Journal of Positive School Psychology
2022, Vol. 6, No. 1, 12-24

http://journalppw.com/jppw
https://doi.org/10.47602/jpsp.v6i1.262

Research Article

Exploring Academic and Character Strengths in Students with Sex Chromosome Aneuploidies

Talia Thompson[1,2], Shanlee Davis[1,2], Stephanie Takamatsu[1], Susan Howell[1,2], and Nicole Tartaglia[1,2]

Abstract

Children with sex chromosome aneuploidies (SCAs) are often characterized in the literature by limitations and pathologies related to the genetic diagnosis. This study aimed to broaden the SCA phenotype by describing parent reported character and academic strengths. Parents of children with SCAs ages 3-21 (*N*=377) responded to an electronic survey asking them to describe their child's strengths in academic settings. Responses were coded for strengths-based content and analyzed using a mixed-methods content analysis approach. We identified overarching qualitative themes of Social Strengths and Assets for Learning. Quantitative results showed a pattern of overlapping strengths among the trisomy SCAs (perseverance and love of learning), with some significant differences between children with supernumerary X chromosomes (strengths in kindness) and those with an additional Y chromosome (strengths in curiosity, humor, and teamwork). Suggestions for future strengths-based research and educational practices to address academic, developmental, and psychosocial risks are explored.

Keywords: Sex chromosome aneuploidy; positive psychology; character strengths; mixed methods; Klinefelter syndrome; school psychology

Students with rare genetic conditions, such as sex chromosome aneuploidies (SCAs), may require significant supports in the school setting. As such, it is important for educators, clinicians, and families to have a solid understanding of how students with these conditions might present in the classroom and how to best support their potential needs. It is critical that educators understand the genetics and medical features of a student's condition(s) as well as risk factors for neurodevelopmental and learning problems associated with the diagnosis. However, much of the current data available on genetic conditions is deficit focused, which overshadows areas of potential strength related to a diagnosis. The field of positive psychology has the potential to expand our understanding of genetic differences and better support students with these conditions.

Sex Chromosome Aneuploidies

SCAs are caused by the presence of extra sex (X or Y) chromosomes in the karyotype. Several genetic diagnoses fall under this umbrella term with a collective prevalence rate of 1 in 500 live births (Hamerton et al., 1975). Trisomy SCAs (47,XXY/Klinefelter syndrome, 47,XYY/Jacob syndrome, 47,XXX/trisomy X) are more common and are associated with a more variable phenotypic presentation. Although the three trisomy conditions have substantial overlap in features, research has documented differences in patterns of weaknesses in cognitive-behavioral and motor profiles due to genetic and hormonal effects of having an extra X and/or Y chromosome (Urbanus et al., 2020). For example, children with 47,XYY tend to report stronger overall cognitive and motor skills, but more impaired language, increased hyperactivity, and higher rates of

[1] University of Colorado, School of Medicine, Aurora, CO, USA
[2] eXtraordinarY Kids Clinic and Research Program, Children's Hospital Colorado, Aurora, CO, USA
Corresponding Author: Talia Thompson, Children's Hospital Colorado, 13123 East 16th Avenue, Box B140, Aurora, CO 80045, USA.
Email: talia.thompson@childrenscolorado.org

autism spectrum disorder (ASD) than children with extra X chromosomes (Leggett et al., 2010; Ross et al., 2009; Tartaglia et al., 2012; Tartaglia et al., 2017). Girls with 47,XXX have been shown to have higher rates of overall cognitive impairment and more concerning psychological problems, while boys with 47,XXY tend to demonstrate more specific deficits in verbal reasoning abilities and gross motor skills (Leggett et al., 2010; Ross et al., 2009). Tetra and pentasomy SCAs (48,XXYY, 48,XXXY, 48,XXXX, 49,XXXYY, and 49,XXXXX) occur less frequently and are characterized by greater risks for developmental delays and medical problems and more significant educational needs (Tartaglia et al., 2011).

In general, students with SCAs have increased risks for learning disabilities and academic challenges, medical problems and hormone dysfunction, as well as increased risk for psychological disorders that can impact school functioning such as ADHD, ASD, anxiety, and depression (Tartaglia et al., 2020; Urbanus et al., 2020). Natural history studies have documented a pattern of educational struggles and current surveys have shown high rates of early interventions and special education supports in this student population (Bender et al., 1993; Linden & Bender, 2002; Rovet et al., 1996; Thompson et al., 2020). Research on the cognitive and behavioral phenotypes of SCAs has historically been rooted in a medical model, focused on identifying risks for atypical developmental patterns and educational failure. Although the ultimate goal is to inform treatment and educational priorities and to improve quality of life, deficit-oriented research fails to capture a holistic understanding of children with SCAs and possible positive outcomes associated with the diagnosis. Furthermore, solely focusing on associated problems might impact parent perceptions of the child after diagnosis, child self-concept, or teacher expectancies (Riggan et al., 2020; Rosenthal, 1991).

Character strengths

Positive psychology provides a unique and innovative framework for understanding the SCA phenotype. In direct contrast with the dominant pathological approach to psychology, positive psychology proposes a classification system for understanding the positive traits of humans including six broad virtues shown to be valued worldwide: wisdom, courage, humanity, justice, temperance, and transcendence (Peterson & Seligman, 2004). Each virtue is comprised of multiple specific character strengths that constitute the best aspects of an individual's personality (Table 1). Although people can exhibit a variety of strengths, 'signature strengths' are those that are most essential to one's identity and behavior; strengths that are most often expressed and observed by others. Interventions that aim to identify student's signature strengths and promote the practice and application of those strengths in daily life have been shown to improve overall wellbeing (Schutte & Malouff, 2019).

Table 1. Positive psychology character strengths and virtues

Virtue	Strengths
Wisdom:	Creativity
Strengths that help to acquire and apply knowledge	Curiosity
	Judgment
	Love of learning
	Perspective
Courage:	Bravery
Strengths that help to exert will and meet adversity	Perseverance
	Honesty
	Zest
Humanity:	Love
Strengths that help to connect with others in one-on-one relationships	Kindness
	Social intelligence
Justice:	Teamwork
Strengths that help to succeed in community and work in groups	Fairness
	Leadership
Temperance:	Forgiveness
Strengths that help to protect against excess and manage behavior	Humility
	Prudence
	Self-regulation
Transcendence:	Appreciation of beauty and excellence
Strengths that help to provide meaning and connection with the wider world	Gratitude
	Spirituality
	Hope
	Humor

Note. (Niemiec & McGrath, 2019; Peterson & Seligman, 2004)

To date, there has been a notable lack of research on the academic or character strengths of children with SCAs. As rates of prenatally diagnosed SCA rise with the increased use of non-invasive prenatal testing (Howard-Bath et al., 2018), parents, clinicians, and educators require a more complete understanding of these diagnoses. Recognition of the positive traits of children with SCA has the potential to enlighten clinical practice and guide research aimed at developing motivating, ecologically valid, and strengths-based interventions to optimize child and family outcomes by ameliorating risks for developmental, educational, and psychosocial problems.

