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Comparison of gut microbiota in autism spectrum disorders and neurotypical boys in China: A case-control study

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ABSTRACT

Background: Autism spectrum disorders (ASDs) are a set of complex neurobiological disorders. Growing evidence has shown that the microbiota that resides in the gut can modulate brain development via the gut–brain axis. However, direct clinical evidence of the role of the microbiota–gut–brain axis in ASD is relatively limited.
Methods: A case-control study of 71 boys with ASD and 18 neurotypical controls was conducted at China-Japan Friendship Hospital. Demographic information and fecal samples were collected, and the gut microbiome was evaluated and compared by 16S ribosomal RNA gene sequencing and metagenomic sequencing.
Results: A higher abundance of operational taxonomic units (OTUs) based on fecal bacterial profiling was observed in the ASD group. Significantly different microbiome profiles were observed between the two groups. At the genus level, we observed a decrease in the relative abundance of *Escherichia, Shigella, Veillonella, Akkermansia, Provindencia, Dialister, Bifidobacterium, Streptococcus, Ruminococcaceae UCG_002, Megasphaera, Eubacterium_coprostanol, Citrobacter, Ruminiclostridium_5, and Ruminiclostridium_6* in the ASD cohort, while *Eisenbergiella, Klebsiella, Faecalibacterium,* and *Blautia* were significantly increased. Ten bacterial strains were selected for clinical discrimination between those with ASD and the neurotypical controls. The highest AUC value of the model was 0.947.
Conclusion: Significant differences were observed in the composition of the gut microbiome between boys with ASD and neurotypical controls. These findings contribute to the knowledge of the alteration of the gut microbiome in ASD patients, which opens the possibility for early identification of this disease.

Introduction

Autism spectrum disorders (ASDs) are a set of complex neurobiological disorders that impair social interactions and communication and lead to restricted, repetitive, and stereotyped patterns of behavior, interests, and activities [1]. The prevalence of ASD has been steadily increasing in recent years, which may to some extent be due to greater awareness of the disease by health and education professionals and changes in diagnostic criteria. Treatments and educational interventions usually last for the entire lives of those who are diagnosed with ASD [2]. This represents a serious public health problem due to its increasing prevalence and huge financial costs.

ASD are etiologically heterogeneous. However, the exact causes of ASD are unclear and are believed to involve a combination of genetic and environmental risk factors. Many studies highlight the possibility of environmental risk factors and associated medical comorbidities that

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contribute to the disease. Some studies have even observed an association between ASD severity and gastrointestinal (GI) symptoms, such as constipation, diarrhea, and alternating constipation/diarrhea [3,4].

In recent years, growing evidence has shown that the microbiota that resides in the gut can modulate brain development via the gut–brain axis [5]. As far as this concept is concerned, increasing interest has developed focusing on the potential effects of the microbiota–gut–brain axis in neurodevelopmental disorders, especially in the etiology and pathogenesis of ASD.

In an attempt to clarify the role of gut microbiota in the appearance and development of ASD, some clinical studies have reported that compared with neurotypical controls, subjects with autism suffer from altered profiles and abundances of gut microbiota [6–8]. The human gut harbors up to 100 trillion microorganisms, including at least 1000 different species of known bacteria [9]. Colonization of the gut microbiota starts at the moment of birth, when the newborn is exposed to a complex microbiota during delivery [10]. Many ASD-affected children undergo extensive oral antibiotic treatment before 3 years of age [11], which is thought to cause the proliferation of anaerobic bacteria, destabilize gut microbiota [12] and provide opportunities for competitive potential pathogens to contribute to ASD [13,14]. *Bacteroidetes, Clostridia*, and *Desulfovibrio* are common bacteria that may promote GI symptoms and autistic behaviors. In addition to altering the intestinal immune system, these bacteria also produce certain potent neurotoxins that directly contribute to the pathologies of ASD.

Several findings support a microbiota–gut–brain connection in a mouse model of ASD. However, direct clinical evidence for the role of the microbiota–gut–brain axis in ASD is relatively limited, and consensus across studies has not yet been established. The sample size of previous studies has been relatively small since recruitment is difficult. To better understand the microbial profiles associated with ASD in the context of the Chinese diet, we conducted this case-control study including 71 ASD children and 18 neurotypical controls, representing the largest sample size of the gut microbiota of children with autism in China.

Methods and materials

Participants

This study was reviewed and approved by the Institutional Review Board of China-Japan Friendship Hospital. All written informed consent was obtained from the parents. The parents and the enrolled participants visited the Department of Pediatrics, China-Japan Friendship Hospital, and were provided the questionnaire data sheets. The fecal samples were obtained within three days after the visit.

According to the results of the Autism Behavior Checklist (ABC) screening tool of ASD and the Diagnostic and Statistical Manual, Fifth Edition (DSM-V) diagnostic criteria [15], the neuropsychiatric status of the affected subjects was established by two experienced experts in the Department of Pediatrics of China-Japan Friendship Hospital. The controls were evaluated in the same manner as the ASD subjects to exclude those with any developmental disturbances, including ASD.

Enrollment criteria

Autism group

1) Age 3 to 6.
2) Male.
3) ASD diagnosed according to the ABC screening tool and DSM-V criteria.
4) No usage of any type of antibiotic medications or probiotics within at least one month prior to the sample collection.

Control group

1) Age and sex-matched neurotypical healthy subjects.

Exclusion criteria

1) Treated with anti-inflammatory or antioxidant drugs.
2) Diagnosed with neurological diseases, severe head injuries, and gastrointestinal diseases such as celiac disease, chronic diarrhea, and chronic constipation, etc.

Sample collection, DNA extraction, and sequencing

Fecal specimens from participants were collected with sterilized 2-ml tubes containing pure ethanol, aliquoted, and frozen at 80 °C until DNA extraction. Total DNA extraction from fecal samples (250 mg, wet weight) was performed using a PSP Spin StooL DNA Kit/PSP Spin StooL DNA PLus Kit according to the manufacturer's instructions. Bacterial 16S rRNA amplicon sequencing of the V1–V2 gene region [16,17] was performed on the Illumina MiSeq platform for identification and relative quantification of bacterial taxa.

Metagenomic sequencing was also performed in this study [18], which can analyze the genetic composition and function of the microbial population in a specific environment and identify which strains are contained in the mixed environment. Metagenomic sequencing and technical advances have enabled culture-free, high-resolution strain and subspecies analyses at high throughput and in complex environments [19]. During metagenomic sequencing, 20 μg of microbial metagenomic DNA was taken, and a sequencing library was constructed using the NEXTflex® Rapid DNA Sequencing Kit (Bioo Scientific, USA, NOVA-5144-02) and following the manufacturer's instructions. After the library was constructed, Qubit 2.0 was used for preliminary quantification, the library was diluted to 2 ng/μL, Agilent 2100 was used to detect the insert size of the library, and the qPCR method [20] was used to accurately quantify the effective concentration of the library (the library was effective concentration> 3 mmol/L) to ensure the quality of the library. The constructed library was sequenced on the Illumina HiSeq 2500 platform. The study utilized MOCAT [21] preprocessing to filter low-quality sequences, sequences that were too short, and error sequences to obtain clean data. Starting from the clean data after quality control of each sample, metagenomic assembly was carried out [22]. Starting from the gene catalog, bowtie was used to compare the sequence with the MicroNR library to obtain the species annotation information of unigenes, and the gene abundance table was combined to obtain the species abundance table of different classification levels.

Statistical analysis

All statistical analyses were performed using SPSS 20.0. Several types of statistical analyses were carried out, depending on the research question being addressed. Continuous data among the groups were analyzed by using ANOVA (generalized linear model). Frequencies were calculated, and a chi-square test was performed to examine the frequency differences among the groups. The calculation of P values was performed with Kruskal–Wallis H-tests and Welch's t-tests. The P values were corrected by false discovery rates (FDRs) to control for multiple hypothesis testing. Principal coordinates were computed for the unweighted distance matrices and used to generate principal coordinate analysis (PCoA) plots using evenly sampled OTU abundances. According to the QIIME (Quantitative Insights into Microbial Ecology) tutorial (http://qiime.org/), high-throughput sequencing analysis of bacterial rRNA genes was processed using the QIIME (version 1.9.1) software suite [23].

Alpha diversity metrics from the final OTU table without singletons were obtained within the QIIME pipeline. Linear discriminant effect size (LEfSe) analysis was used to explore potential bacterial biomarkers

Table 1
Demographics of the children with ASD and the controls.

	ASD group (n = 71)	Control group (n = 18)	P-value
Sex (Male/Female)	71/0	18/0	1.000
Age (years, mean ± SD)	4.28 ± 1.52	4.62 ± 1.29	P = 0.792
ABC score	79.82 ± 13.10	8.77 ± 7.29	P < 0.001

associated with different groups. LEfSe combines the Kruskal-Wallis test pairwise Wilcoxon rank-sum test with linear discriminant analysis (LDA). It ranks features by effect size, which places features that explain most of the biological differences at the top. Relative operating characteristic (ROC) curves and area under the curve (AUC) values were used to evaluate the performance of the predictive model.

To further distinguish between ASD and their comparisons, we used a machine learning method based on the random forest algorithm to build a classifier from the same input feature set. Random forest classifier is a kind of ensemble learning algorithm belonging to bagging. It can establish an unbiased estimate of the error during the generation of the tree. Random forest classifier was found to be more accurate than both the alternative cross-validation-based estimator of the underlying algorithm's error [24].

Species relative abundances and patient profiles were analyzed using the random forest package in R [25,26]. A forest was trained by supervised learning. In the forest, each tree finds an ideal split for a set of randomly chosen features that predict the outcome of each sample as the expected outcome. The data partition split by every tree in a forest is used to vote on a predicted overall outcome of each sample. Every tree was used to vote on an outcome to prevent any single tree that may have memorized the data from having a dominant prediction. In this study, outcomes were children with ASD or neurotypical children. For all analyses, differences were considered statistically significant when the FDR corrected P value was <0.05.

Results

Demographics of the children with ASD and the controls

This study enrolled a total of 71 children with ASD and 18 controls. The demographic characteristics of the study participants are listed in Table 1. Age and sex were not significantly different between the two groups (P > 0.05).

Altered gut microbiome profile

We compared the alpha diversity of the gut microbiota in the two groups. As shown in Fig. 1, significant increases in bacterial richness, including observed species and the Shannon and Simpson diversity indices (all P values < 0.05), were observed in the ASD group; no significant variations between the two groups were observed for the Chao1 diversity index.

The total distribution of bacteria demonstrated that the taxonomy was significantly different at the phylum level, with increases in *Firmicutes, Proteobacteria,* and *Actinomycetes* and decreases in *Bacteroidetes* in the ASD group (all P values < 0.05). Notably, the *Bacteroidetes/Firmicutes* ratio was significantly lower in the ASD group (P value < 0.001).

LEfSe analysis showed significantly different microbiome profiles in the two groups. A histogram of the LDA scores was computed, which indicated the effect size of each differentially abundant taxon. At the genus level, significantly higher *Escherichia, Shigella, Veillonella,*

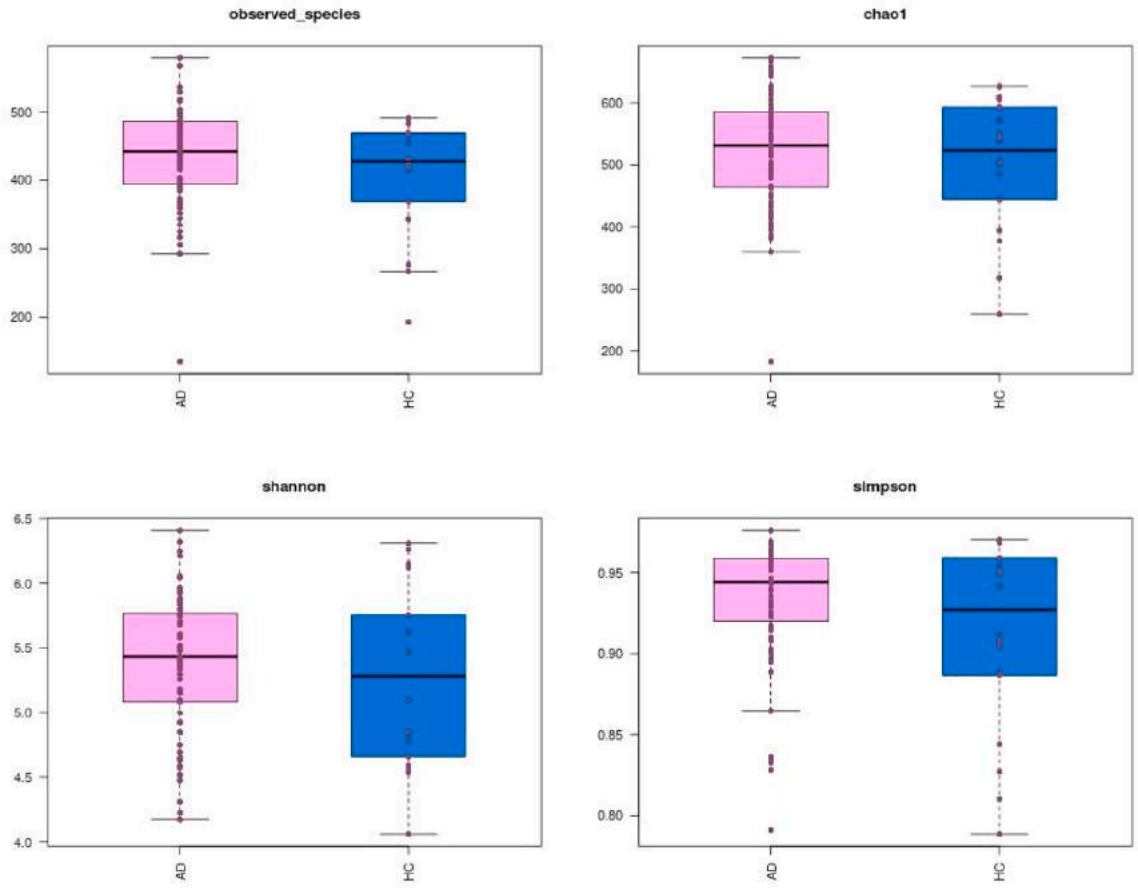

Fig. 1. Comparison of diversity index of two groups on the OTU profile.

