of Developmental and Behavioral Pediatrics, 33*(4), 309–318. http://www.ncbi.nlm.nih.gov/entrez/query.fcgi?cmd=Retrieve&db=PubMed&dopt=Citation&list_uids=22333574.

Tartaglia, N. R., Wilson, R., Miller, J. S., Rafalko, J., Cordeiro, L., Davis, S., … Ross, J. (2017). Autism spectrum disorder in males with sex chromosome aneuploidy: XXY/Klinefelter syndrome, XYY, and XXYY. *Journal of Developmental and Behavioral Pediatrics, 38*(3), 197–207. https://doi.org/10.1097/DBP.0000000000000429.

Thompson, T., Davis, S., Takamatsu, S., Howell, S., & Tartaglia, N. (2021). Exploring academic and character strengths in students with sex chromosome aneuploidies. *Journal of Positive School Psychology*, 1–13 (FirstView articles) https://journalppw.com/index.php/JPPW/article/view/262.

Thompson, T., Howell, S., Davis, S., Wilson, R., Janusz, J., Boada, R., & Tartaglia, N. (2020). Current survey of early childhood intervention services in infants and young children with sex chromosome aneuploidies. *American Journal of Medical Genetics Part C: Seminars in Medical Genetics, 184*(2), 414–427. https://doi.org/10.1002/ajmg.c.31785

Turriff, A., Levy, H. P., & Biesecker, B. (2011). Prevalence and psychosocial correlates of depressive symptoms among adolescents and adults with Klinefelter syndrome. *Genetics in Medicine, 13*(11), 966–972. https://doi.org/10.1097/GIM.0b013e3182227576.

U.S. Census Bureau. (2015). Current population survey, digest of education statistics. https://nces.ed.gov/programs/raceindicators/indicator_rda.asp.

Urbanus, E., van Rijn, S., & Swaab, H. (2020). A review of neurocognitive functioning of children with sex chromosome trisomies: Identifying targets for early intervention. *Clinical Genetics, 97*(1), 156–167.

Van Rijn, S., Swaab, H., Aleman, A., & Kahn, R. (2008). Social behavior and autism traits in a sex chromosomal disorder: Klinefelter (47XXY) syndrome. *Journal of Autism and Developmental Disorders, 38*(9), 1634–1641. https://link.springer.com/content/pdf/10.1007%2Fs10803-008-0542-1.pdf.

Walzer, S., Bashir, A. S., & Silbert, A. R. (1990). Cognitive and behavioral factors in the learning disabilities of 47,XXY and 47,XYY boys. *Birth Defects Original Article Series, 26*(4), 45–58. http://www.ncbi.nlm.nih.gov/pubmed/2090328.

Walzer, S., Graham, J. M., Jr., Bashir, A. S., & Silbert, A. R. (1982). Preliminary observations on language and learning in XXY boys. *Birth Defects Original Article Series, 18*(4), 185–192. http://www.ncbi.nlm.nih.gov/entrez/query.fcgi?cmd=Retrieve&db=PubMed&dopt=Citation&list_uids=7159718.

Wigby, K., D'Epagnier, C., Howell, S., Reicks, A., Wilson, R., Cordeiro, L., & Tartaglia, N. (2016). Expanding the phenotype of Triple X syndrome: A comparison of prenatal versus postnatal diagnosis. *American Journal of Medical Genetics: Part A, 170*(11), 2870–2881. https://doi.org/10.1002/ajmg.a.37688.

Willcutt, E. G., McGrath, L. M., Pennington, B. F., Keenan, J. M., DeFries, J. C., Olson, R. K., & Wadsworth, S. J. (2019). Understanding comorbidity between specific learning disabilities. *New Directions in Child and Adolescent Development, 2019*(165), 91–109. https://doi.org/10.1002/cad.20291.

Zirkel, P., & Huang, T. (2018). State rates of 504-only students in K–12 public schools: An update. *West's Education Law Reporter, 354*, 621–625.