This study aimed to characterize the SCA

phenotype from a positive psychology perspective through a mixed-methods content analysis of parent reported academic and character strengths in children and young adults with SCAs. This study was primarily exploratory, as there have been no research studies to date specifically examining character strengths in children with SCAs. However, based on the literature, we did hypothesize that academic strengths in science, technology, engineering, and math (STEM) would be coded more frequently than in language arts, based on known profiles of weakness in verbal reasoning as well as high rates of reading disabilities for children and adults with SCAs (Simpson et al., 2014). We also hypothesized there would be differences in strengths profiles between children with supernumerary X versus Y chromosomes within the trisomy conditions, as has been documented in previous literature regarding differences in risk profiles (Urbanus et al., 2020).

Method

Study Design and Participant Recruitment

This international electronic survey study used a mixed-methods content analysis design (Creswell & Clark, 2017; Park & Peterson, 2006) to both generate and test hypotheses about strengths for students with SCAs. Participants were recruited through email listservs and social media websites for the eXtraordinarY Kids clinic of Children's Hospital Colorado and the Association for X and Y Chromosome Variations (AXYS) advocacy organization. Participants were included if they were parents or guardians of a child with a diagnosed sex chromosome aneuploidy (SCA), ages three to 21-years, and were able to read and respond to survey questions in English. All participants provided consent for research prior to completing the survey, results were kept anonymous, and the study was approved by the Colorado Multiple Institutional Review Board (#19-0555).

Instrument

Data included in this analysis came from a larger, international, electronic survey on early therapies, school supports, and educational outcomes for children with SCAs (Thompson et al., 2020). The survey was administered via REDCap electronic data capture tool hosted at the University of Colorado School of Medicine and was developed by an interdisciplinary team of clinicians and researchers in SCAs and then piloted with parents of children with SCA conditions to ensure both content and face validity. Although the larger survey included a mixture of true/false, multiple choice, and open-text questions, the data reported here are limited to one open-ended survey question on a child's strengths. Because the broader survey focused on education related topics, parents were asked to consider their child's strengths in context of the current educational/work setting. Questions differed slightly by age group: parents of preschool and school-aged children were asked, 'What are your child's strengths in the classroom?' and parents of children who had completed high school were asked, 'What are your child's strengths in the school or work setting?' Questions were purposely open-ended, allowing respondents to provide free-text responses of any length, encouraging emergent parent perspectives and mitigating potential bias caused by predetermined categories or rating forms.

Table 2. Study sample demographics

Demographic Variable	*N*=377 (%)
SCA Condition	
XXY	197(52.3)
XXX	51 (13.5)
XYY	46(12.2)
XXYY	56(14.9)
XXXY	22(5.8)
XXXX	2(<1)
Other (pentasomy SCAs, mosaic SCA)	3(<1)
Timing of diagnosis	
Prenatal	153(40.6)
Postnatal	215(57)
Unreported	9(2.4)
Age	
Child, M±SD	11.5±5.5
Parent, M±SD	45.8±9.0
Caregiver	
Mother	336(89.1)
Father	32(8.5)
Other/Unreported	9(2.4)
Child Sex	
Male	322(85.4)
Female	55(14.6)
Country of Residence	
USA	304(80.6)
Europe	29(7.7)
Canada	17(4.5)
Australia/New Zealand	9(2.4)
Asia	4(1.1)
Other/ Unreported	14(3.7)
Highest Education Completed: Respondent	
Advanced degree (e.g., Master's, PhD, MD)	130(34.5)
Bachelor's degree	125(33.2)
<Bachelor's degree	107(28.4)
Unreported	15(4)

Analytic Plan

Qualitative Analysis

A positive psychology content analysis (Park & Peterson, 2006) guided the qualitative strand of analysis to characterize parent reported strengths in children with SCAs. Survey responses were downloaded from REDCap and uploaded to Excel spreadsheets and qualitative analytic software (ATLAS.ti) for storage and analysis. The first author developed a preliminary deductive codebook based on the 24 positive psychology character strengths (Peterson & Seligman, 2004). The initial codebook also included major developmental domains (cognitive, language, motor, social) and performance strengths (talents), such as specific academic subject areas, artistic abilities, and athleticism in order to capture the broad spectrum of potential parent responses. Inductive, "first cycle" open coding was also used to capture any emergent strengths not covered by the existing coding structure, including the use of in-vivo (verbatim) codes named directly from the language of the participants rather than researcher imposed terms (Saldaña, 2015).

The coding team included a primary coder who coded all survey responses (psychologist with expertise in positive psychology, qualitative methods, and SCAs) and two reliability coders who each co-coded 100 (27%) of responses (endocrinologist with expertise in SCAs and qualitative methods; psychologist with expertise in child development and SCAs). All coders were unaware of group (SCA condition) although gendered pronouns which might reveal a condition could not be avoided. The coding team met multiple times; first for consensus coding of 20 responses to establish a working codebook with code definitions. Next, the team coded another 20 responses independently before meeting again to review results and update the codebook to ensure it accurately represented consensus codes and code definitions. Using this updated codebook, each member co-coded another 60 responses before calculating intercoder agreement (ICA) in ATLAS.ti. ICA reached a Krippendorff's α-binary coefficient >.90; supporting the trustworthiness of the coding process and the qualitative results (O'Connor & Joffe, 2020).