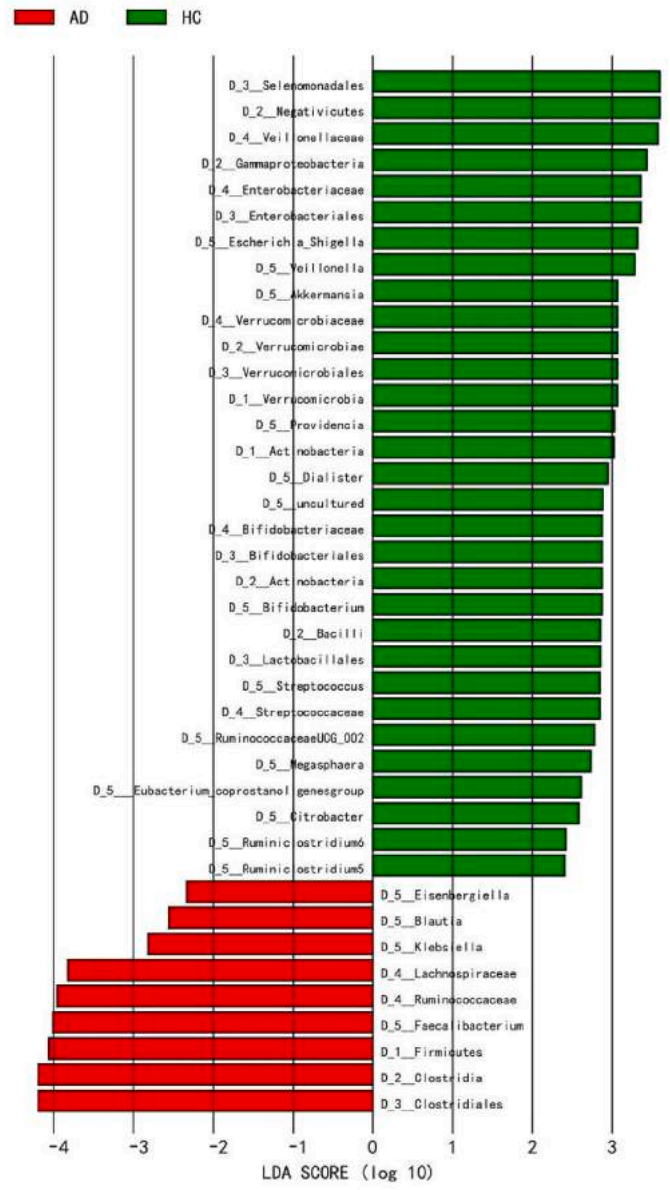

Fig. 2. LEfSe analysis between two groups.

Akkermansia, Providencia, Dialister, Bifidobacterium, Streptococcus, Ruminococcaceae UCG_002, Megasphaera, Eubacterium_coprostanol, Citrobacter, Ruminiclostridium_5, and *Ruminiclostridium_6* were observed in the control group, while *Eisenbergiella, Klebsiella, Faecalibacterium,* and *Blautia* were higher in the ASD group (all P value < 0.05) as shown in Fig. 2.

We further performed PCoA analysis of the bacterial beta diversity. As shown in Fig. 3, based on weighted UniFrac distances, the microbiome of the ASD group was distinct from that of the control group.

Consistent with the 16S rRNA analysis at the genus level, most of the identified differential species belonged to the genus *Clostridium*. We constructed a stochastic forest machine algorithm to build a prediction model of the identified bacterial strains for clinical discrimination (ASD vs. comparison group). Ten bacterial strains, namely, *Prevotella buccae, Bifidobacterium longum, Streptococcus thermophilus, Enterobacter cloacae, Klebsiella oxytoca, Eubacterium hallii, Clostridium ramosum, Erysipelotrichaceae bacterium 6_1_45, Eubacterium siraeum,* and *Lautropia mirabilis*, were selected to predict the risk of ASD in children (Fig. 4). The ROC curves are presented in Fig. 4 (Right). The highest AUC value was 0.947, with a sensitivity of 0.818 and specificity of 0.948.

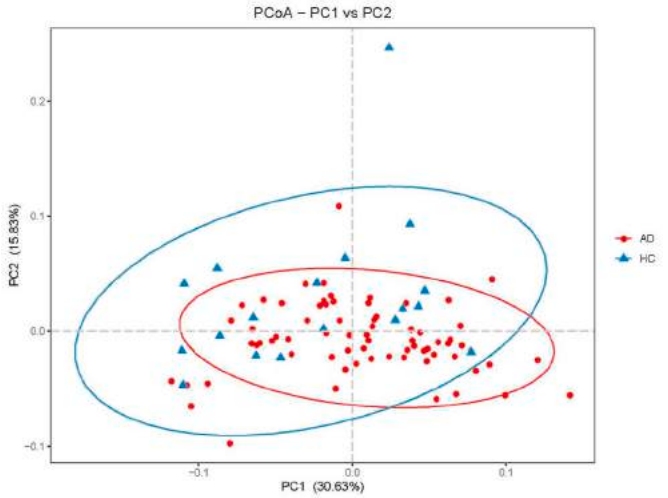

Fig. 3. PCoA of bacterial diversity based on weighted distance.

We utilized sample from an independent cohort to verify the random forest classifier derived from this study. The independent cohort include 15 asd children, with a mean age 4.17 ± 0.89 (range 3–6). Results showed that the *Streptococcus* , Ruminiclostridium_5 , Dialister , *Escherichia_Shigella* was significantly lower in affected group than those in control group, which was consistent with the results of this study population, as shown in Fig. 5.

Discussion

ASD is a complex neurodevelopmental disorder. The microbiota plays a key role in regulating normal host metabolism, physiology, nutrition, and even brain function. Emerging evidence has revealed that the gut microbiota profile [6–9] in individuals with ASD is different from that in neurotypical populations. To explore the profile of gut microbiota in ASD children, the current study enrolled 71 ASD children and 18 neurotypical controls. The sample size of this study is the largest of its kind in China.

Existing studies have revealed alterations in gut microbiota in ASD individuals compared with neurotypical controls. Consistent with previous clinical studies [27–29], the results of this study demonstrated a higher abundance of OTUs in fecal bacterial profiling in ASD patients.

The alterations in gut microbiota were related to the intensity of autistic symptomatology, indicating that bacterial metabolites may be involved in the development and severity of ASD [6]. To some extent, these findings verified the “gut–brain microbiome axis” concept, which assumes that there is a bidirectional interaction between gut microbiota and the brain. However, it is considered that this interaction is caused by hormones and neurotransmitters from the gut endocrine system. However, the specific underlying mechanisms have not yet been elucidated.

Consistent with the most recently published articles on the gut microbiome of Chinese ASD children [30], ASD patients exhibited a decreased *Bacteroidetes/Firmicutes* ratio.

Consistent with a previous study, *Ruminococcus* and *Faecalibacterium* were more abundant in ASD children than in the controls [31]. Studies have revealed that *Faecalibacterium* is related to the up- and down-regulation of some genes involved in the expression of interferon (IFN) gamma. IFN is a cytokine related to ASD that is exposed during fetal development [32]. As an underlying mechanism, IFN-gamma may play an indirect role in brain plasticity and synapse formation [33]. However, its connection with the pathogenesis of autism remains to be further investigated.

The percentage of *Dialister* was higher in neurotypical children, in line with previous studies. However, the evidence from a previous study was not enough to observe a statistically significant difference [34]. This

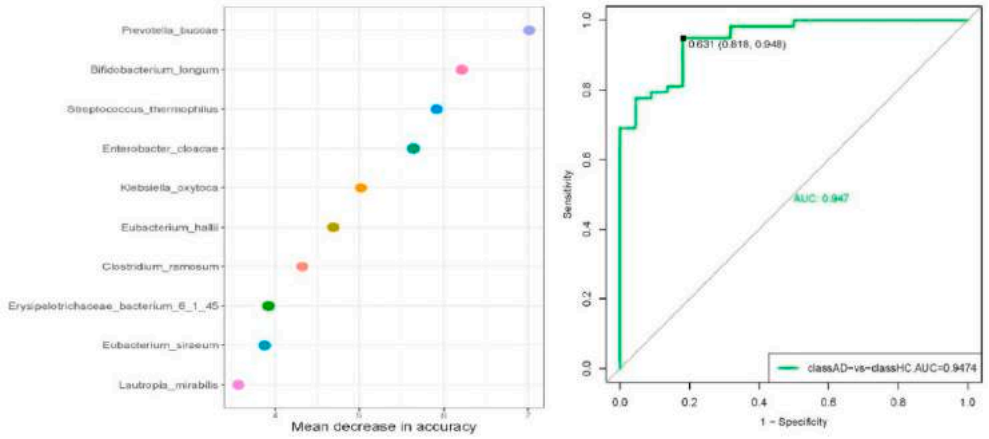

Fig. 4. Prediction model constructed by stochastic forest machine algorithm (Left) and ROC curves for clinical discrimination (Right).

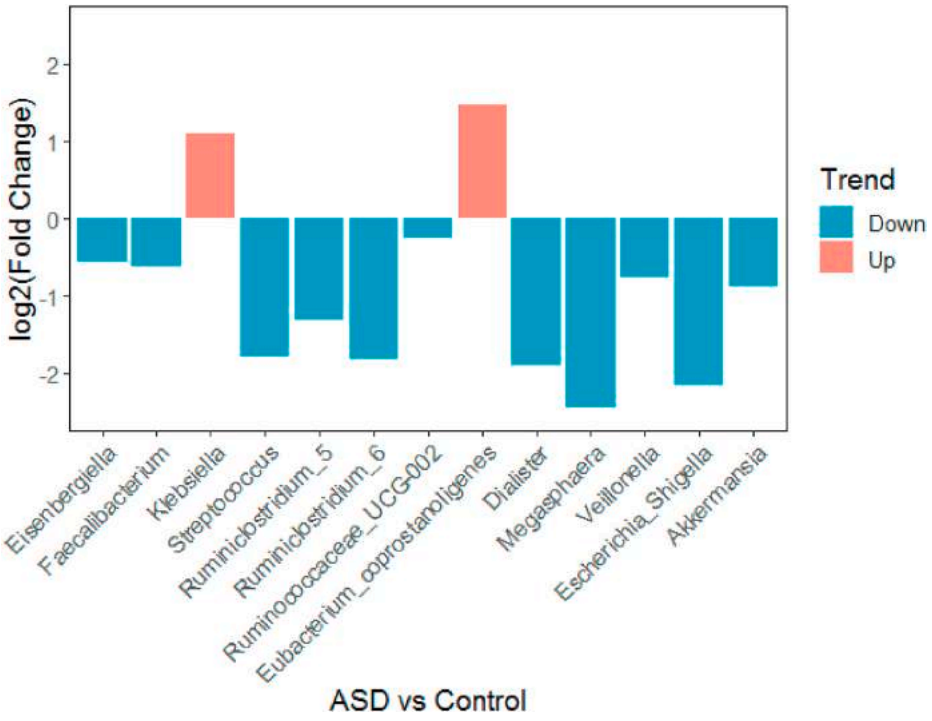

Fig. 5. Fold change of mean relative abundance for ASD versus HC.

study reported a lower abundance of protective bacteria, such as *Bifidobacterium*. A number of studies have shown that some species of *Bifidobacterium* produce GABA (gamma aminobutyric acid) [35], the concentrations of which have been found to be low in ASD children. GABA is highly related to glutamate metabolism, which is the main excitatory neurotransmitter in the central nervous system [36]. Some studies have revealed that lower glutamate concentrations correlate with the severity of social and behavioral symptoms of ASD [37].

Consistent with recent studies, higher *Veillonella, Akkermansia, and Providencia* were observed in neurological children than in the ASD group. *Veillonella* colonizing the intestine may supplement the Cori cycle by providing another lactic acid treatment method, thereby converting systemic lactic acid into SCFAs and re-entering the circulation [38]. *Akkermansia* induces immunoglobulin G1 (IgG1) antibodies and antigen-specific T cell responses, metabolic modulation, immune regulation and gut health protection [39]. Studies have shown that *Akkermansia muciniphila* can be a next-generation probiotic [40]. Members of the *Provindencia* family have been considered pathobionts, with the *Providencia stuartii* species being the dominant taxon. These strains swarm on semisolid (viscous) surfaces and adhere to and invade host cells, determining the specificity of disease pathogenesis and therapy [41].

The current study found an increase in *Faecalibacterium* and *Klebsiella. Faecalibacterium prausnitzii* is a late colonizer of the healthy human gut and a major butyrate producer [42,43]. *Klebsiella* is gaining recognition as a cause of several human infections. Recent studies of carbapenemase-producing and colistin-resistant strains demonstrate a potential reservoir of multidrug-resistant genes [44].

To our knowledge, this is the first study using the random forest algorithm to build a classifier to identify the characteristics of the gut microbiome of children with ASD, which is important for the early assessment of ASD risk. In addition, the new approach of diagnosis classifiers based on the AUC value makes it possible to plan personalized treatment and prevention strategies for ASD via microbiota modulation. Restoring the balance of the microbiota–gut–brain axis offers promising beneficial therapeutic effects on autistic deficits; however, direct clinical

evidence for a role of the microbiota–gut–brain axis in ASD is relatively limited. In the future, more randomized double-blind clinical studies are required to determine the role of the microbiota–gut–brain axis in the etiology of ASD. As a potential mediator of risk factors in ASD, the microbiota is positioned at the intersection between the environment and genes. The composition and function of the microbiome are dependent on genetic background and influenced by environmental factors, including feeding patterns, age, infection, antibiotic treatment, etc. Further study could focus on the mediating effect of the microbiome on the development of ASD.

Conclusions

Significant differences were observed in the composition of the gut microbiome between boys with ASD and neurotypical controls. These findings contribute to knowledge of the alteration of the gut microbiome in ASD patients, which opens the possibility for early identification of this disease.

Availability of data and materials

The datasets used and/or analyzed during the current study are available as supplemental materials.

Declaration of competing interest

The authors declare that they have no competing interests.

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Abbreviations

ABC	Autism Behavior Checklist
ANOVA	Analysis of Variance
ASD	Autism spectrum disorders
AUC	Area Under Curve
DNA	DeoxyriboNucleic Acid
DSM-V	Diagnostic and Statistical Manual, Fifth Edition
FDR	False Discovery Rates
GABA	Gamma aminobutyric acid
GI	gastrointestinal
IFN	interferon
LDA	Linear Discriminant Analysis
LEfSe	Linear discriminant effect size
OTUs	Operational Taxonomic Units
PCoA	Principal coordinate Analysis
QIIME	Quantitative Insights into Microbial Ecology
RNA	Ribonucleic Acid
ROC	Relative Operating Characteristic
SPSS	Statistical Product and Service Solutions

Ethical statement and consent to participate

The study protocol and data collection instruments were approved by the ethical review boards of China-Japan Friendship Hospital. Informed consent was obtained prior to interviews.

Consent for publication

Not applicable.

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Authors' contributions

FY, XYG, PFX, DNH, and QZ coordinated the study, participated in the study design, supervision, and data collection. ZYW, SMC, GCL, ZTL, and LMW conducted the experiment and data analysis. FY, XYG, and ZYW participated in the drafting of the manuscript. PFX, QZ and LMW coordinated the study, participated in study design, provided supervision, and contributed to the drafting of the final manuscript. All the authors read and approved the final manuscript.

References

[1] World Health Organization. International statistical classification of diseases and health related problems, Tenth Revision (ICD-10). Geneva: World Health Organization; 2004.
[2] Cdc. What is autism spectrum disorder?. Available online: https://www.cdc.gov/ncbddd/autism/facts.html. [Accessed 14 March 2020].
[3] Hsiao EY. Gastrointestinal issues in autism spectrum disorder. Harv. Rev. Psychiatr. 2014;22(2):104–11.
[4] McElhanon BO, McCracken C, Karpen S, Sharp WG. Gastrointestinal symptoms in autism spectrum disorder: a meta-analysis. Pediatrics 2014;133(5):872–83.
[5] Martin CR, Osadchiy V, Kalani A, Mayer EA. The brain-gut-microbiome Axis. Cell Mol. Gastroenterol. Hepatol. 2018;6:133–48.
[6] Xu M, Xu X, Li J, Li F. Association between gut microbiota and autism spectrum disorder: a systematic review and meta-analysis. Front Psychiatr 2019;10:473.
[7] Son JS, Zheng LJ, Rowehl LM, Tian X, Zhang Y, Zhu W, et al. Comparison of fecal microbiota in children with autism spectrum disorders and neurotypical siblings in the simons simplex collection. PloS One 2015;10:E0137725.
[8] Fattorusso A, Di Genova L, Dell'isola GB, Mencaroni E, Esposito S. Autism spectrum disorders and the gut microbiota. Nutrients 2019;11:521.
[9] Bermon S, Petriz B, Kajeniene A, Prestes J, Castell L, Franco OL. The microbiota: an exercise immunology perspective. Exerc Immunol Rev 2015;21:70–9.
[10] Di Mauro A, Neu J, Riezzo G, Raimondi F, Martinelli D, Francavilla R, et al. Gastrointestinal function development and microbiota. Ital J Pediatr 2013;39:15–22.
[11] Lopez CA, Kingsbury DD, Velaz-quez EM, Baumler AJ. Collateral damage: microbiota-derived metabolites and immune function in the antibiotic era. Cell Host Microbe 2014;16:156–63.
[12] Niehus R, Lord C. Early medical history of children with autism spectrum disorders. J Dev Behav Pediatr 2006;27:S120–7.
[13] Finegold SM, Dowd SE, Gontcharova V, Liu C, Henley KE, Wolcott RD, et al. Pyrosequencing study of fecal microflora of autistic and control children. Anaerobe 2010;16:444–53.
[14] Williams BL, Hornig M, Parekh T, Lipkin WI. Application of novel PCR-based methods for detection, quantification, and phylogenetic characterization of *Sutterella* species in intestinal biopsy samples from children with autism and gastrointestinal disturbances. mBio 2012;3:e00261. 11.
[15] Hudziak JJ, Novins DK. Proposed criteria for autism spectrum disorder in the DSM-5. J Am Acad Child Adolesc Psychiatry 2012;51:343.
[16] Edwards U, Rogall T, Blöcker H, Emde M, Böttger EC. Isolation and direct complete nucleotide determination of entire genes. Characterization of a gene coding for 16S ribosomal RNA. Nucleic Acids Res 1989;17:7843–53.
[17] Fierer N, Hamady M, Lauber CL, Knight R. The influence of sex, handedness, and washing on the diversity of hand surface bacteria. Proc Natl Acad Sci USA 2008;105:17994–9.
[18] Franzosa EA, Morgan XC, Segata N, Waldron L, Reyes J, Earl AM, Giannoukos G, Boylan MR, Ciulla D, Gevers D, Izard J, Garrett WS, Chan AT, Huttenhower C. Relating the metatranscriptome and metagenome of the human gut[J]. Proc Natl Acad Sci U S A 2014;111(22):E2329–38.
[19] Van Rossum T, Ferretti P, Maistrenko OM, Bork P. Diversity within species: interpreting strains in microbiomes. Nat Rev Microbiol 2020 Sep;18(9):491–506.
[20] Mehta B, Venables S, Roffey P. Comparison between magnetic bead and qPCR library normalisation methods for forensic MPS genotyping[J]. Int J Leg Med 2017;132(3):1–8.
[21] Kultima JR, Sunagawa S, Li J, et al. MOCAT: a metagenomics assembly and gene prediction toolkit[J]. PloS One 2012;7(10):e47656.
[22] Luo R, Liu B, Xie Y, et al. SOAPdenovo2: an empirically improved memory-efficient short-read de novo assembler[J]. GigaScience 2012;1(1):18.

[23] Caporaso JG, Kuczynski J, Stombaugh J, Bittinger K, Bushman FD, Costello EK, et al. QIIME allows analysis of high-throughput community sequencing data. Nat Methods 2010;7:335–6.
[24] Wolpert DH, Macready WG. An efficient method to estimate bagging's generalization error[J]. Mach Learn 1999;35(1):41–55.
[25] Breiman L. Random forests. Mach Learn 2001;45:5–32.
[26] Liaw A. Classification and regression by random forest. R News 2002;2:18–22.
[27] Finegold SM, Molitoris D, Song Y, Liu C, Vaisanen ML, Bolte E, et al. Gastrointestinal microflora studies in late-onset autism. Clinical infectious diseases : an official publication of the Infectious Diseases. Society of Americavol. 35; 2002. p. S6–16.
[28] Song Y, Liu C, Finegold SM. Real-time PCR quantitation of *clostridia* in feces of autistic children. Appl Environ Microbiol 2004;70:6459–65.
[29] Parracho HM, Bingham MO, Gibson GR, McCartney AL. Differences between the gut microflora of children with autistic spectrum disorders and that of healthy children. J Med Microbiol 2005;54:987–91.
[30] Kang DW, Park JG, Ilhan ZE, Wallstrom G, Labaer J, Adams JB, et al. Reduced incidence of *Prevotella* and other fermenters in intestinal microflora of autistic children. PloS One 2013;8:e68322.
[31] Inoue R, Sakaue Y, Sawai C, Sawai T, Ozeki M, Romero-Pérez GA, et al. A preliminary investigation on the relationship between gut microbiota and gene expressions in peripheral mononuclear cells of infants with autism spectrum disorders. Biosci Biotechnol Biochem 2016;80:2450–8.
[32] Goines PE, Croen LA, Braunschweig D, Yoshida CK, Grether J, Hansen R, et al. Increased mid gestational IFN-, IL-4 and IL-5 in women bearing a child with autism:A case-control study. Mol Autism 2011;2:13.
[33] Shatz CJ. MHC class I: an unexpected role in neuronal plasticity. Neuron 2009;64:40–5.
[34] Kang DW, Adams JB, Gregory AC, Borody T, Chittick L, Fasano A, et al. Microbiota Transfer Therapy alters gut ecosystem and improves gastrointestinal and autism symptoms: an open-label study. Microbiome 2017;5:10.
[35] Srikantha P, Hasan Mohajeri M. The possible role of the microbiota-gut-brain-axis in autism spectrum disorder. Int J Mol Sci 2019;20:2115.
[36] Shen J. Modeling the glutamate-glutamine neurotransmitter cycle. Front Neuroenergetics 2013;5:1.
[37] Horder J, Petrinovic MM, Mendez MA, Bruns A, Takumi T, Spooren W, et al. Glutamate and GABA in autism spectrum disorder-a translational magnetic resonance spectroscopy study in man and rodent models. Transl Psychiatry 2018;8:106.
[38] Scheiman J, Luber JM, Chavkin TA, MacDonald T, Tung A, Pham LD, Wibowo MC, Wurth RC, Punthambaker S, Tierney BT, Yang Z, Hattab MW, Avila-Pacheco J, Clish CB, Lessard S, Church GM, Kostic AD. Meta-omics analysis of elite athletes identifies a performance-enhancing microbe that functions via lactate metabolism. Nat Med 2019 Jul;25(7):1104–9. https://doi.org/10.1038/s41591-019-0485-4. Epub 2019 Jun 24.
[39] Ansaldo E, Slayden LC, Ching KL, Koch MA, Wolf NK, Plichta DR, Brown EM, Graham DB, Xavier RJ, Moon JJ, Barton GM. *Akkermansia* muciniphila induces intestinal adaptive immune responses during homeostasis. Science 2019;364(6446):1179–84. Jun. 21.
[40] Zhai Q, Feng S, Arjan N, Chen W. A next generation probiotic, *Akkermansia muciniphila*. Crit Rev Food Sci Nutr 2019;59(19):3227–36.
[41] Kurmasheva N, Vorobiev V, Sharipova M, Efremova T, Mardanova A. The potential virulence factors of *Providencia stuartii*: motility, adherence, and invasion. BioMed Res Int 2018:3589135. . [Accessed 21 February 2018].
[42] Coretti L, Paparo L, Riccio MP, Amato F, Cuomo M, et al. Gut microbiota features in young children with autism spectrum disorders. Front Microbiol 2018;9:3146. Dec. 19.
[43] Xu M, Xu X, Li J, Li F. Association between gut microbiota and autism spectrum disorder: a systematic review and meta-analysis. Front Psychiatr 2019;10:473. Jul. 17.
[44] Rodríguez-Medina N, Barrios-Camacho H, Duran-Bedolla J, Garza-Ramos U. *Klebsiella variicola*: an emerging pathogen in humans. Emerg Microb Infect 2019;8(1):973–88.

2. L’étude de cohorte (cohort study)

Adherence to Screen Time and Physical Activity Guidelines is Associated with Executive Function in US Toddlers Participating in the STRONG Kids 2 Birth Cohort Study

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Objective To test the hypothesis that healthy weight status and adherence to American Academy of Pediatrics (AAP) guidelines for diet and physical activity would extend to greater executive function (EF) at age 24 months.

Study design Parents of 24-month-old children from the STRONG Kids 2 cohort study (n = 352) completed the Behavioral Rating Inventory of Executive Function for Preschoolers (BRIEF-P) and reported physical activities, diet, and screen time. Toddlers met AAP guidelines if they consumed at least 5 servings of fruits and vegetables, were physically active, refrained from sugar-sweetened beverages, and limited daily screen time to <60 minutes. Relationships between EF, 24-month weight status, and meeting AAP guidelines were tested independent of child sex, ethnicity, socioeconomic status, weight status at birth, and maternal pregnancy weight status.

Results Weight-for-length *z*-score had no effect on EF. Toddlers meeting the screen time guideline had greater EF (β, −0.125; 95% CI, 0.234 to −0.008), inhibitory self-control (β, −0.142; 95% CI, −0.248 to −0.029), and emergent metacognition (β, −0.111; 95% CI, −0.221 to 0.002), indicated by lower BRIEF-P scores. Those with more minutes of screen time had poorer overall EF (β, 0.257; 95% CI, 0.118-0.384), inhibitory self-control (β, 0.231; 95% CI, 0.099-0.354), cognitive flexibility (β, 0.217; 95% CI, 0.082-0.342), and emergent metacognition (β, 0.257; 95% CI, 0.120-0.381). Daily physical activity was associated with greater emergent metacognition (β, −0.116; 95% CI, −0.225 to −0.005).

Conclusions Meeting AAP guidelines for physical activity and screen time was related to greater EF in a demographically homogenous sample of toddlers. Future randomized control trials and more diverse samples are needed to confirm the directionality of this relationship. *(J Pediatr 2023;252:22-30)*.

Clinical trial registration ClinicalTrials.gov, NCT03341858.

Executive function (EF), defined as neurocognitive processes pertinent to the regulation of goal-directed behaviors,[1] is linked to overweight and obesity in childhood.[2] Low EF is implicated in lower academic success as early as preschool age,[3] as well as in poorer physical health (including overweight), financial instability, criminal offenses, and substance dependence in adulthood.[4] Inhibitory control is a domain of EF that allows one to regulate behavior, attention, thoughts, and emotions[1] and has been heavily studied in regard to weight status. This is likely due to its role in the regulation of energy intake and subsequently, prevention of excess weight gain.[2] Indeed, several longitudinal studies have observed poor performance on various aspects of inhibitory control in preschool ages to be predictive of body mass index (BMI) *z*-scores throughout childhood.[5,6] Few studies have explored the relationship between early-life weight status in multiple EF domains that are thought to develop rapidly over early childhood, such as working memory (which allows individuals to hold on to information for application to problem-solving situations) and cognitive flexibility (which allows for switching of perspective or focus)[1]; however, children with higher weight status as young as 5-9 years exhibit poorer EF in these domains.[7,8] By advancing our understanding of the relationship of weight status

3-moa	3 months of age	FFQ	Food Frequency Questionnaire
24-moa	24 months of age	FITS	Feeding Infants and Toddlers Study
AAP	American Academy of Pediatrics		
BMI	Body mass index	SES	Socioeconomic status
BRIEF-P	Behavioral Rating Inventory of Executive Function for Preschoolers	SPARK	Sports, Play, and Active Recreation for Kids
		SSB	Sugar-sweetened beverages
EF	Executive function	WFLZ	Weight-for-length *z*-score

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with these important predictors of success across the lifespan, we add to the understanding of the possible impact of obesity on cognitive development.

Because the prevalence of overweight and obesity tends to rise with age,[9] it is also important to consider how habits of diet and play that emerge around toddlerhood could influence EFs directly and indirectly through their effects on weight status. The American Academy of Pediatrics (AAP) provides evidence-based guidelines for the prevention of childhood obesity in the form of the *Bright Futures* initiative, which encourages (1) 5 or more servings of fruits and vegetables per day; (2) reduced or eliminated intake of sugar-sweetened beverages (SSB); (3) less than 1 hour of screen time daily; and (4) participation in daily physical activity or at least 60 minutes of moderate-to-vigorous physical activity daily.[10] Emerging evidence in older children suggests that adherence to these guidelines positively impacts cognitive function. Specifically, the amount of screen time[11,12] and physical activity,[13] as well as various aspects of diet quality,[14] have been linked to EF in school-aged and adolescent children, although not independently of weight status. A better understanding of the interrelationships among these factors in toddlerhood is needed, as that stage marks a sensitive period of cognitive development[15] and rapid brain growth.[16]

The primary aim of the present analysis was to examine how toddler weight status is related to EF. A secondary aim was to address the possible direct and indirect (through weight status) relationships among adherence to childhood obesity prevention guidelines, health behaviors, and better EF at 24 months of age (24-moa). A sub-aim of these analyses was to explore the impact of additional early-life factors, delivery mode, and feeding mode at 3 months of age (3-moa) as covariates of the relationship between weight status and EF. Determining the relationships among weight status, health behaviors, obesity prevention guidelines, and EF in toddlers could inform interventions aimed at improving adherence to guidelines.

Methods

Data were obtained for mothers and 24-moa dyads enrolled in the STRONG Kids 2 birth cohort study (ClinicalTrials.gov: NCT03341858).[17] This study was approved by the University of Illinois Institutional Review Board (13 448). Women were recruited during their third trimester of pregnancy between May 2013 and January 2017 from the Francis Nelson Center in conjunction with Carle Foundation Hospital (Champaign, IL), Christie Clinic (Champaign, IL), Decatur Memorial Hospital (Decatur, IL), Provena United Samaritans Medical Center, and Danville Polyclinic (Danville, IL) at prenatal visits or birthing classes provided by the sites. For birthing classes, recruitment was conducted by STRONG Kids research staff. For prenatal visits, recruitment materials were distributed by trained clinic or hospital staff. In addition, participants were recruited through local newsletters and other media outlets in the Champaign-Urbana, Danville, and Decatur areas.

Interested mothers were contacted by trained research staff to schedule a meeting during their next prenatal visit to discuss further details of the study and to confirm or decline enrollment. In either case, mothers were provided with a $15 gift card for their time. Online informed consent forms were completed by all participant parents or guardians. Infants with birth conditions that affect feeding or who were born prematurely (<37 weeks) or with a low birth weight (<2.5 kg) were excluded from the cohort study. After using these exclusionary criteria and addressing missing data a final sample of 356 children was retained for analyses (see the EF Measurement section and **Figure 1**; available at www.jpeds.com).