Table 3. Qualitative results; themes, categories, codes, sample quotes

Social Strengths	Extraordinary Kindness		Eager to Please	
Codes	sweet, gentle, kindness, love		helpful, eager to please, rule follower, good behavior, teamwork, social intelligence	
Quotes	'He is polite, gentle, and shares things with others' 'Heart of gold' 'He is very kind and will easily work and play with other children'		'She is a people pleaser, loves to try to help people' 'Extremely helpful and polite to teachers and staff.' 'Her teachers always love her because she makes them presents each day.'	
Assets for Learning	Strengths in STEM	Creative Thinkers	Loves to Learn	Hard-working
Codes	math, science, technology, engineering, visual-spatial reasoning	creativity, visual arts, music, dance, performing arts	love of learning, curiosity, cognitive, smart/intelligent, motivated	Hard-working, perseverance
Quotes	'He loves science and hands-on based learning. He has an aptitude for robotics and engineering well beyond his years.' 'Math is his favorite subject. He calls himself a 'Math Master.' 'He excels in science - scored top marks in Year 3 and is one of the top scientists in his class'	'She is very creative and thinks 'outside the box'' 'Very imaginative' 'He is very creative and an amazing artist' 'He has an amazing imagination' 'She's very creative in singing, drama'	'Eagerness to learn and be challenged.' 'Loves school!! Is really motivated to learn new things.' 'He loves to read, has a very curious mind and is willing to try' 'He is like a sponge and soaks up information he hears on subjects that he likes.'	'Her strengths are how hard she studies and how hard she works' 'Always tries his best even when he doesn't know or understand the material' 'Persistent, tenacious, problem solver'

Any disagreements on codes were discussed by the group and survey responses were recoded to better reflect the consensus coding structure. After ICA was determined, the primary coder used the updated codebook to independently code all remaining responses (N=277). Finally, the entire team (including genetic counselor with expertise in SCAs; developmental behavioral pediatrician and principal investigator with expertise in SCAs) engaged in data reduction through "second cycle" coding (Saldaña, 2015); collapsing and combining codes, developing code networks, and examining code co-occurrences to develop categories and broad themes describing parent reported strengths in children with SCAs.

Quantitative Analysis

In the quantitative strand of the study, code frequency data for strengths by academic subjects and the 24 positive psychology character strengths were exported to Excel. The most commonly coded academic subjects were collapsed into three overarching subject area code groups (science/technology/engineering/math=STEM; reading/writing/spelling=Language Arts; music/visual arts/dance/performing arts=Fine Arts). Descriptive statistics (frequencies of codes and proportions) were calculated for each trisomy SCA group, adjusted for sample size, and then plotted in bar charts for visual comparisons. Next, the top ten total most frequently applied character strength codes, as well as the three broad academic subject area code groups, were transformed to binary data, coded as present or not for each participant (1=yes, 0=no), and uploaded into statistical software (SPSS.26) to compare frequencies between the trisomy SCA groups using Pearson's Chi-square or Fisher's exact analysis, depending on the cell size. Statistical analysis was limited to the top ten character strengths codes as frequencies for other character strengths codes were too low for meaningful or valid analysis. Significant findings ($p<.05$) were analyzed pairwise with post hoc Chi-square and Fisher's exact comparisons. We did not adjust for multiple comparisons as cell sizes were relatively small and this study was meant to be exploratory and hypothesis generating (Feise, 2002).

Mixed Methods Analysis

Results from the qualitative and quantitative strands were merged through a joint display (interpretive matrix). Aligning the data sets highlighted areas of possible congruence, discordance, or expansion, providing a more robust representation of strengths in children with SCAs (Guetterman et al., 2015).

Results

Demographics

Sample demographics are described in Table 2.

550 individuals consented to participate and 428 provided codable answers for the survey question on child strengths. Fifty-one respondents did not meet inclusion criteria (one had a different chromosomal trisomy, 50 were out of age window for these analyses; under 3-years), resulting in a total N of 377 respondents.

Table 4. Code frequencies, ranks, and comparisons by group

		Group				Pairwise comparisons		
Rank	Code	XXY *N*=197	XXX *N*=51	XYY *N*=46	*p*	XXY v XXX	XXY v XYY	XXX v XYY
Character strengths codes								
1	Kindness	27(13.7)	12(23.5)	6(13)	.199			
2	Love of learning	23(11.7)	10(23.8)	9(19.6)	.193			
3	Perseverance	25(12.7)	11(21.6)	7(15.2)	.279			
4	Social intelligence	22(11.2)	8(15.7)	5(10.9)	.687			
5	Creativity	23(11.7)	5(9.8)	4(8.7)	.821			
6	Self-regulation	13(6.6)	1(2)	1(2.2)	.369			
7	Teamwork	11(5.6)	1(2)	7(15.2)	.028*	.469	.033*	.025*
8	Curiosity	10(5.1)	0(0)	7(15.2)	.005*	.222	.024*	.004*
9	Zest	8(4.1)	2(3.9)	1(2.2)	1.00			
10	Humor	4(2)	1(2)	4(8.7)	.066			
Academic subject area code groups								
1	STEM	63(32)	6(11.8)	13(28.3)	.016*	.005*	.725	.071
2	Fine Arts	23(11.7)	9(17.6)	3(6.5)	.271			
3	Language Arts	16(8.1)	4(7.8)	3(6.5)	1.00			

Note. Positive psychology character strength and academic subject area code frequencies ranked by total frequency count and compared by SCA group *N* (%). *$p<.05$

Table 5. Joint display: Alignment of data strands with linking relationships

QUAL	Link	QUANT		
Theme: Category		Strengths Code/ Academic Code Group	Rank	Group differences
Social Strengths: Extraordinary kindness	*Congruent*	Kindness	1[a]	*p*=.199†
Social Strengths: Eager to Please	*Congruent*	Social Intelligence	4[a]	*p*=.687†
	Expansion	Teamwork (XYY>XXY & XYY>XXX)	7[a]	*p*=.028*
Assets for Learning: Strengths in STEM	*Congruent* *Expansion*	STEM (XXY>XXX)	1[b]	*p*=.016*
Assets for Learning: Loves to learn	*Congruent*	Love of Learning	2[a]	*p*=.193†
	Expansion	Curiosity (XYY>XXY & XYY>XXX)	8[a]	*p*=.005*
Assets for Learning: Creative thinkers	*Congruent*	Creativity	5[a]	*p*=.821†
		Fine Arts	2[b]	*p*=.271†
Assets for Learning: Hard-working students	*Congruent*	Perseverance	3[a]	*p*=.279†

Note: Rank indicates overall frequency ranking for total N in character strength codesa, and frequency rating for respondents who described any academic strengths for academic subject area code groupsb
†Non-significant Chi-square results comparing, 47,XXY, 47,XXX, and 47,XYY groups.
* Significant Chi-square results comparing 47,XXY, 47,XXX, and 47,XYY groups; p<.05

Codable answers included one-word responses, short phrases and lists, and multiple sentences describing the child.