Measures

Demographics. Caregivers responded to a survey that provided their highest level of education, maternal prepregnancy height and weight, childbirth mode, child's birth weight and length, child's ethnicity/race, and household income at 24-moa. Mother's level of education and household income were used to compute composite socioeconomic status (SES) scores, which were divided into low, medium, and high SES. Some caregivers failed to disclose socioeconomic (n = 32), ethnicity (n = 16), prepregnancy weight (n = 13), birth weight (n = 7), and delivery mode (n = 1). There was no significant difference in child sex (mean difference, 0.03; 95% CI, −0.14 to 0.07), ethnicity (mean difference, 0.07; 95% CI, −0.16 to 0.03), 24-moa Weight-for-length *z*-score (WFLZ) (mean difference, 0.02; 95% CI, −0.21 to 0.24), age (mean difference, 0.01; 95% CI, −0.04 to 0.03), and 6-week high SES (mean difference <0.00; 95% CI, −0.10 to 0.10) or low SES (mean difference, 0.02; 95% CI, −0.04 to 0.08) between families lost to follow-up by 24-moa.

Anthropometrics. All 24-moa measurements were obtained during home visits by trained research personnel. After requesting that shoes and any excess, heavy clothing be removed, the average of 2 measurements of height and weight obtained with a portable stadiometer (Seca 213) and a digital scale (HealthOmeter 349KLX), respectively, were recorded. WFLZ was computed using the World Health Organization's child growth standards. Children were classified as obese (≥3.0), overweight (≥2.0), normal weight (−2.0 to <2.0), and underweight (<-2.0) based on established criteria.[18] Maternal prepregnancy height and weight were self-reported and BMI was used to classify mothers as underweight (BMI <18.5 kg/m^2), normal weight (18.5≤ BMI <25), overweight (25≤ BMI <30), or obese (BMI ≥30). Twenty-three participants at 24-moa had missing height and weight measurements.

EF. Parents completed the Behavioral Rating Inventory of EF for Preschoolers (BRIEF-P),[19] which consists of 63 questions on a single-rating scale of frequency—never (1), sometimes (2), or often (3)—of everyday behaviors at home or daycare. A lower score is indicative of better EF—that is, the child exhibits lower frequency of behavioral problems related to EF. Scoring was completed based on recommendations from the survey authors. In brief, the responses were aggregated to compute 5 clinical subscales

representing various dimensions of EF (Inhibit, Shift, Emotional Control, Working Memory, and Plan/Organize). According to the scoring recommendations, clinical scales consisting of 3 broader indices were then created: Inhibitory Self-Control (composed of Inhibit and Emotional Control scales), Cognitive Flexibility of Shift and Emotional Control, Emergent Metacognition of Working Memory, and Plan/Organize, along with Overall Executive Function (composed of all 5 scales).

In adherence to the BRIEF-P manual instructions, toddlers missing >12 items overall or >2 items on a scale were excluded from the analyses, and the remaining missing values (0.5%) on this survey were imputed with the response "never," at the guideline provided by the BRIEF-P manual. Survey results were assessed for validity using Negativity and Inconsistency indices.[19] Raw index scores were converted to T-scores and percentiles as instructed in the BRIEF-P manual for descriptive purposes. Raw scores are used in all analyses, as several of our toddlers fell just below 24-moa (n = 183; minimum age, 23.41 months).[20] Age was not associated with any BRIEF-P index and thus was not included as a covariate in the models.

Physical Activity. Parents reported the number of days per week that their child participated in various physical activities for at least 15 minutes using the Sports, Play, and Active Recreation for Kids (SPARK) survey.[21] Total activities reported were summed and used to determine weekly physical activity. Those reporting 1 activity per day were classified as meeting the AAP guideline for toddlers to engage in active play every day. In this case, it was assumed that if at least 7 activities were reported in a week, the child was participating in at least 1 activity per day. Thirteen participants failed to complete the SPARK survey.

Screen Time. The Common Sense Media Survey was used by parents to report their child's screen time.[22] This survey consists of both ratings and open-ended questions to determine the frequency and duration of various types of media, respectively. Minutes of media use that involved screens (ie, TV, DVD, shows on a computer or cellphone, games on a console, computer, cellphone, handheld device, and other uses of apps and computers) were summed to determine total screen time. Outlier values for screen time, as determined by those 3 SD above the mean (2075 minutes/day), were winsorized to the next highest value within 3 SD (685 minutes/day). Those reporting no more than 60 minutes of screen time were classified as meeting the AAP guideline for screen time. One participant failed to complete the Common Sense Media Survey.

Dietary Intake. At 3-moa, mothers completed survey items on feeding mode (ie, exclusive breastfeeding, formula feeding, or both) from the CDC Survey on Infant Feeding Practices Study II.[23] At 24-moa, parents completed the NutritionQuest Child Block Food Frequency Questionnaire (FFQ) for Ages 2-7, consisting of 90 questions pertaining to the child's usual eating habits in the previous 6 months. Food lists developed by NutritionQuest were obtained from National Health and Nutrition Examination Survey II dietary recall data, which provided approximate daily servings of fruits and vegetables and kcal from sugary beverages.[24] Children above (1602 kcal) or below (396 kcal) 2 SD of the mean for total kcal were considered outliers (n = 7) and were excluded, based on typical energy intake of ~1470 kcal for age 2-5 years.[25] Children who consumed at least 5 servings of fruits and vegetables were classified as meeting the AAP guideline for fruit and vegetable servings. The AAP recommends limited consumption of sugary beverages[10]; thus, children who did not consume any energy from sugary beverages were classified as meeting the guideline. Fifty-one participants failed to complete the FFQ, and 8 did not complete the CDC Survey on Infant Feeding Practices Study II.

Statistical Analyses

A path analysis with the structural equation modeling technique was performed with MPlus version 8.4 to assess the direct and indirect relationships between AAP guidelines and heath behaviors with EF (**Figures 2** and **3**). Direct effects were tested to examine AAP guidelines and continuous health behavior relationships with BRIEF-P indices. Indirect effects were tested to explore the possible mediating effect of WFLZ at 24-moa on the relationships of AAP guidelines and continuous health behaviors with BRIEF-P indices. All path coefficients were freely estimated in the models. A threshold of $P = .05$ was considered significant. P values were corrected for multiple comparisons using a Benjamini–Hochberg procedure with a false discovery rate of 0.1 to determine significance after correction. Because individual testing was the primary interest of this study (ie, to determine relationships between individual components of EF with specific AAP guidelines or continuous health behaviors), P values were considered the primary indicator for hypothesis testing.[26] The full information maximum likelihood approach was used to handle missing data. Unlike traditional maximum likelihood estimation, which requires complete data, this method uses all observed variables for each case, allowing computation of parameter estimates even in the presence of missing data. The full information maximum likelihood approach has been shown to produce unbiased parameter estimates and standard errors when data are at least missing at random.[27] Selection of the covariates—child sex, SES, birth WFLZ, ethnicity, and maternal pre-pregnancy weight status—was based on a priori expected association with EF and WFLZ at 24-moa.[10,19,20] The independent-samples t-test and ANOVA were used to examine these relations in our sample. These analyses and descriptive statistics were conducted in SPSS 28.0 (IBM). Direct and indirect effects were tested using a bootstrap estimation approach with 5000 samples. Bootstrap SEs and CIs of the direct and indirect effects were calculated. Post hoc power analysis revealed that our model was sufficiently powered (95.4%) to detect an effect on overall EF, based on number

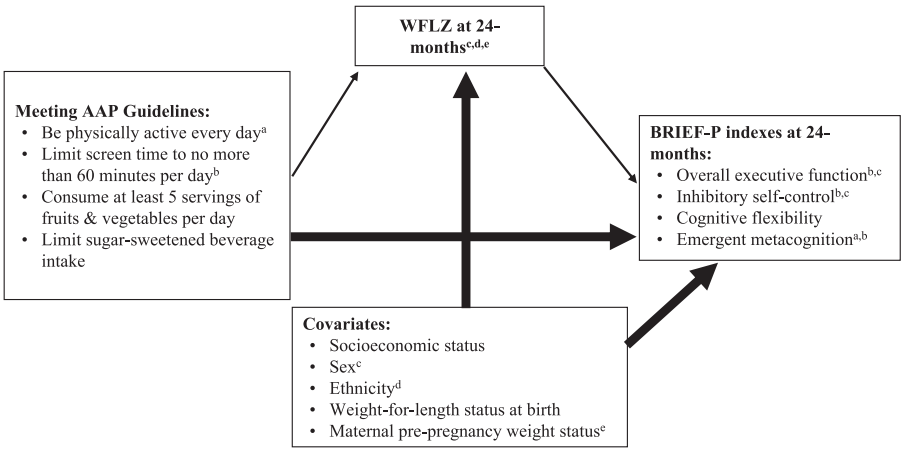

Figure 2. Path model A. Direct and indirect effects (through weight status) of AAP guidelines for physical activity, screen time, and fruit, vegetable, and SSB intakes on EF.

of predictors included in the model (n = 10), observed R^2 of 6.7%, probability level of .05, and sample size of 356.[28,29] Additionally, post hoc power analyses were performed to evaluate the sufficiency of meeting the guideline for fruits and vegetable consumption and 24-moa WFLZ groups owing to their uneven group sizes.[30]

Results

Participant recruitment and data analysis flow can be found in **Figure 1**. Demographic data, weight status, and EF percentiles standardized for age and sex are presented in **Table I**. Adherence to AAP guidelines can be found in **Table II**. Eight percent of toddlers met none of the guidelines, 25% met 1 guideline, 38% met 2 guidelines, 28% met 3 guidelines, and 1% met all 4 AAP guidelines. Note that EF scores were all below the 50th percentile for frequency of problematic behaviors related to poorer EF (**Table I**). Toddlers of mothers with obesity or overweight prior to conception had higher WFLZ at 24-moa (mean difference, 0.73; 95% CI, 0.39 to 1.07) and toddlers who had a higher WFLZ at birth (≥2.0) exhibit higher WFLZ at 24-moa (mean difference, 0.82; 95% CI, −1.63 to 0.01). The *t*-test showed no significant difference in EF by any covariate or in WFLZ at 24-mo by 3-moa feeding mode, delivery mode, ethnicity, or SES in this sample (**Table III**; available at www.jpeds.com). Sex, ethnicity, SES, WFLZ at birth, and maternal pregnancy weight status were included as covariates in subsequent analyses.

Model A Results: Adherence to AAP Guidelines and EF

Adherence to guidelines was not associated with WFLZ. Toddlers adhering to the screen time guideline had significantly greater emergent metacognition (β, −0.111; 95% CI, −0.221 to 0.002), inhibitory self-control (β, −0.142; 95% CI, −0.248 to −0.029), and overall EF abilities (β, −0.125; 95% CI, −0.234 to −0.008), indicated by lower BRIEF-P scores. Those who met the guideline of daily physical activity had greater emergent metacognition (β, −0.116; 95% CI, −0.225 to −0.005). Those meeting the guideline to limit

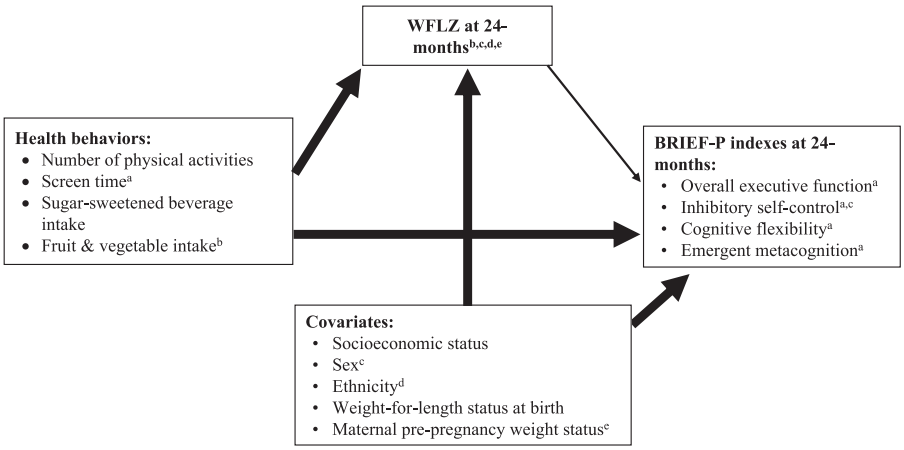

Figure 3. Path model B. Direct and indirect effects (through weight status) of physical activity, screen time, and fruit, vegetable, and SSB intakes on EF.

Table I. Maternal and child characteristics and demographics

Characteristics	Total N	Values
Child sex, n (%)	356	
Male		177 (49.7)
Female		179 (50.3)
Child race, n (%)	340	
Caucasian		255 (75.7)
Asian		18 (5.3)
Black		18 (5.3)
American Indian or Alaskan Native		1 (0.3)
Multiple races		45 (13.4)
SES, n (%)	324	
Low		15 (4.6)
Medium		121 (37.3)
High		188 (58.0)
Child age, mo, mean ± SE	356	24.1 (0.03)
Delivery mode, n (%)	355	
Cesarean		83 (23.4)
Vaginal		272 (76.6)
Feeding mode at 3-moa, n (%)	348	
Exclusively breastfed		238 (68.4)
Mixed feeding		61 (17.5)
Exclusively formula-fed		49 (14.1)
Child WFLZ*		
Birth weight category, n (%)	349	
Underweight		36 (10.3)
Normal weight		299 (85.7)
Overweight		14 (4.0)
24-month weight category, n (%)	333	
Underweight		12 (3.6)
Normal weight		254 (76.3)
Overweight		67 (20.1)
Maternal prepregnancy weight status, n (%)	343	
Normal weight		168 (49.0)
Overweight		85 (24.8)
Obese		90 (26.2)
Standardized EF scores†, mean (SE)	356	
Overall EF		47.2 (1.7)
Inhibitory self-control		45.1 (1.5)
Cognitive flexibility		44.1 (1.5)
Working memory		47.8 (1.8)
Raw EF scores‡, mean (SE)	356	
Overall EF		91.3 (1.0)
Inhibitory self-control		38.4 (0.4)
Cognitive flexibility		29.3 (0.3)
Working memory		38.8 (0.5)

*WFLZ characterized by weight category using the following World Health Organization standard cutoffs: <-2.0, underweight; −2.0 to <2.0, normal weight; ≥2.0 to <3.0, overweight; ≥3.0, obese.
†Data are average percentile scores standardized for sex and age (SE). Lower than 50th percentile indicates reporting fewer problems related to EF relative to the BRIEF-P standard population mean.
‡Data are average raw scores (SE) used in analyses.

Table II. Adherence to AAP guidelines (%) and mean participant physical activities, screen time, and fruit, vegetable, and SSB intakes

Measures	N	Reported frequency, mean (SE)	Guidelines	% adherence (n)
Physical activities, n/wk	343	10.8 (0.5)	Physically active every day	75.5 (259)
Screen time, min/d	355	98.2 (6.4)	No more than 60 minutes of screen time daily	54.1 (192)
Fruits and vegetables, servings/d	305	2.8 (0.1)	At least 5 servings of fruits and vegetables daily	3.9 (12)
SSB, kcal/d	305	11.5 (1.7)*	Limit SSB	68.9 (210)

*Includes only those who reported any SSB intake (n = 95).

SSB had numerically greater emergent metacognition (β, −0.118; 95% CI, −0.240 to 0.001), inhibitory self-control (β, −0.107; 95% CI, −0.232 to 0.020), and overall EF (β, −0.113; 95% CI, −0.237 to −0.010), although the differences were not statistically significant. There was no significant difference in BRIEF-P indices for those meeting the guideline for fruit and vegetable intake. WFLZ at 24-moa was not associated with any BRIEF-P index, nor did it have any mediating effects on the relationships between AAP guidelines and BRIEF-P (**Table IV**; available at www.jpeds.com). AAP guidelines and covariates explained 10.9% of the variance in WFLZ at 24-moa. AAP guidelines, WFLZ, and covariates explained 6.7%, 6.8%, and 6.8% of the variance in overall EF, inhibitory self-control, and emergent metacognition, respectively. Male children (β, −0.127; 95% CI, −0.237 to −0.023), children of Caucasian ethnicity (β, 0.125; 95% CI, 0.007 to 0.242), and mothers with overweight or obesity prior to pregnancy (β, 0.232; 95% CI, 0.121 to 0.351) had higher WFLZ at 24-moa. Males also had poorer inhibitory self-control (β, −0.117; 95% CI, −0.223 to −0.018). Correction for multiple comparisons abrogated all significant relationships with EF and WFLZ in the model (**Table IV**).

Model B Results: Associations of Physical Activity, Screen Time, and Diet with EF

Toddlers consuming more servings of fruits and vegetables had significantly higher WFLZ at 24-moa (β, 0.131; 95% CI, 0.006-0.252). Although not statistically significant, toddlers with more screen time had higher WFLZ (β, 0.116; 95% CI, −0.012 to 0.233). Screen time was significantly associated with each BRIEF-P index, such that toddlers with more screen time had poorer overall EF (β, 0.257; 95% CI, 0.118-0.384), inhibitory self-control (β, 0.231; 95% CI, 0.099-0.354), cognitive flexibility (β, 0.217; 95% CI, 0.082-0.342), and emergent metacognition (β, 0.257; 95% CI, 0.120-0.381). Intake of SSB, consumption of fruit or vegetables, and physical activity were not associated with any BRIEF-P index. SSB and number of physical activities also were not associated with WLZ at 24-moa (**Table V**; available at www.jpeds.com). WFLZ at 24-moa had no effect on any BRIEF-P index, nor did it have any mediating effect on the relationships between health behaviors and BRIEF-P. Health behaviors, WFLZ, and covariates explained 8.1%, 7.9%, 5.8%, and 7.7% of the variance in overall EF, inhibitory self-control, cognitive flexibility, and emergent metacognition, respectively, and health behaviors and covariates explained 11.7% of the variance in WFLZ at 24-moa. Male children (β, −0.120; 95% CI, −0.230 to −0.021), children of Caucasian ethnicity (β, 0.121; 95% CI, 0.005-0.240), and mothers with overweight or obesity prior to pregnancy (β, 0.213; 95% CI, 0.101-0.329) had higher WFLZ at 24-moa. Males also had poorer inhibitory self-control (β, −0.106; 95% CI, −0.209

to −0.007). After correction for multiple comparisons, all effects remained significant in the model except for the direct association between fruit and vegetable servings with higher WFLZ at 24-moa (**Table V**).

Discussion

The current study expands on prior findings of poorer EFs linked to higher weight status[2,5,7,8] and factors influencing weight regulation in older children,[2,11-14] by addressing these relationships in a large cohort of toddlers for each EF domain individually and as a composite score. The results suggest that associations between health behaviors and EFs may precede observed relationships between EFs and weight status. Toddlers meeting the guideline to limit screen time to no more than 1 hour per day had better inhibitory self-control and emergent metacognition, as well as overall EF. Lower total screen time also was predictive of higher overall EF and each domain of EF. Meeting the guideline to be physically active every d was associated with greater emergent metacognition. Although associations specific to meeting guidelines was abrogated on correction for multiple comparisons, this should be interpreted with caution, owing to the interest in individual testing of relationships between EFs and guidelines. Alternatively, this result may be simply a further indicator (aside from effect sizes) of the weaker association of EFs with guidelines compared with continuous variables, which had greater variability. Overall, these findings suggest that obesity risk factors are relevant, and that adherence to guidelines for daily physical activity and screen time limitations may be advantageous not only for weight regulation, but also for cognitive development.

Although much of the work surrounding EFs and weight status has focused on the causal relationship of the former on the latter, this relationship is likely to be birectional. Indeed, up-regulation of inflammatory cytokines and other obesity-associated biomarkers may have consequences for brain growth and development.[31,32] A study of 9- to 11-year-olds showed that the relationship between increased BMI and lower EF was mediated by cortical thickness of the prefrontal cortex, the maturation of which has been linked to EF development.[33] Despite these previous observations in older children, there was no association between weight status and any domain of EF in the current sample of toddlers; however, a high proportion of the current sample was of healthy weight at 24-moa. Post hoc power analyses revealed low power (17%) to detect an effect on overall EF, based on a Cohen *d* value of 0.110.

Therefore, our results may indicate that the relationship between greater weight status and EF emerges later in childhood; toddlers must rely heavily on their caregiver for dietary intake, but this reliance tends to decrease throughout childhood as they naturally become more independent. In this sample, toddlers of mothers with overweight or obesity prior to conception had higher WFLZ at 24-moa. Although noncausal, this may suggest a role for caregiver weight status on weight management in early childhood and may be a reflection of the relevance of family-wide interventions for promoting optimal health behaviors in young children. Longitudinal work in this area could elucidate the age at which relationships between EF and weight status emerge, and whether parent weight status and behaviors continue to track with child weight status throughout early life.

Although a priori covariates were implemented for regression analyses, we also explored other early-life associations with EF in an effort to promote the generalizability of our results, regardless of diet and delivery mode during infancy. Comparing groups who were exclusively breastfed to those who were formula-fed or mixed-fed at 3-moa revealed no significant differences in any domain of EF. In contrast, a recent study reported that each month of exclusive breastfeeding was associated with a decreased risk of clinically defined working memory deficit in 6-year-olds, even after adjusting for SES, among other factors; however, no relationship was found for inhibition or overall EF.[34] It is possible that the relationship between breastfeeding and EF is not observable until the child is older (6 years vs 2 years). On the other hand, previous work in a large sample of infants (n = 11 134) suggested that the negative relationship of delivery via cesarean with cognitive outcomes may be rescued by age 3 years.[35] Consistent with this work, our results showed no relationship between infant delivery mode and EF. The present study sample demonstrated an especially high proportion of exclusively breastfed infants at 3-moa (68%) and vaginally delivered infants (77%), whereas only 40% of infants aged <6 months are exclusively breastfed worldwide,[36] and ∼68% of children in the US are delivered vaginally.[37] Considering this and the cross-sectional nature of the present study, longitudinal analyses exploring relationships of early-life feeding and delivery mode with EFs throughout childhood in a more diverse sample are needed.

Many parents and guardians in this sample reported toddler SSB intake, screen time, and physical activities in meeting AAP guidelines similar to previous findings in a national, cross-sectional analysis of the 2008 Feeding Infants and Toddlers Study (FITS) of >600 2-year-olds (defined as age 24-35.9 months). In the FITS, ∼70% of the toddlers did not meet the recommended 5 servings of fruits and vegetables per day, ∼55% consumed SSB on a daily basis, ∼20% exceeded 2 hours of screen time per day, and only 30% engaged in active play outside for at least 1 hour per day.[38] Our current sample yielded an especially low adherence rate (5%) for consumption of 5 servings of fruits and vegetables per day. Surprisingly, post hoc power analyses indicated that the power to detect a significant difference between those meeting and not meeting the guideline for fruits and vegetables for overall EF was likely sufficient (74.4%), based on a Cohen *d* of 0.301. However, no significant relationships were found between EFs and fruits and vegetable guideline or intake. Regardless, it is still possible that low adherence and lack of variability in the current sample impacted these findings, possibly owing to imprecise assessment in serving sizes for toddlers, given that the Block FFQ Ages 2-7 does not inquire about food serving sizes (only beverages).

Furthermore, as young children tend to meet recommendations for daily fruit servings but not for vegetable servings (and especially nonstarchy vegetables),[39] a less crude analysis of diet quality is likely required to detect an effect and should be considered in future analyses.

Even on inquiry of beverage portions, SSB also was not related to EFs, even though those meeting the guideline for SSB exhibited trend toward better EF. Many parents reported no intake of SSB, and those who did report intake of SBB reported very little (11.5 kcal/day). Recent NHANES data suggest that males and females aged 2-5 years consumed 65 kcal and 59 kcal from SSB daily, respectively, an average that is likely skewed by the older ages, because intake of SSB tends to increase across childhood age groups. Even for children aged 6-11 years, the daily kcal from SSB almost doubles, to 133 kcal in males and 104 kcal in females.[40] Therefore, considering our highly educated sample of families and the young age of our sample, it is possible that these kcal estimations of SSB are fairly accurate but are not necessarily generalizable. Regardless of study limitations, the lack of relationship between diet and EF was surprising, given the evidence in older children showing relationships between SSB, fruit and vegetable intake, and diet quality (of which these are a hallmarks) and EF.[14]

Limiting screen time to no more than 60 minutes per day was not associated with cognitive flexibility, and the relationships with the other indices were not as strong as those with the continuous screen time variable. Although noncausal, this may indicate that more than 60 minutes of screen time by toddlers negatively impacted EF. Because the emotional control subscale overlaps for cognitive flexibility and inhibitory self-control, the lack of association between limiting screen time and cognitive flexibility also may indicate greater influence of the inhibition subscale in the relationship between screen time and inhibitory self-control (composed of inhibit and emotional control subscales). Mechanisms for the relationship between screen time and EFs are unclear, however. Nathanson and Fries proposed two mechanisms: one dependent on the specific content, some of which may alter the child's perception of the social world (eg, fast-paced, fantasy television that requires and rewards inefficient information processing),[12] and the other involves the indirect effect of screen time on EF through other behaviors. In fact, screen time has been negatively correlated with both physical activity and sleep time in toddlerhood.[12,41]

Our results showing greater working memory in toddlers who were more physically active supports observations that physical activity interventions can improve EF in school-aged children;[13,38,42] however, these findings are novel in suggesting that this relationship begins in toddlerhood and may be specific to emergent metacognition. Physical activity has numerous health benefits relevant to development, including the up-regulation of important neurodevelopmental growth factors.[43] In fact, magnetic resonance imaging studies comparing school-aged children, characterized by their fitness level and/or involvement in a physical activity intervention, exhibited functional[44] and structural changes,[45] respectively, in areas of the brain associated with EF, along with better performance on cognitive control tasks.

This sample of toddlers was a highly homogenous demographic representation, and future studies should aim to recruit a more ethnically and socioeconomically diverse sample. Given the greater risk for high screen use, obesity, low physical activity, and poor diet in children of non-Caucasian ethnicities/races and lower SES,[46,47] this is especially important to address in future studies. Another limitation of the present study was the methods of assessment for physical activity and diet. Measurement of physical activity was through parental report of number of activities engaged in for at least 15 minutes per week. As a result, actual time spent doing physical activity could not be determined, and the actual AAP Bright Futures guideline for 60 minutes of moderate-to-vigorous physical activity per day could not be determined. Diet was assessed through FFQ, which introduces bias related to assumption of serving sizes and reliance on parent memory of diet over the last 6 months. Future work should aim to collect time spent doing physical activities and more precise measures of SSB, fruit, and vegetable intake.

Toddlers with less screen use who meet the AAP guidelines for both screen time and physical activity exhibited better EF, suggesting a potential role of these health behaviors in cognitive development. Based on observations in older children, these study results suggest that relationships between more sedentary and less active play with poorer EF emerges earlier in life compared with weight status. However, owing to the aforementioned limitations and the cross-sectional study design, these results should be interpreted with caution. In addition, a majority (67%) of toddlers met multiple guidelines, suggesting that health behaviors in toddlerhood tend to coincide. Future studies should explore interactions and coincidence of health behaviors, as well as their effects on EF development. Exploring the interplay of these factors and growth trajectories over time could be foundational to the understanding of their impact on early-life cognitive development. Regardless of limitations, this work is among the few studies to explore multiple EF domain relationships with weight status and health behaviors in a large sample of toddlers, emphasizing the need for future, robust studies to determine the significance of building healthy habits from a young age to promote early EF development. ■

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Data Statement

Data sharing statement available at www.jpeds.com.