Qualitative Findings

Qualitative analysis revealed two overarching themes: Social Strengths and Assets for Learning. Seven descriptive subcategories were developed directly from the qualitative codes and included constructs that aligned with preestablished character strengths from the literature as well as novel inductive categories, specific to our study sample and using the language provided by our participants. (see Table 3 for examples of parent quotes and codes that comprised each theme).

Social Strengths

Parents reported an overwhelming number of strengths that help their children to engage and connect with others; similar to the positive psychology virtue of Humanity (Niemiec & McGrath, 2019). Specifically, parents described features of extraordinary kindness and an eagerness to please.

Extraordinary kindness. Many parent respondents described their child as kind, loving, empathetic, or sweet. Parents described children greeting classmates each day with a smile, caring for younger children and animals, acting friendly and polite, using gentle behaviors, and sharing with others. Numerous respondents noted a strong social motivation in their children; a desire to connect with peers and make friends.

Eager to please. A common theme throughout survey responses was an eagerness to please teachers and friends. Children were often described as helpful in the classroom; team players who assisted classmates with work, supported peers, and aimed to please teachers through remarkably good behavior. Parents described empathy and thoughtful social overtures.

Assets for Learning

Parents described character strengths that help their children to acquire and apply their knowledge; analogous to the positive psychology virtue of Wisdom (Niemiec & McGrath, 2019). Parents described strengths in STEM, creative thinkers, a love for learning, and hardworking students.

Strengths in STEM. One quarter of all parents noted their child had marked strengths in STEM subjects. Math and science were particularly common, and parents described mathematical minds, abilities to build and design, and strong visual spatial reasoning skills that supported scientific inquiry.

Creative thinkers. Parents described creative children with good imaginations that encouraged interesting play and academic discoveries. Many noted 'outside the box' thinking; children who were able to solve problems in new and innovative ways. Further, the arts were frequently noted as areas of strength, particularly visual arts and music.

Loves to learn. A love of learning and curiosity were frequently reported along with descriptions of bright, smart, and intelligent children. A strong motivation to learn new things coupled with a genuine love of school, particularly when in a supportive environment, was common throughout the responses.

Hard-working students. Many parents described their children as industrious students who stuck with difficult tasks. Respondents used phrases such as 'never gives up' and 'tenacious' to describe a noteworthy

sense of persistence. Over thirty parents specifically used the words 'hard-working,' while many others referred to children who tried hard or gave maximum effort. Responses suggested a strong sense of energy and perseverance required to meet the challenges of the school day.

Quantitative Results

Statistical analysis revealed substantial overlap of strengths across all trisomy SCAs, with some unique patterns by condition. All parents volunteered responses that were coded as 'character strengths' as described in the positive psychology literature. Character strength code frequencies, adjusted for sample size, showed all trisomy SCAs had similarly high rates of strengths in perseverance and love of learning. Kindness was the most frequently coded character strength in boys with 47,XXY and girls with 47,XXX, and curiosity, teamwork, and humor were particular strengths in boys with 47,XYY (Figure 1). Fewer parents provided responses that were later coded as 'academic subject area strengths' (47,XXY=98, 47,XXX=19, 47,XYY=19). STEM was the most frequently coded academic strength for both the 47,XXY and 47,XYY groups and Fine Arts was the most frequently coded academic area for the 47,XXX group (Figure 2). Chi-square analysis (Table 4) revealed three codes with significant group differences: curiosity (p=.005), teamwork (p=.028), and STEM (p=.016). Post hoc pairwise comparisons revealed parents of boys with 47,XYY were more likely to describe behaviors coded as curious and teamwork, while parents of boys with 47,XXY were more likely to describe behaviors coded as STEM than girls with 47,XXX but not more than boys with 47,XYY. However, sample sizes for 47,XXX and 47,XYY were limited and small Chi-square and Fisher's exact cell sizes suggest results should be interpreted with caution.

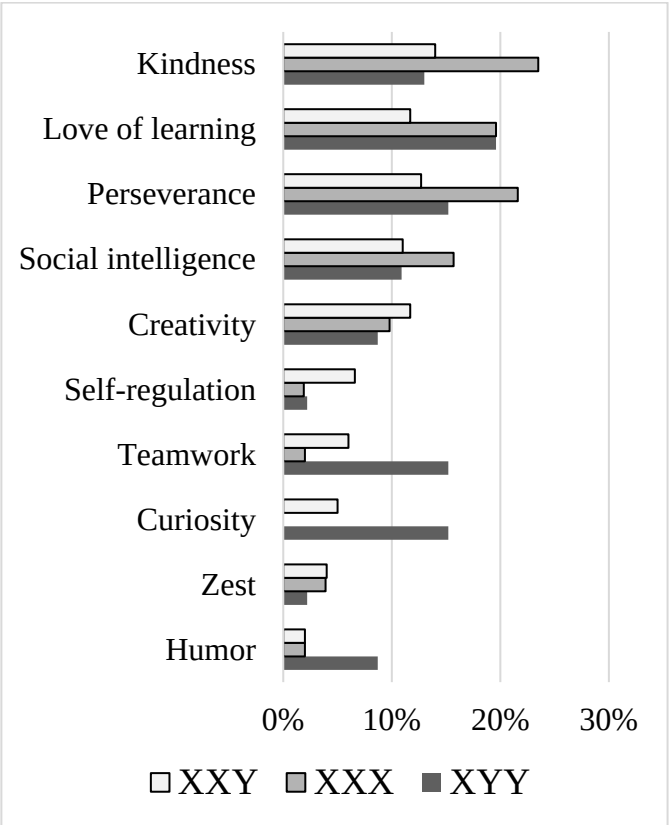

Figure 1. Top ten parent reported character strengths in trisomy SCAs

Note. Results are based on number of parent responses to a free-text question about child strengths. Rather than a traditional yes/no response about each trait, parents were asked to describe their child's strengths in an open-ended manner and responses were later coded for character strengths content. Responses could have more than one code assigned. X axis depicts frequencies of character strengths codes within each trisomy SCA group, adjusted for group sample sizes. For example, 27/197 parents (~14%) of boys with 47,XXY spontaneously described strengths that we later coded as 'Kindness' making it the most frequently assigned character strength code for the 47,XXY group.