References

1. Diamond A. Executive functions. Annu Rev Psychol 2013;64:135-68. https://doi.org/10.1146/annurev-psych-113011-143750
2. Liang J, Matheson BE, Kaye WH, Boutelle KN. Neurocognitive correlates of obesity and obesity-related behaviors in children and adolescents. Int J Obes (Lond) 2014;38:494-506. https://doi.org/10.1038/ijo.2013.142
3. Blair C, Razza RP. Relating effortful control, executive function, and false belief understanding to emerging math and literacy ability in kindergarten. Child Dev 2007;78:647-63. https://doi.org/10.1111/j.1467-8624.2007.01019.x
4. Moffitt TE, Arseneault L, Belsky D, Dickson N, Hancox RJ, Harrington H, et al. A gradient of childhood self-control predicts health, wealth, and public safety. Proc Natl Acad Sci U S A 2011;108:2693-8. https://doi.org/10.1073/pnas.1010076108
5. Francis LA, Susman EJ. Self-regulation and rapid weight gain in children from age 3 to 12 years. Arch Pediatr Adolesc Med 2009;163:297-302. https://doi.org/10.1001/archpediatrics.2008.579
6. Graziano PA, Calkins SD, Keane SP. Toddler self-regulation skills predict risk for pediatric obesity. Int J Obes (Lond) 2010;34:633-41. https://doi.org/10.1038/ijo.2009.288
7. Wirt T, Schreiber A, Kesztyüs D, Steinacker JM. Early life cognitive abilities and body weight: cross-sectional study of the association of inhibitory control, cognitive flexibility, and sustained attention with BMI percentiles in primary school children. J Obes 2015;2015:534651. https://doi.org/10.1155/2015/534651
8. Li N, Yolton K, Lanphear BP, Chen A, Kalkwarf HJ, Braun JM. Impact of early-life weight status on cognitive abilities in children. Obesity (Silver Spring) 2018;26:1088-95. https://doi.org/10.1002/oby.22192
9. Ogden CL, Carroll MD, Lawman HG, Fryar CD, Kruszon-Moran D, Kit BK, et al. Trends in obesity prevalence among children and adolescents in the United States, 1988-1994 through 2013-2014. JAMA 2016;315:2292-9. https://doi.org/10.1001/jama.2016.6361
10. Hagan JF Jr., Shaw JS, Duncan PM, eds. Bright futures: guidelines for health supervision of infants, children, and adolescents. 4th ed. Elk Grove Village (IL): American Academy of Pediatrics; 2017.
11. Lillard AS, Peterson J. The immediate impact of different types of television on young children's executive function. Pediatrics 2011;128:644-9. https://doi.org/10.1542/peds.2010-1919
12. Nathanson AI, Fries PT. Television exposure, sleep time, and neuropsychological function among preschoolers. Media Psychol 2014;17:237-61. https://doi.org/10.1080/15213269.2014.915197
13. Hillman CH, Pontifex MB, Castelli DM, Khan N, Raine LBR, Scudder M, et al. Effects of the FITKids randomized controlled trial on executive control and brain function. Pediatrics 2014;134:e1063-71. https://doi.org/10.1542/peds.2013-3219
14. Cohen JFW, Gorski MT, Gruber SA, Kurdziel LBF, Rimm EB. The effect of healthy dietary consumption on executive cognitive functioning in children and adolescents: a systematic review. Br J Nutr 2016;116:989-1000. https://doi.org/10.1017/S0007114516002877
15. Nelson CA 3rd, Zeanah CH, Fox NA, Marshall PJ, Smyke AT, Guthrie D. Cognitive recovery in socially deprived young children: the Bucharest Early Intervention Project. Science 2007;318:1937-40. https://doi.org/10.1126/science.1143921
16. Knickmeyer RC, Gouttard S, Kang C, Wilber K, Smith J, Hamer R, et al. A structural MRI study of human brain development from birth to 2 Years. J Neurosci 2008;28:12176-82. https://doi.org/10.1523/JNEUROSCI.3479-08.2008
17. Fiese BH, Musaad S, Bost KK, McBride B, Lee S, Teran-Garcia M, et al. The STRONG Kids 2 birth cohort study: a cell-to-society approach to dietary habits and weight trajectories across the first 5 years of life. Curr Dev Nutr 2019;3:nzz007. https://doi.org/10.1093/cdn/nzz007
18. World Health Organization. WHO child growth standards: length/height-for-age, weight-for-age, weight-for-length, weight-for-height and body mass index-for-age: methods and development. Accessed September 6, 2019. https://www.who.int/publications/i/item/924154693X
19. Gioia GA, Espy KA, Isquith PK. Behavior Rating Inventory of Executive Function – pre-school version (BRIEF-P). Lutz (FL): Psychological Assessment Resources Inc; 2003.
20. Iwinski S, Donovan SM, Fiese B, Bost K. The impact of household chaos and dietary intake on executive function in young children. Nutrients 2021;13:4442. https://doi.org/10.3390/nu13124442
21. Mckenzie TL, Sallis JF, Rosengard P. Beyond the stucco tower: design, development, and dissemination of the SPARK physical education programs. Quest 2009;61:114-27. https://doi.org/10.1080/00336297.2009.10483606
22. Common Sense Media. Zero to eight: children's media use in America 2013. Accessed September 8, 2019. https://www.commonsensemedia.org/research/zero-to-eight-childrens-media-use-in-america-2013
23. Shealy KR, Scanlon KS, Labiner-Wolfe J, Fein SB, Grummer-Strawn LM. Characteristics of breastfeeding practices among US Mothers. Pediatrics 2008;122(Suppl 2):S50-5. https://doi.org/10.1542/peds.2008-1315f
24. NutritionQuest, Assessment & Analysis Services. Questionnaires and screeners. Accessed August 28, 2019. https://www.nutritionquest.com/assessment/list-of-questionnaires-and-screeners/
25. US Department of Agriculture, Agricultural Research Service. Nutrient intakes from food and beverages: mean amounts consumed per individual, by gender and age. What We Eat in America, NHANES 2015-2016. 2018. Accessed June 29, 2022. https://www.ars.usda.gov/ARSUserFiles/80400530/pdf/1516/tables_1-36_2015-2016.pdf
26. Rubin M. When to adjust alpha during multiple testing: a consideration of disjunction, conjunction, and individual testing. Synthese 2021 https://doi.org/10.1007/s11229-021-03276-4
27. Kane SP. Post. ClinCalc. Accessed July 2, 2022. https://clincalc.com/stats/Power.aspx. Updated November 10, 2018.
28. Enders CK, Bandalos DL. The relative performance of full information maximum likelihood estimation for missing data in structural equation models. Struct Equation Model 2001;8:430-57.
29. Soper DS. Post-hoc statistical power calculator for multiple regression [Software]. 2022. Accessed June 29, 2022. https://www.danielsoper.com/statcalc
30. Cohen J. Statistical power analysis for the behavioral sciences. 2nd ed. Hillsdale (NJ): Lawrence Earlbaum Associates; 1988.
31. Shields GS, Moons WG, Slavich GM. Inflammation, self-regulation, and health: an immunologic model of self-regulatory failure. Perspect Psychol Sci 2017;12:588-612. https://doi.org/10.1177/1745691616689091
32. Ronan L, Alexander-Bloch A, Fletcher PC. Childhood obesity, cortical structure, and executive function in healthy children. Cereb Cortex 2020;30:2519-28. https://doi.org/10.1093/cercor/bhz257
33. Al-Khalaf SY, O'Neill SM, O'Keeffe LM, Henriksen TB, Kenny LC, Cryan JF, et al. The impact of obstetric mode of delivery on childhood behavior. Soc Psychiatry Psychiatr Epidemiol 2015;50:1557-67. https://doi.org/10.1007/s00127-015-1055-9
34. Keim SA, Sullivan JA, Sheppard K, Smith K, Ingol T, Boone KM, et al. Feeding infants at the breast or feeding expressed human milk: long-term cognitive, executive function, and eating behavior outcomes at age 6 years. J Pediatr 2021;233:66-73.e1. https://doi.org/10.1016/j.jpeds.2021.02.025
35. Miller AL, Lee HJ, Lumeng JC. Obesity-associated biomarkers and executive function in children. Pediatr Res 2015;77:143-7. https://doi.org/10.1038/pr.2014.158
36. UNICEF/WHO. Global Breastfeeding Scorecard, 2017. Global Breastfeeding Collective. 2017. Tracking progress for breastfeeding policies and programmes. Accessed June 29, 2022. https://www.who.int/publications/m/item/global-breastfeeding-scorecard-2017-tracking-progress-for-breastfeeding-policies-and-programmes
37. Centers for Disease Control and Prevention. National Center for Health Statistics. Method of delivery. 2020. Accessed June 29, 2022. https://www.cdc.gov/nchs/fastats/delivery.htm
38. Briefel RR, Deming DM, Reidy KC. Parents' perceptions and adherence to children's diet and activity recommendations: the 2008 feeding infants and toddlers study. Prev Chronic Dis 2015;12:E159. https://doi.org/10.5888/pcd12.150110
39. US Department of Agriculture and US Department of Health and Human Services. Dietary guidelines for Americans, 2020-2025. Accessed June 29, 2022. https://www.dietaryguidelines.gov/resources/2020-2025-dietary-guidelines-online-materials
40. Rosinger A, Herrick K, Gahche J, Park S. Sugar-sweetened beverage consumption among U.S. youth, 2011-2014. NCHS Data Brief 2017:1-8.

41. Venetsanou F, Kambas A, Gourgoulis V, Yannakoulia M. Physical activity in pre-school children: trends over time and associations with body mass index and screen time. Ann Hum Biol 2019;46:393-9. https://doi.org/10.1080/03014460.2019.1659414
42. Álvarez-Bueno C, Pesce C, Cavero-Redondo I, Sánchez-López M, Martínez-Hortelano JA, Martínez-Vizcaíno V. The effect of physical activity interventions on children's cognition and metacognition: a systematic review and meta-analysis. J Am Acad Child Adolesc Psychiatry 2017;56:729-38. https://doi.org/10.1016/j.jaac.2017.06.012
43. Arvidsson D, Johannesson E, Andersen LB, Karlsson M, Wollmer P, Thorsson O, et al. A longitudinal analysis of the relationships of physical activity and body fat with nerve growth factor and brain-derived neural factor in children. J Phys Act Health 2018;15:620-5. https://doi.org/10.1123/jpah.2017-0483
44. Chaddock L, Hillman CH, Pontifex MB, Johnson CR, Raine LB, Kramer AF. Childhood aerobic fitness predicts cognitive performance one year later. J Sports Sci 2012;30:421-30. https://doi.org/10.1080/02640414.2011.647706
45. Chaddock-Heyman L, Erickson KI, Voss MW, Knecht AM, Pontifex MB, Castelli DM, et al. The effects of physical activity on functional MRI activation associated with cognitive control in children: a randomized controlled intervention. Front Hum Neurosci 2013;7:72. https://doi.org/10.3389/fnhum.2013.00072
46. Duch H, Fisher EM, Ensari I, Harrington A. Screen time use in children under 3 years old: a systematic review of correlates. Int J Behav Nutr Phys Act 2013;10:102. https://doi.org/10.1186/1479-5868-10-102
47. Byrd AS, Toth AT, Stanford FC. Racial disparities in obesity treatment. Curr Obes Rep 2018;7:130-8. https://doi.org/10.1007/s13679-018-0301-3

50 Years Ago in *The Journal of Pediatrics*

The Continued Importance of the Meckel Scan

Jaros R, Schussheim A, Levy LM. Preoperative diagnosis of bleeding Meckel's diverticulum utilizing 99m technetium pertechnetate scinti-imaging. J Pediatr 1973;82:45-9.

Meckel diverticulum results from incomplete obliteration of the vestigial vitelline duct and is the most common congenital abnormality of the gastrointestinal tract, affecting approximately 2% of the population.[1] Meckel diverticulum is located in the ileum and usually contains ectopic gastric mucosa, which can cause ulceration of the adjacent ileal mucosa and rectal bleeding but also can contain pancreatic, duodenal, or colonic mucosa. Fifty years ago, children with rectal bleeding of unknown etiology would undergo an exploratory laparotomy in search of a Meckel diverticulum or other cause. In 1973, ^{99}technetium pertechnetate scintigraphy, now known as the Meckel scan, was first used in pediatric patients perioperatively to diagnose Meckel diverticulum with ectopic gastric mucosa. Jaros et al used the technology in 5 children with rectal bleeding of unknown etiology. Two of these children had positive scans and subsequently had surgery to excise a Meckel diverticulum; 3 children had normal scans and avoided unnecessary laparotomies.

Although most patients remain asymptomatic, a recent study of 945 children with Meckel diverticulum found that 60% presented with bowel obstruction including intussusception, 36% presented with bleeding, and 8% presented with inflammation, which can appear as appendicitis-like symptoms in a patient with a normal appendix.[2] Many imaging modalities, including angiography, video capsule endoscopy, and small-bowel enteroscopy, can detect a Meckel diverticulum but are not commonly used for this purpose. Although there have been technical advances to the scan since 1973, when Polaroid films augmented a 90-minute digital recording, it maintains the same underlying technology. Now, premedication with H_2 antagonists often is used to enhance tracer uptake into gastric tissue, and a Meckel scan has high sensitivity and specificity, both approaching 100%.[3] Today, the Meckel scan remains the study of choice in hemodynamically stable patients with rectal bleeding and should be used for prompt recognition and differentiation of this congenital anomaly from other conditions with similar clinical presentations.

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References

1. Hansen CC, Søreide K. Systematic review of epidemiology, presentation, and management of Meckel's diverticulum in the 21st century. Medicine (Baltimore) 2018;97(35):e12154.
2. Alemayehu H, Hall M, Desai AA, St Peter SD, Snyder CL. Demographic disparities of children presenting with symptomatic Meckel's diverticulum in children's hospitals. Pediatr Surg Int 2014;30:649-53.
3. Irvine I, Doherty A, Hayes R. Bleeding meckel's diverticulum: A study of the accuracy of pertechnetate scintigraphy as a diagnostic tool. Eur J Radiol 2017;96:27-30.

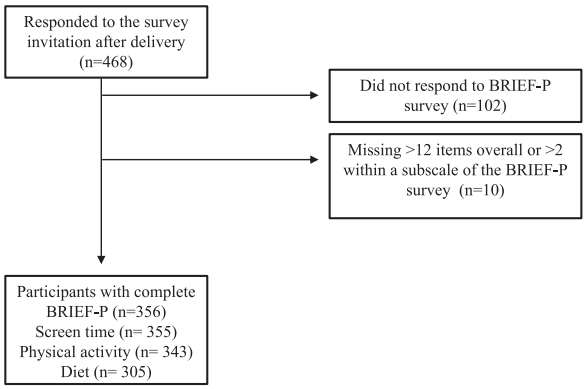

Figure 1. Study recruitment and data analysis flow.

Table III. Means of the BRIEF-P indices and WFLZ at 24-moa by sex, ethnicity, SES, WFLZ at birth, maternal prepregnancy weight status, delivery mode, and feeding mode at 3-moa

Variables	Overall EF	Cognitive flexibility	Inhibitory self-control	Emergent metacognition	WFLZ at 24-moa
Sex					
Females (n)	89.67 ± 1.34 (87.06-92.25)	28.93 ± 0.44 (28.10-29.80)	37.60 ± 0.59 (36.44-38.75)	38.14 ± 0.65 (36.86-39.40)	0.55 ± 0.10 (0.35-0.74)
Males (n)	93.03 ± 1.38 (90.32-95.74)	29.71 ± 0.44 (28.86-30.59)	39.25 ± 0.62 (38.01-40.45)	39.53 ± 0.67 (38.19-40.82)	0.81 ± 0.11 (0.59-1.04)
P value	.078	.215	.057	.137	.084
Ethnicity					
Caucasian (n)	90.91 ± 1.12 (88.72-93.11)	29.13 ± 0.36 (28.44-29.82)	38.28 ± 0.50 (37.32-39.25)	38.67 ± 0.56 (37.60-39.78)	0.73 ± 0.86 (0.57-0.90)
Other (n)	90.82 ± 1.90 (87.26-94.61)	29.48 ± 0.66 (28.19-30.82)	37.95 ± 0.90 (36.22-39.75)	38.44 ± 0.90 (36.68-40.20)	0.38 ± 0.16 (0.06-0.70)
P value	.970	.622	.755	.818	.054
SES*					
Low (n)	99.40 ± 5.96 (86.63-112.17)	31.93 ± 2.02 (27.60-36.27)	42.00 ± 2.69 (36.24-47.76)	41.93 ± 2.74 (36.07-47.80)	0.67 ± 0.29 (0.05-1.29)
Medium (n)	90.57 ± 1.68 (87.24-93.90)	28.81 ± 0.53 (27.77-29.85)	38.08 ± 0.76 (36.59-39.58)	38.80 ± 0.82 (37.17-40.43)	0.61 ± 0.14 (0.34-0.88)
High (n)	91.66 ± 1.27 (89.15-94.17)	29.59 ± 0.42 (28.75-30.42)	38.59 ± 0.58 (37.44-39.73)	38.80 ± 0.62 (37.58-40.01)	0.71 ± 0.10 (0.51-0.92)
P value†	.206	.127	.219	.403	.839
WFLZ at birth					
<2.0 (n)	91.43 ± 5.80 (80.59-103.08)	28.93 ± 1.52 (26.06-32.07)	37.43 ± 2.32 (32.92-42.07)	39.43 ± 2.98 (33.80-45.36)	**−0.08 ± 0.41 (−0.87-0.74)**
≥2.0 (n)	91.11 ± 0.99 (89.20-93.06	29.29 ± 0.32 (28.66-29.93)	38.35 ± 0.45 (37.48-39.23)	38.71 ± 0.48 (37.78-39.66)	**0.74 ± 0.08 (0.59-0.89)**
P value	.956	.815	.688	.809	**.040**
Maternal prepregnancy weight status					
Healthy weight (n)	93.14 ± 2.06 (89.06-97.23)	29.34 ± 0.62 (28.11-30.58)	39.39 ± 0.93 (37.60-41.22)	39.89 ± 1.02 (37.85-41.88)	**0.49 ± 0.09 (0.32-0.66)**
Overweight or obese (n)	89.44 ± 1.10 (88.27-92.63)	29.19 ± 0.37 (28.48-29.91)	37.98 ± 0.50 (37.01-38.96)	38.37 ± 0.53 (37.33-39.42)	**1.22 ± 0.15 (0.91-1.51)**
P value	.242	.827	.175	.182	**<.001**
Delivery mode					
Vaginal (n)	91.22 ± 1.08 (89.16-93.37)	29.29 ± 0.35 (28.61-29.98)	38.32 ± 0.48 (37.41-39.28)	38.83 ± 0.53 (37.83-39.89)	−0.65 ± 0.09 (0.49-0.83)
Cesarean (n)	92.05 ± 2.07 (87.86-96.00)	29.51 ± 0.65 (28.22-30.77)	38.90 ± 0.91 (37.18-40.70)	38.98 ± 1.01 (37.83-39.89)	0.76 ± 0.16 (0.43-1.08)
P value	.726	.761	.572	.896	.584
Feeding mode at 3-moa					
Exclusively breastfed (n)	91.20 ± 1.11 (89.10-93.40)	29.31 ± 0.36 (28.62-30.04)	38.23 ± 0.50 (37.27-39.27)	38.79 ± 0.56 (37.72-39.91)	0.63 ± 0.09 (0.45-0.81)
Mixed- or formula-fed (n)	90.88 ± 1.74 (87.47-94.40)	29.19 ± 0.59 (28.07-30.38)	38.47 ± 0.77 (36.95-39.99)	38.51 ± 0.85 (36.89-40.20)	0.72 + 0.13 (0.45-0.99)
P value	.871	.854	.783	.780	.589

All values are reported as mean ± SE (95% CI). All statistics were from an independent-samples *t*-test with SE, 95% CIs, and *P* values calculated using the bootstrap with 5000 samples unless noted otherwise. Bold type denotes a significant result ($P < .05$).
*SES is a composite score of mother's level of education and household income, which was divided into low, medium, and high SES.
†Represents *P* value for between-group difference by ANOVA.

Table IV. Path model A: Standardized regression coefficients of the direct and indirect effects of adherence to AAP guidelines on BRIEF-P indices, with WFLZ at 24-moa as a mediator variable

Variables	Point estimate (95% CI)	SE	*P* value	Adjusted *P* value*
Direct path from guidelines to WFLZ				
Physically active every day	−0.005 (−0.116 to 0.106)	0.057	.94	1.0
No more than 60 minutes of screen time	−0.055 (−0.166 to 0.053)	0.057	.33	.637
At least 5 servings of fruits and vegetables	0.096 (−0.032 to 0.219)	0.065	.14	.392
Limit SSB	0.107 (−0.028 to 0.234)	0.066	.11	.411
Direct path from WFLZ to BRIEF-P indices				
Overall EF	−0.003 (−0.123 to 0.112)	0.06	.96	1.0
Cognitive flexibility	−0.033 (−0.152 to 0.086)	0.061	.58	.902
Inhibitory self-control	0.006 (−0.116 to 0.124)	0.060	.93	1.0
Emergent metacognition	0.011 (−0.108 to 0.123)	0.059	.86	1.0
Effects of guidelines on overall EF				
Physically active every day				
Total	−0.082 (−0.190 to 0.029)	0.056	.14	.413
Direct	−0.082 (−0.190 to 0.029)	0.056	.14	.436
Indirect	0.00002 (−0.007 to 0.008)	0.003	.997	.997
No more than 60 min of screen time				
Total	**−0.125 (−0.233 to −0.008)**	**0.057**	**.03**	.56
Direct	**−0.125 (−0.234 to −0.008)**	**0.057**	**.03**	.42
Indirect	0.0002 (−0.009 to 0.010)	0.005	.97	1.0
At least 5 servings of fruits and vegetables				
Total	−0.053 (−0.154 to 0.046)	0.051	.30	.622
Direct	−0.053 (−0.156 to 0.047)	0.052	.31	.62
Indirect	−0.0003 (−0.016 to 0.014)	0.007	.97	1.0
Limit SSB				
Total	−0.114 (−0.237 to 0.010)	0.064	.08	.373
Direct	−0.113 (−0.237 to 0.010)	0.065	.08	.407
Indirect	−0.003 (−0.017 to 0.014)	0.008	.97	1.0
Effects of guidelines on cognitive flexibility				
Physically active every day				
Total	−0.044 (−0.155 to 0.065)	0.056	.43	.753
Direct	−0.044 (−0.155 to 0.065)	0.056	.43	.730
Indirect	0.0002 (−0.008 to 0.009)	0.004	.97	1.0
No more than 60 min of screen time				
Total	−0.090 (−0.202 to 0.027)	0.059	.12	.395
Direct	−0.092 (−0.205 to 0.026)	0.059	.12	.420
Indirect	0.002 (−0.007 to 0.014)	0.005	.71	1.0
At least 5 servings of fruits and vegetables				
Total	−0.056 (−0.148 to 0.034)	0.047	.23	.56
Direct	−0.053 (−0.148 to 0.040)	0.048	.27	.630
Indirect	−0.003 (−0.020 to 0.011)	0.007	.66	.999
Limit SSB				
Total	−0.090 (−0.226 to 0.046)	0.069	.19	.507
Direct	−0.086 (−0.224 to 0.051)	0.070	.22	.560
Indirect	−0.004 (−0.023 to 0.010)	0.008	.66	.973
Effect of guidelines on inhibitory self-control				
Physically active every day				
Total	−0.044 (−0.153 to 0.071)	0.057	.44	.725
Direct	−0.044 (−0.154 to 0.071)	0.058	.44	.704
Indirect	−0.00003 (−0.007 to 0.008)	0.004	.99	1.0
No more than 60 min of screen time				
Total	**−0.143 (−0.248 to −0.029)**	**0.056**	**.01**	.560
Direct	**−0.142 (−0.248 to −0.029)**	**0.056**	**.01**	.280
Indirect	−0.0003 (−0.010 to 0.010)	0.005	.95	1.0
At least 5 servings of fruits and vegetables				
Total	−0.047 (−0.146 to 0.049)	0.050	.35	.632
Direct	−0.047 (−0.148 to 0.051)	0.051	.35	.653
Indirect	0.001 (−0.014 to 0.016)	0.007	.94	1.0
Limit SSB				
Total	−0.107 (−0.231 to 0.019)	0.064	.10	.400
Direct	−0.107 (−0.232 to 0.020)	0.065	.10	.431
Indirect	0.001 (−0.017 to 0.016)	0.008	.94	1.0
Effects of guidelines on emergent metacognition				
Physically active every day				
Total	**−0.116 (−0.224 to −0.004)**	**0.056**	**.04**	.448
Direct	**−0.116 (−0.225 to −0.005)**	**0.056**	**.04**	.373
Indirect	0.001 (−0.008 to 0.007)	0.004	.99	1.0
No more than 60 min of screen time				
Total	**−0.112 (−0.222 to 0.002)**	**0.057**	**.047**	.376
Direct	**−0.111 (−0.221 to 0.002)**	**0.057**	**.049**	.343

(*continued*)

Table IV. Continued

Variables	Point estimate (95% CI)	SE	*P* value	Adjusted *P* value*
Indirect	−0.001 (−0.010 to 0.010)	0.005	.90	1.0
At least 5 servings of fruits and vegetables				
Total	−0.053 (−0.148 to 0.045)	0.049	.28	.627
Direct	−0.054 (−0.150 to 0.046)	0.050	.28	.603
Indirect	0.001 (−0.013 to 0.016)	0.007	.88	1.0
Limit SSB				
Total	−0.117 (−0.239 to 0.003)	0.062	.06	.373
Direct	−0.118 (−0.240 to 0.001)	0.063	.06	.336
Indirect	0.001 (−0.015 to 0.016)	0.007	.88	1.0

Statistics in bold type denote a significant result ($P < .05$) before correction for multiple comparisons. Covariates included in the model were child sex, SES, ethnicity, WFLZ at birth, and maternal prepregnancy weight status.

*P values are corrected for multiple comparisons using a Benjamini–Hochberg procedure with a false discovery rate of 0.1 to determine significance after correction.

Table V. Path model B: Standardized regression coefficients of the direct and indirect effects of health behaviors on BRIEF-P indices, with WFLZ at 24-moa as a mediator variable

Variables	Point estimate (95% CI)	SE	*P* value	Adjusted *P* value*
Direct path from health behaviors to WFLZ				
Number of physical activities	−0.024 (−0.148 to 0.086)	0.060	.69	.991
Screen time	0.116 (−0.012 to 0.233)	0.062	.06	.336
Fruit and vegetable servings	**0.131 (0.006-0.252)**	**0.062**	**.04**	.249
SSB kcal	−0.091 (−0.229 to 0.014)	0.062	.14	.713
Direct path from WFLZ to BRIEF-P indices				
Overall EF	−0.032 (−0.154 to 0.088)	0.062	.60	1.0
Cognitive flexibility	−0.061 (−0.179 to 0.059)	0.061	.32	.943
Inhibitory self-control	−0.015 (−0.139 to 0.110)	0.062	.82	.937
Emergent metacognition	−0.021 (−0.143 to 0.096)	0.061	.73	.973
Effects of health behaviors on overall EF				
Number of physical activities				
Total	−0.028 (−0.168 to 0.088)	0.065	.67	.987
Direct	−0.029 (−0.170 to 0.088)	0.065	.66	.999
Indirect	0.001 (−0.008 to 0.012)	0.005	.17	.680
Screen time				
Total	**0.254 (0.114-0.378)**	**0.067**	**<.001**	**.008**
Direct	**0.257 (0.118-0.384)**	**0.067**	**<.001**	**.056**
Indirect	−0.004 (−0.023 to 0.011)	0.008	.65	1.0
Fruit and vegetable servings				
Total	−0.067 (−0.179 to 0.056)	0.059	.26	.971
Direct	−0.063 (−0.179 to 0.063)	0.061	.30	.988
Indirect	−0.004 (−0.026 to 0.013)	0.009	.65	1.0
SSB kcal				
Total	−0.045 (−0.169 to 0.125)	0.075	.54	1.0
Direct	−0.048 (−0.172 to 0.125)	0.075	.52	1.0
Indirect	0.003 (−0.009 to 0.022)	0.008	.70	.98
Effects of health behaviors on cognitive flexibility				
Number of physical activities				
Total	−0.035 (−0.171 to 0.075)	0.063	.57	1.0
Direct	−0.037 (−0.175 to 0.075)	0.063	.56	1.0
Indirect	0.001 (−0.009 to 0.014)	0.005	.78	.929
Screen time				
Total	**0.210 (0.076-0.336)**	**0.067**	**.002**	**.014**
Direct	**0.217 (0.082-0.342)**	**0.067**	**.001**	**.028**
Indirect	−0.007 (−0.026 to 0.008)	0.009	.41	.998
Fruit and vegetable servings				
Total	−0.031 (−0.142 to 0.090)	0.060	.60	1.0
Direct	−0.024 (−0.139 to 0.100)	0.061	.70	.956
Indirect	−0.008 (−0.030 to 0.008)	0.010	.40	1.0
SSB kcal				
Total	−0.031 (−0.144 to 0.126)	0.068	.65	1.0
Direct	−0.037 (−0.151 to 0.123)	0.068	.59	1.0
Indirect	0.006 (−0.006 to 0.028)	0.008	.51	1.0
Effects of health behaviors on inhibitory self-control				
Number of physical activities				
Total	−0.005 (−0.133 to 0.101)	0.059	.93	.93
Direct	−0.005 (−0.134 to 0.101)	0.059	.93	.964
Indirect	0.0004 (−0.008 to 0.011)	0.004	.93	.947
Screen time				
Total	**0.229 (0.098-0.351)**	**0.064**	**<.001**	**.019**
Direct	**0.231 (0.099-0.354)**	**0.064**	**<.001**	**.014**
Indirect	−0.002 (−0.020 to 0.015)	0.008	.84	.922
Fruit and vegetable servings				
Total	−0.088 (−0.200 to 0.032)	0.059	.14	.653
Direct	−0.086 (−0.202 to 0.039)	0.061	.16	.689
Indirect	−0.002 (−0.021 to 0.016)	0.009	.83	.93
SSB kcal				
Total	−0.025 (−0.162 to 0.163)	0.082	.76	.946
Direct	−0.027 (−0.166 to 0.166)	0.083	.75	.977
Indirect	0.001 (−0.011 to 0.019)	0.007	.86	.926
Effects of health behaviors on emergent metacognition				
Number of physical activities				
Total	−0.039 (−0.181 to 0.072)	0.065	.54	1.0
Direct	−0.040 (−0.183 to 0.074)	0.065	.54	1.0
Indirect	0.001 (−0.008 to 0.011)	0.004	.91	.962

(*continued*)

Table V. Continued

Variables	Point estimate (95% CI)	SE	*P* value	Adjusted *P* value*
Screen time				
Total	**0.255 (0.118-0.012)**	**0.066**	**<.001**	**.011**
Direct	**0.257 (0.120-0.381)**	**0.067**	**<.001**	**.009**
Indirect	−0.002 (−0.021 to 0.012)	0.008	.76	.925
Fruit and vegetable servings				
Total	−0.062 (−0.171 to 0.062)	0.058	.29	1.0
Direct	−0.059 (−0.172 to 0.067)	0.059	.32	.996
Indirect	−0.003 (−0.172 to 0.014)	0.009	.76	.967
SSB kcal				
Total	−0.059 (−0.172 to 0.094)	0.068	.38	1.0
Direct	−0.061 (−0.175 to 0.092)	0.068	.37	1.0
Indirect	0.002 (−0.010 to 0.020)	0.007	.79	.922

Statistics in bold type denote a significant result ($P < .05$). Covariates included in the model were child sex, SES, ethnicity, WFLZ at birth, and maternal prepregnancy weight status.
**P* values are corrected for multiple comparisons using a Benjamini–Hochberg procedure with a false discovery rate of 0.1 to determine significance after correction.

3. L’étude controlee randomisée (randomized controlled trial)

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Parent, child, and family outcomes following Acceptance and Commitment Therapy for parents of autistic children: A randomized controlled trial

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Abstract

Emerging research shows that Acceptance and Commitment Therapy (ACT) may improve mental health for caregivers. Parents of autistic children, adolescents, and adults ($N = 54$) were randomly assigned to either complete a brief group-based ACT intervention or remain on the waitlist. Participants completed surveys immediately prior to randomization, and 3-, 7-, and 17-weeks post-randomization. The primary outcome was depression symptoms and secondary outcomes included stress, goal attainment, positive affect, ACT psychological processes, child mental health, and family functioning. Mixed effects linear models testing Group × Time interaction indicated the Treatment group ($n = 27$) demonstrated greater post-intervention improvements than the Waitlist group ($n = 27$) in parent depression ($p = .03$, $d = -0.64$) and family distress ($p = .04$, $d = -0.57$). Treatment group parents also reported greater short-term gains in positive affect ($p = < .001$, $d = 0.77$) and personal goal attainment ($p = .007$, $d = 0.80$), compared to the Waitlist group. Although there was no significant Group × Time interaction for other outcomes, stress ($b = -2.58$, $p = .01$), defusion ($b = -3.78$, $p = .001$), and experiential avoidance ($b = -4.22$, $p = .01$) showed improvement for the Treatment group, but not the Waitlist group, at post-intervention. All Treatment group improvements were maintained at follow-up. Results suggest that a brief ACT group intervention is efficacious for improving some aspects of mental health for parents of autistic children.