Integrated Mixed Methods Results

Qualitative themes and categories were linked with statistical results in a joint display (interpretive matrix) (Table 5). Results from the two strands of analysis were mostly congruent, where thematic findings were supported by the frequencies of strengths codes and academic code groups. However, the qualitative themes did expand on some of the quantitative results by explaining *how* children with SCAs demonstrate specific character strengths. For instance, many students with SCAs demonstrated their strengths of social intelligence and teamwork through an eagerness to please teachers and classmates. Statistical comparisons expanded upon thematic findings for specific trisomy conditions; more frequent codes for STEM in boys with 47,XXY and more frequent codes for curiosity and teamwork in boys with 47,XYY elaborated upon the learning profiles and behavioral phenotypes for these two specific trisomy SCA conditions.

Discussion

This is the first study to systematically examine parent reported strengths in a relatively large, international sample of youth with SCAs. Parents described themes that aligned closely with VIA scales for character strengths, such as 'extraordinary kindness', 'creative thinking', and 'love for learning'.

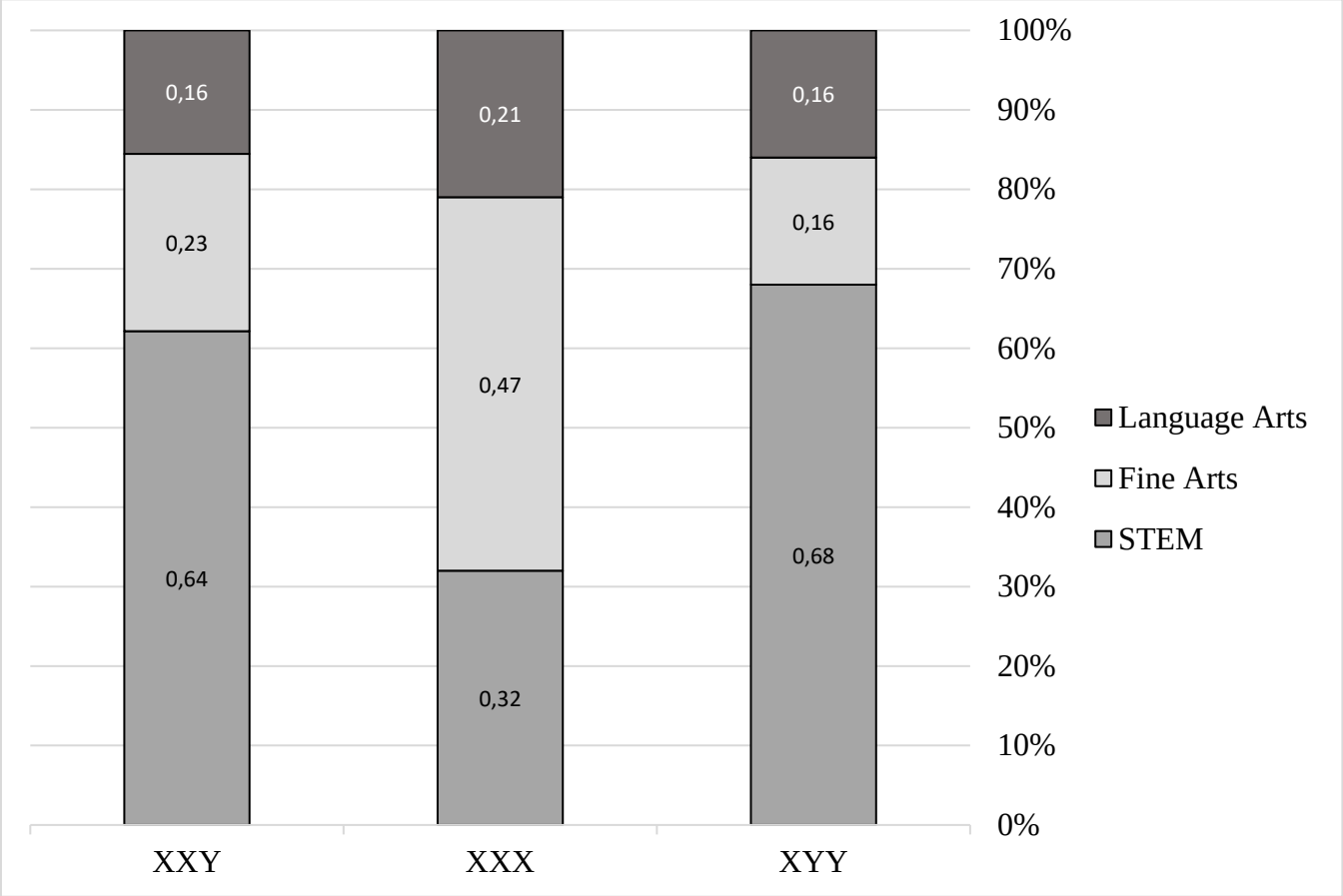

Figure 2. Parent reported academic strengths in trisomy SCAs

Note. Results are based on number of parent responses to a free-text question about child strengths. Rather than a traditional yes/no response about each trait, parents were asked to describe their child's strengths in an open-ended manner. The numbers of parents who spontaneously volunteered answers coded as academic strengths (as opposed to character or other strengths) were as follows: 47,XXY=98, 47,XXX=19, 47,XYY=19. Bars indicate percent of each academic strength codes assigned to each subject area across the trisomy SCA groups. For example, 63 of the 98 parents (64%) who provided academic subject area strengths in the 47,XXY group described strengths we coded as 'STEM', making it the most frequently coded academic subject area for the 47,XXY group

However, using an inductive open-ended survey question with open and in-vivo coding elicited some novel themes that may be unique to children with SCAs, such as 'eager to please', 'success in STEM', and 'hardworking students'. Across the trisomy conditions, parents described similar strengths profiles with frequent codes for love of learning and perseverance. However, as expected, some interesting genetic differences were identified. Specifically, kindness was the most frequently coded character strength for children with 47,XXY and 47,XXX, while teamwork, curiosity, and humor were more common for boys with 47,XYY. STEM subjects were the most commonly coded academic strengths for boys with 47,XXY and 47,XYY, while fine arts was most commonly reported for girls with 47,XXX.

Our findings are consistent in some ways with prior research on cognitive phenotypes associated with SCA conditions. However, the qualitative nature of our study allows for a richer and nuanced description than simply describing IQ scores, as many of our parents described assets for learning such as curiosity, love of learning, and creativity. Previous research on developmental outcomes in SCAs has documented that cognitive skills tend to fall within the average range and are a relative strength when compared with language and motor skills (Bender et al., 1986). Further, many parents in our sample indicated strengths in STEM-related academic subjects and a proclivity toward the arts. Prior research on neurocognitive profiles suggests students with SCAs have relatively higher visual-spatial skills, although these trends are typically presented as weaknesses in verbal reasoning rather than strengths in nonverbal reasoning as captured in our data (Leggett et al., 2010).

Interestingly, an overwhelming number of parents in our study reported their children possess social strengths, demonstrating empathetic, helpful, and caring behaviors. This contrasts with prior research focused on risk for pathology that has documented a clear pattern of psychosocial deficits in children and adults with SCAs, including impaired social cognition, increased neuroticism, introversion, and social isolation

(Freilinger et al., 2018; Skakkebaek et al., 2014; Skakkebæk et al., 2018; van Rijn, 2019). Similarly, our finding that parents described hardworking students with a courageous persistence was noteworthy in light of prior research documenting a passive, unassertive temperament and a lack of initiative for individuals with SCAs (Skakkebæk et al., 2020; van Rijn et al., 2006). Although our results do not disprove these well-documented risks, our study suggests that current assessments used in clinical and research settings may fail to capture the complete SCA phenotype, including this reported tendency toward extraordinary kindness and hard work. Further, supportive school environments, positive relationships with teachers, and intrinsically motivating tasks may encourage students with SCAs to apply their strengths in order to overcome challenges related to the diagnosis.

Limitations

This study was limited by the survey design, in that we were unable to directly assess child strengths or observe the application of child strengths. Our survey had an open URL and therefore we were unable to determine how many parents were provided the link but chose not to participate and thus cannot provide an overall response rate. Our findings may not reflect the full phenotype due to possible ascertainment bias in the sample. Further, we chose to use a "master coder/reliability coder" approach (Syed & Nelson, 2015), with a primary coder assigning the majority of codes used as data for the final analyses. Although this method has the potential for researcher bias, we utilized best practice in rigorous qualitative coding by ensuring that over one-quarter of all responses obtained a high level of ICA with highly trained co-coders (O'Connor & Joffe, 2020). Finally, although the sample size was more than adequate for a qualitative investigation, it was limited for statistical analysis, especially at the subgroup level where cell sizes were small, and comparisons reported are not generalizable to the general SCA population. Importantly, our findings of social strengths and assets for learning can be attributed, in part, to the nature of our question. By asking parents to comment specifically on their child's strengths in the educational setting, we may have narrowed the possible responses and prohibited parents from commenting on strengths more often noted outside the classroom such as transcendental strengths (e.g., spirituality, gratitude). Nevertheless, our findings support further examination of strengths profiles for children with SCAs to test whether or not our most frequently coded strengths (e.g., kindness, love of learning, perseverance, social intelligence, creativity) are commonly associated with the SCA phenotype and how these, and other strengths, may impact educational outcomes and improve overall quality of life.

Implications

Historically, SCA literature has called for early interventions to address a known pattern of risks for future school problems and behavioral health concerns in children and youth with SCAs (Thompson et al., 2020). Recommendations often include targeting known problem areas, such as language deficits, motor delays, learning disabilities, and psychosocial problems (van Rijn, 2019). While these interventions are undoubtably critical for facilitating independence and achievement, children with SCAs will also benefit from interventions that promote happiness and wellbeing. Research from the field of positive psychology has shown that working to amplify strengths rather than treating deficits can increase positive affect, decrease depressive symptoms, and enhance quality of life (Schutte & Malouff, 2019; Seligman et al., 2005).

For example, in the home setting, brief strengths-based parenting (SBP) interventions have been shown to improve both parent and adolescent mood and life satisfaction even when accounting for child introversion and neuroticism, two known areas of concern in SCA conditions (Freilinger et al., 2018; Jach et al., 2018; Skakkebæk et al., 2018; Waters et al., 2019). In our study, as found in prior research asking parents to openly describe their children (Park & Peterson, 2006), 377 parents described meaningful character strengths in their children with SCAs with very little guidance (e.g. providing examples or training on strengths). These findings suggest that facilitated SBP interventions may be feasible in the SCA population and merit further investigation.

In the school setting, strengths-based approaches have been shown to improve overall student wellbeing, enhance academic achievement, and build student capacity (Tejada-Gallardo et al., 2020). Strengths-based assessments (Buckley & Epstein, 2004) measuring child strengths through self, parent, and teacher rating forms have proven valid and reliable for students with and without learning disabilities (Shogren et al., 2018) and documenting strengths in a standardized manner might indirectly impact a child, as educators presented with psychoeducational reports with an added section on child strengths have been

shown to have a more hopeful attitude about a student's future and tend to predict higher levels of student success (Donovan & Nickerson, 2007). As children with SCAs are at risk for school problems, strengths-based approaches to education should be studied further.

Our results specifically indicate a pattern of potential signature strengths for children with SCAs that can be targeted and enhanced in daily life at home and at school. Students with particular strengths in kindness can be encouraged to purposely conduct daily prosocial acts (Nelson et al., 2016) or journal and reflect on their own kind behaviors at the end each day (Ko et al., 2021). Mindfulness interventions may be particularly effective for empathetic students who report strengths in social intelligence and an eagerness to please, as increased monitoring and awareness of self and others can improve social connections and overall mood (Lindsay et al., 2019; Teal et al., 2019). Those with a strong love of learning can be encouraged to dig deeper into topics of interest through student-centred, project-based learning (Kokotsaki et al., 2016) and creative students may benefit from extracurricular involvement in creative problem solving clubs (Greenberg, 2016; Missett et al., 2013) or receiving explicit encouragement to "be creative" with their school work (Nusbaum et al., 2014). Teachers and parents of students with SCAs who report signature strengths in perseverance can encourage a growth mindset, focusing positive feedback on effort and energy rather than final grades (Haimovitz & Dweck, 2016; Paunesku et al., 2015). Finally, prioritizing highly motivating STEM and fine arts coursework for students with SCAs who struggle with language arts may actually boost overall school performance and confidence, as the application of strengths is ultimately more energizing and fulfilling than remediation of weaknesses (Niemiec, 2017).

Conclusion

In summary, our study adds to the positive psychology, school psychology, and SCA bodies of literature by documenting that parents report a wide variety of academic and character strengths in their children with SCA conditions. Using a positive psychology approach provided fresh insight to the SCA phenotype and potential differences between the trisomy conditions, reframing prior research to highlight strengths as opposed to documenting profiles of deficits. Further, asking parents to comment in an open-ended format revealed novel, previously undocumented insights to children with SCAs as hard-working, curious, creative students with a love for learning, as well as kind, helpful, and caring personalities. There is an opportunity to further investigate the role of positive psychology concepts in other populations of students with rare genetic conditions. Natural history studies can document strengths in the phenotypes through standardized strengths-based assessments. Qualitative methods, such as field observations or photo elicitation, could be used to capture student voice and document strengths as children with rare conditions engage and interact in their school environments (Kozleski, 2017). Finally, strengths based assessments and positive psychology interventions should be used with students with rare conditions in educational settings in order to capitalize on strengths and improve school outcomes. By understanding and promoting the character assets of students with genetic differences, we may be able to protect against risk for future educational failure and psychological problems and ultimately enhance quality of life for children and their families.

Compliance with Ethical Standards

Ethical Standards

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Ethic approval has been obtained before conducting the research.

Declaration of Conflicting Interests

On behalf of all authors, the corresponding author states that there is no conflict of interest.

Funding

Eunice Kennedy Shriver National Institute of Child Health and Human Development, Grant/Award Numbers: K23HD092588, R01HD091251; National Center for Advancing Translational Sciences, Grant/Award Number: UL1 TR002535

Data Availability Statement

Data for this study are available upon reasonable request.

ORCID

Talia Thompson https://orcid.org/0000-0001-6512-9743

Received: February 25, 2021
Accepted: June 2, 2021
Published Online: June 17, 2021

References

Bender, B., Puck, M., Salbenblatt, J., & Robinson, A. (1986). *Cognitive development of children with sex chromosome abnormalities*. College Hill Press.

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.

Buckley, J. A., & Epstein, M. H. (2004). The Behavioral and Emotional Rating Scale-2 (BERS-2): Providing a comprehensive approach to strength-based assessment. *The California School Psychologist, 9*(1), 21-27.

Creswell, J. W., & Clark, V. L. P. (2017). *Designing and conducting mixed methods research*. Sage publications.

Donovan, S. A., & Nickerson, A. B. (2007). Strength-based versus traditional social-emotional reports: Impact on multidisciplinary team members' perceptions. *Behavioral Disorders, 32*(4), 228-237.

Feise, R. J. (2002). Do multiple outcome measures require p-value adjustment? *BMC medical research methodology, 2*(1), 1-4.

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

Greenberg, E. B. (2016). *Destination Imagination: An Examination of Highly Creative Children's Experiences on Their Journey Through Imagination* William James College].

Guetterman, T. C., Fetters, M. D., & Creswell, J. W. (2015). Integrating quantitative and qualitative results in health science mixed methods research through joint displays. *The Annals of Family Medicine, 13*(6), 554-561.

Haimovitz, K., & Dweck, C. S. (2016). Parents' views of failure predict children's fixed and growth intelligence mind-sets. *Psychological science, 27*(6), 859-869.

Hamerton, J. L., Canning, N., Ray, M., & Smith, S. (1975). A cytogenetic survey of 14,069 newborn infants. I. Incidence of chromosome abnormalities. *Clin Genet, 8*(4), 223-243.

Howard-Bath, A., Poulton, A., Halliday, J., & Hui, L. (2018, Dec). Population-based trends in the prenatal diagnosis of sex chromosome aneuploidy before and after non-invasive prenatal testing. *Prenat Diagn, 38*(13), 1062-1068. https://doi.org/10.1002/pd.5363

Jach, H. K., Sun, J., Loton, D., Chin, T.-C., & Waters, L. E. (2018). Strengths and subjective wellbeing in adolescence: Strength-based parenting and the moderating effect of mindset. *Journal of Happiness Studies, 19*(2), 567-586.

Ko, K., Margolis, S., Revord, J., & Lyubomirsky, S. (2021). Comparing the effects of performing and recalling acts of kindness. *The Journal of Positive Psychology, 16*(1), 73-81.

Kokotsaki, D., Menzies, V., & Wiggins, A. (2016). Project-based learning: A review of the literature. *Improving schools, 19*(3), 267-277.

Kozleski, E. B. (2017). The uses of qualitative research: Powerful methods to inform evidence-based practice in education. *Research and Practice for Persons with Severe Disabilities, 42*(1), 19-32.

Leggett, V., Jacobs, P., Nation, K., Scerif, G., & Bishop, D. V. (2010, Feb). Neurocognitive outcomes of individuals with a sex chromosome trisomy: XXX, XYY, or XXY: a systematic review. *Dev Med Child Neurol, 52*(2), 119-129. https://doi.org/DMCN3545 [pii] 10.1111/j.1469-8749.2009.03545.x

Linden, M. G., & Bender, B. G. (2002, Jun 1). Fifty-one prenatally diagnosed children and adolescents with sex chromosome abnormalities. *Am J Med Genet, 110*(1), 11-18. https://doi.org/10.1002/ajmg.10394

Lindsay, E. K., Young, S., Brown, K. W., Smyth, J. M., & Creswell, J. D. (2019). Mindfulness training reduces loneliness and increases social contact in a randomized controlled trial. *Proceedings of the National Academy of Sciences, 116*(9), 3488-3493.

Missett, T. C., Callahan, C. M., & Hertberg-Davis, H. (2013). Evaluating the impacts of destination ImagiNation on the creative problem solving skills of middle school students. *The International Journal of Creativity and Problem Solving, 23*(2), 97-112.

Nelson, S. K., Layous, K., Cole, S. W., & Lyubomirsky, S. (2016). Do unto others or treat yourself? The effects of prosocial and self-focused behavior on psychological flourishing. *Emotion, 16*(6), 850.

Niemiec, R. M. (2017). *Character strenghts interventions: A field guide for practitioners*. Hogrefe Publishing.

Niemiec, R. M., & McGrath, R. E. (2019). *The power of character strengths: Appreciate and ignite your positive personality*. VIA Institute on Character.

Nusbaum, E. C., Silvia, P. J., & Beaty, R. E. (2014). Ready, set, create: What instructing people to "be creative" reveals about the meaning and mechanisms of divergent thinking. *Psychology of Aesthetics, Creativity, and the Arts, 8*(4), 423.

O'Connor, C., & Joffe, H. (2020). Intercoder reliability in qualitative research: debates and practical guidelines. *International Journal of Qualitative Methods, 19*, 1609406919899220.

Park, N., & Peterson, C. (2006, 2006/09/01). Character Strengths and Happiness among Young Children: Content Analysis of Parental Descriptions. *Journal of Happiness Studies, 7*(3), 323-341. https://doi.org/10.1007/s10902-005-3648-6

Paunesku, D., Walton, G. M., Romero, C., Smith, E. N., Yeager, D. S., & Dweck, C. S. (2015, Jun). Mind-set interventions are a scalable treatment for academic underachievement. *Psychol Sci, 26*(6), 784-793. https://doi.org/10.1177/0956797615571017

Peterson, C., & Seligman, M. E. (2004). *Character strengths and virtues: A handbook and classification* (Vol. 1). Oxford University Press.

Riggan, K. A., Close, S., & Allyse, M. A. (2020). Family experiences and attitudes about receiving the diagnosis of sex chromosome aneuploidy in a child. American Journal of Medical Genetics Part C: Seminars in Medical Genetics,

Rosenthal, R. (1991). Teacher Expectancy Effects: A Brief Update 25 Years after the Pygmalion Experiment. *Journal of Research in Education, 1*(1), 3-12.

Ross, J. L., Zeger, M. P., Kushner, H., Zinn, A. R., & Roeltgen, D. P. (2009). An extra X or Y chromosome: contrasting the cognitive and motor phenotypes in childhood in boys with 47,XYY syndrome or 47,XXY Klinefelter syndrome. *Dev Disabil Res Rev, 15*(4), 309-317. https://doi.org/10.1002/ddrr.85

Rovet, J., Netley, C., Keenan, M., Bailey, J., & Stewart, D. (1996, Mar). The psychoeducational profile of boys with Klinefelter syndrome. *J Learn Disabil, 29*(2), 180-196.

Saldaña, J. (2015). *The coding manual for qualitative researchers*. Sage.

Schutte, N. S., & Malouff, J. M. (2019). The impact of signature character strengths interventions: A meta-analysis. *Journal of Happiness Studies, 20*(4), 1179-1196.

Seligman, M. E., Steen, T. A., Park, N., & Peterson, C. (2005). Positive psychology progress: empirical validation of interventions. *American Psychologist, 60*(5), 410.

Shogren, K. A., Shaw, L. A., Raley, S. K., Wehmeyer, M. L., Niemiec, R. M., & Adkins, M. (2018). Assessing character strengths in youth with intellectual disability: Reliability and factorial validity of the VIA-Youth. *Intellectual and developmental disabilities, 56*(1), 13-29.

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 & Child Neurology, 56*(4), 346-353.

Skakkebæk, A., Gravholt, C. H., Chang, S., Moore, P. J., & Wallentin, M. (2020). Psychological functioning, brain morphology, and functional neuroimaging in Klinefelter syndrome. American Journal of Medical Genetics Part C: Seminars in Medical Genetics,

Skakkebaek, A., Gravholt, C. H., Rasmussen, P. M., Bojesen, A., Jensen, J. S., Fedder, J., Laurberg, P., Hertz, J. M., Ostergaard, J. R., Pedersen, A. D., & Wallentin, M. (2014). Neuroanatomical correlates of Klinefelter syndrome studied in relation to the neuropsychological profile. *Neuroimage Clin, 4*, 1-9. https://doi.org/10.1016/j.nicl.2013.10.013

Skakkebæk, A., Moore, P. J., Pedersen, A. D., Bojesen, A., Kristensen, M. K., Fedder, J., Hertz, J. M., Østergaard, J. R., Wallentin, M., & Gravholt, C. H. (2018). Anxiety and depression in Klinefelter syndrome: The impact of personality and social engagement. *PLoS One, 13*(11), e0206932.

Syed, M., & Nelson, S. C. (2015). Guidelines for establishing reliability when coding narrative data. *Emerging Adulthood, 3*(6), 375-387.

Tartaglia, N., Ayari, N., Howell, S., D'Epagnier, C., & Zeitler, P. (2011, Jun). 48,XXYY, 48,XXXY and 49,XXXXY syndromes: not just variants of Klinefelter syndrome. *Acta Paediatr, 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., Boada, C., Alston, A., Crawford, L., & 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*.

Tartaglia, N. R., Ayari, N., Hutaff-Lee, C., & Boada, R. (2012, May). Attention-deficit hyperactivity disorder symptoms in children and adolescents. with sex chromosome aneuploidy: XXY, XXX, XYY, and XXYY. *Journal of Developmental and Behavioral Genetics, 33*(4), 309-318.

Tartaglia, N. R., Wilson, R., Miller, J. S., Rafalko, J., Cordeiro, L., Davis, S., Hessl, D., & Ross, J. (2017, Apr). Autism Spectrum Disorder in Males with Sex Chromosome Aneuploidy: XXY/Klinefelter Syndrome, XYY, and XXYY. *J Dev Behav Pediatr, 38*(3), 197-207. https://doi.org/10.1097/dbp.0000000000000429

Teal, C., Downey, L. A., Lomas, J. E., Ford, T. C., Bunnett, E. R., & Stough, C. (2019). The role of dispositional mindfulness and emotional intelligence in adolescent males. *Mindfulness, 10*(1), 159-167.

Tejada-Gallardo, C., Blasco-Belled, A., Torrelles-Nadal, C., & Alsinet, C. (2020). Effects of School-based Multicomponent Positive Psychology Interventions on Well-being and Distress in Adolescents: A Systematic Review and Meta-analysis. *Journal of Youth and Adolescence*, 1-18.

Thompson, T., Howell, S., Davis, S., Wilson, R., Janusz, J., Boada, R., Pyle, L., & 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*.

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. (2019, Mar). A review of neurocognitive functioning and risk for psychopathology in sex chromosome trisomy (47,XXY, 47,XXX, 47, XYY). *Curr Opin Psychiatry, 32*(2), 79-84. https://doi.org/10.1097/yco.0000000000000471

van Rijn, S., Swaab, H., Aleman, A., & Kahn, R. S. (2006, Jun). X Chromosomal effects on social cognitive processing and emotion regulation: A study with Klinefelter men (47,XXY). *Schizophr Res, 84*(2-3), 194-203. https://doi.org/10.1016/j.schres.2006.02.020

Waters, L. E., Loton, D., & Jach, H. K. (2019). Does strength-based parenting predict academic achievement? The mediating effects of perseverance and engagement. *Journal of Happiness Studies, 20*(4), 1121-1140.