Lay abstract

Parents of autistic children commonly experience difficulties with their own mental health. This study looked at the effects of a brief group-based Acceptance and Commitment Therapy program, developed for parents of autistic children, youth, and adults. ACT focuses on increasing psychological flexibility, which is the ability to be mindful and accepting of difficult thoughts and experiences, shown to be important for mental wellness. Participants included 54 parents of autistic people, ages 3–34. Parents were randomly divided into two groups: a Treatment group that received the intervention right away, and a Waitlist group that completed the program after the Treatment group completed the trial. All parents filled out questionnaires right before the program began, and at 3, 7, and 17 weeks after randomization. Compared to the group that was waiting to participate in the program, parents in the Treatment group reported greater improvements in depression and family distress, and these improvements were still present 4 months later. Parents in the Treatment group also reported short-term improvements in their positive feelings and personal goals, compared to those waiting. Results showed that ACT may help improve some aspects of mental health for parents of autistic children, but further research is recommended.

Keywords

acceptance and commitment therapy, autism, caregivers, interventions—psychosocial/behavioral, mental health

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Parents of autistic children[1] commonly experience greater levels of stress, anxiety, and depression compared to parents of children who do not have a disability, and their levels of chronic stress have been shown to surpass levels experienced by parents of children with other developmental disabilities (Bitsika & Sharpley, 2004; Estes et al., 2009). Given the high stress experienced by parents of autistic children (Hayes & Watson, 2013), it follows that parents would experience their own challenges that require parent-focused support.

Acceptance and Commitment Therapy (ACT) has emerged as an intervention with a strong evidence base supporting its use for caregivers (Han et al., 2021). The ACT framework focuses on helping individuals improve psychological flexibility, which involves increasing openness and mindful awareness in the present moment, and acceptance of difficult experiences (Hayes et al., 2006). The overall aim of ACT is not to decrease psychological distress, but to increase one's effectiveness in pursuing values that are personally meaningful, even in the context of difficult symptoms or circumstances (Harris, 2006). In addition to rewards and positive impacts of raising an autistic child that many parents report (Kayfitz et al., 2010), parents of autistic children also describe making significant adjustments to their lives to support their child's care, such as career changes, or devoting very limited time to social or personal activities (Meirsschaut et al., 2010). They may have difficulty navigating inaccessible care systems, unsupportive social systems, or adjusting to a parenting experience that can involve a high level of daily childcare demands (Meirsschaut et al., 2010). As a result, parents may experience difficult thoughts and feelings in response to these real challenges and stressors. Acceptance, therefore, may be a more appropriate framework than treatment modalities that focus on problem-solving. The use of psychological acceptance has also been shown to be an important protective factor in the well-being of parents of autistic children (Jones et al., 2014; Weiss et al., 2012). In addition, parents of autistic children report that they employ more avoidance coping (i.e. cognitive distancing/minimizing; escape behaviors) than parents of children without autism, which may be related to increased strain on the family system (Sivberg, 2002). Avoidance coping, although sometimes adaptive in the short term, is associated with increased stress and mental health problems over time (Hastings et al., 2005). As such, these parents may benefit from interventions that help decrease the use of psychological avoidance and promote psychological acceptance.

There is a growing literature demonstrating the effectiveness of ACT among parents of autistic children. In the first published study of ACT for parents of autistic children, Blackledge and Hayes (2006) delivered a 2-day workshop to 20 parents. Participants demonstrated post-intervention improvements in self-reported depression, distress, experiential avoidance, and cognitive fusion. In another study, two 4 h ACT workshops were delivered to five mothers of children with intellectual and developmental disabilities who displayed high levels of challenging behavior (Reid et al., 2016). Analysis of post-intervention interviews revealed that parents reported a better ability to cope with stress, increased emotional wellbeing for themselves and their child, and the ability to take a more mindful perspective on their difficulties. In a multiple baseline repeated measures analysis of three mothers of autistic children who participated in six 90-min sessions of ACT, parents increased in their use of values-guided behaviors (Gould et al., 2018) post-training and 6 months later.

In India, Poddar and colleagues (2015) delivered ACT individually to five mothers of autistic children over 10 sessions. Improvements in parent self-reports of depressive and anxious symptoms, psychological flexibility, and quality of life were noted post-intervention. In Iran, an 8-week group ACT intervention provided to 12 mothers was superior to 8 weeks of individual counseling for reducing self-reports of depression and experiential avoidance, although group assignment was not random (Joekar et al., 2016).

In a study with a slightly larger sample, Lunsky and colleagues (2018) delivered a brief ACT workshop to 29 mothers of autistic children and young adults. In addition to having ACT expertise, facilitators were also parents of autistic children. Post-intervention, improvements were seen in parents' self-reports of depression, stress, perceived physical health, and social isolation, and improvements in all but social isolation were maintained at follow-up. A second study of this cohort reported that mothers also indicated improvements in cognitive fusion, psychological flexibility, and values-consistent action, which were maintained at follow-up (Fung et al., 2018a).

One of the only randomized controlled trials (RCTs) of ACT for parents focused on parents of children with cerebral palsy, where researchers added a 4 h ACT training for parents who were already receiving a parenting intervention, compared to a randomly assigned waitlist control group and a parent training only group. The addition of ACT training resulted in improved parent reports of child quality of life and reduced parental depression and stress, compared to the waitlist group (Whittingham et al., 2015). This study did not compare receiving only ACT to a control condition, making it difficult to determine the relative impact of ACT on the improvements observed. In the only RCT to date testing the efficacy of ACT for parents of autistic children (age 5–13), nine parents receiving Applied Behavior Analysis (ABA) participated in a 4 h ACT workshop, compared to nine parents who received ABA only (Hahs et al., 2018). The ACT group demonstrated significantly greater improvements compared to the ABA-only group in self-reports of mindfulness, experiential avoidance, cognitive fusion, consistency toward personal values, and depression, but no follow-up was employed.

ACT has yet to be evaluated in parents of autistic children using both a randomized controlled design and a large

enough sample to increase generalizability to the larger population of parents of autistic children. Further, while there is evidence that in families of autistic children, parent-focused interventions can also serve to improve child behavior and mental health (Neece, 2014; Singh et al., 2006, 2014) and family adjustment (Tonge et al., 2006), no studies have examined child and family outcomes following ACT interventions for parents. Finally, as ACT is meant to improve psychological flexibility and positive functioning while not necessarily decreasing symptoms, it is essential that evaluations of ACT include outcome measures that assess positive aspects of parent functioning as well, rather than focusing only on reduction of problems.

Aim

The current study tested the efficacy of a brief, manualized group-based ACT workshop developed for parents of children, adolescents, and adults[2] with neurodevelopmental disabilities, including autism. A randomized controlled design compared a Treatment group that completed the program immediately to a Waitlist control condition. The primary outcome was reduction in parent depression, and secondary outcomes included other aspects of parent mental health, positive functioning, and goal attainment. We also investigated the extent to which the intervention impacted the mental health of children of participating parents, and overall family functioning. We predicted that parents who were offered the ACT program would report improvements in their mental health, positive functioning, goal attainment, and in their child's mental health and family's functioning, relative to the waitlist condition.

Method

Participants

Participants included 54 parents of autistic children, adolescents, and adults ages 3 to 34 years. To be included in the study, parents had to provide documentation of their child's autism diagnosis from a health care practitioner qualified to diagnose autism in Ontario, Canada (i.e. clinical psychologist, pediatric/general practitioner physician, or psychiatrist). Additional inclusion criteria were the ability to speak English and identify therapy goals, and no prior exposure to ACT.

Program description

The intervention was a three-session workshop, developed by Fung and colleagues (2018b) and described in detail by Lunsky and colleagues (2018). The first session involved a 3 h introduction to the ACT processes, and participants were introduced to mindfulness through an experiential guided imagery exercise. The second full-day session occurred the following day and included didactic, experiential, and mindfulness activities to demonstrate the concepts of ACT, and linked material to the experience of parenting an autistic person. Guided activities were interspersed with paired sharing, group discussion, and videos. At the end of session 2, participants committed to engaging in one action that aligned with their values. A 3 h refresher session was conducted 1 month later.

Measures

Sample characteristics. The parent-report *Social Communication Questionnaire–Lifetime version* (SCQ; Rutter et al., 2003) was used to measure autism symptoms. The parent-report *Scales of Independent Behavior–Revised* Short Form (SIB-R; Bruininks et al., 1996) measured adaptive functioning. The SIB-R uses a standard score ($M = 100$, $SD = 15$, Range: 0–200).

Primary outcome

Depression. Symptoms of depression were measured using the 7-item Depression subscale of the *Depression Anxiety Stress Scale–21* (DASS-21; Lovibond & Lovibond, 1995). Item responses reference the past week and use a 4-point scale (0 = Never to 3 = Almost Always). Subscale scores can fall into the ranges of normal, mild, moderate, severe, or extremely severe. These severity labels characterize the range of scores in the population; a mild score does not indicate mild level of disorder and is below the typical severity of those seeking help (Lovibond & Lovibond, 1995). Therefore, a "moderate" score (7 or higher) was used to define depression of clinical concern. Internal consistency for Depression was good in the current sample ($\alpha = 0.89$).

Secondary outcomes

Parent mental health and positive functioning. The 7-item Stress subscale of the DASS-21 was used to measure symptoms of stress. Internal consistency was $\alpha = 0.84$ in the current sample.

Two subscales of the *Parenting Stress Index–Fourth Edition* (PSI-4; Abidin, 2012) were used. The 6-item Isolation subscale measures perceived social isolation and the 5-item Health subscale measures perceived physical health in relation to parenthood. Parents were asked to reference the past week. Items were rated on 5-point scale (1 = Strongly Agree to 5 = Strongly Disagree). The PSI-4 has been previously used with parents of autistic children (Feinberg et al., 2014; Lunsky et al., 2018; Thullen & Bonsall, 2017). In this sample, internal consistency was acceptable (Isolation $\alpha = 0.76$, Health $\alpha = 0.70$).

The *Positive & Negative Affect Schedule* (PANAS; Crawford & Henry, 2004) is a 20-item measure that assesses respondents' experiences of different positive and negative emotions over the past week. Only the positive affect subscale was used, which has items that include 10

positive (i.e. active, determined) mood words, and respondents report the extent to which they experienced each mood over the last week (1 = Not at all to 5 = Extremely). The PANAS demonstrated excellent internal consistency in the current sample (α = .90 for Positive Affect) and in previous studies with parents of autistic children the Positive Affect subscale has been associated with other positive parental outcomes (e.g. optimism, psychological well-being; Ekas et al., 2010).

To assess goal attainment, at screening, participants identified three goals they hoped to attain through completing the workshop. At baseline, goals were refined with the assistance of a clinically-trained graduate student to ensure they were Specific, Measurable, Attainable, Relevant, and Timed, and *Goal Attainment Scaling*, a procedure typically used in rehabilitation intervention, was used to track progress (Bovend'Eerdt et al., 2009). For each goal, operationalized definitions were created to describe the participant's target behaviors if their goal was achieved (0), slightly exceeded (+1), greatly exceeded (+2), not quite achieved (−1) or nowhere near (−2), to create an individualized reference guide. At each assessment time point, participants were electronically sent their guide, which they referenced to select the number that represented their progress toward each goal.

For the analysis, participants' highest goal attainment score at each time point was used (i.e. the goal for which the participant indicated the highest achievement rating could differ between time points). We analyzed participants' highest achieved goal (and thus the goal they were prioritizing) rather than totaling achievement of all goals, as the three goals may not have been considered equally important to participants. Focusing on one priority goal allowed for the examination of what meaningful change in one area could look like. Other research using goal attainment scaling has found utility in selecting the highest-ranking goal for analysis (Jennings et al., 2018; McGarrigle & Rockwood, 2020). To capture the effect of the program on whether goals were attained or not, the resulting values were dichotomized into 0 (*not quite achieved* or *nowhere near achieved*) and 1 (*achieved* or *slightly exceeded* or *greatly exceeded*). Past research on goal scaling has also analyzed reports in this manner (e.g. Chiarello et al., 2020; Jennings et al., 2018).

ACT process measures. The *Acceptance & Action Questionnaire-II* (AAQ-II; Bond et al., 2011) was used to measure psychological flexibility and acceptance. The AAQ-II is a 7-item questionnaire on which items are rated on a 7-point scale to provide an overall score. Higher scores indicate greater levels of acceptance and less experiential avoidance. The AAQ-II showed excellent internal consistency, α = .92. The AAQ and AAQ-II have been used previously in studies of parents of autistic children (Blackledge & Hayes, 2006; Hahs et al., 2018 respectively).

The *Valued Living Questionnaire* (VLQ; Wilson et al., 2010) is a two-part questionnaire that measures the extent to which respondents are in contact with their chosen values in everyday life. First, the participant indicates the importance they place on 10 life areas (e.g. friendship, work, spirituality), on a 10-point scale from Not at all important to Very important. In the second part, they indicate how consistent their actions currently are in each valued area on a 10-point scale, ranging from Completely inconsistent to Completely consistent. In this sample, the VLQ demonstrated acceptable internal consistency (Importance α = .69, Consistency α = .83, Valued Living Composite α = .76). The VLQ has been used in other studies with parents (Fung et al., 2018a).

The *Cognitive Fusion Questionnaire* (CFQ; Gillanders et al., 2014) is a 7-item measure used to assess the degree to which respondents report currently feeling distressed by, getting caught up with, or struggling to let go of thoughts. Statements are rated on a 7-point Likert-type scale (1 = Never true to 7 = Always true). Internal consistency was excellent in the current sample, α = .93. The CFQ has been used in studies of parents of autistic children (Fung et al., 2018a; Hahs et al., 2018).

The *Bangor Mindful Parenting Scale* (BMPS; Jones et al., 2014) is a 15-item questionnaire that measures parents' degree of mindfulness in their parenting and interactions with their children. Respondents rate items on a 4-point scale (0 = Never true to 3 = Always true). It has been used previously in studies of parents of children with autism and other disabilities (Lunsky et al., 2015). Internal consistency for the total score was α = .79 in the current sample.

Youth mental health and family functioning. The *Strengths & Difficulties Questionnaire* (SDQ; Goodman, 2001) assesses mental health difficulties broadly in children and youth. In line with SDQ guidelines, when administered at baseline, parents were asked to reference the past 6 months, and at subsequent time points the reference period was the past 1 month. The measure has subscales for Emotional Symptoms, Conduct Problems, Hyperactivity-Inattention, Peer Problems, Prosocial Behavior, Impact, and Total Difficulties. Respondents provided item ratings on a 3-point scale (1 = Not true to 3 = Certainly true). Internal consistencies were acceptable for most subscales of the SDQ in the current sample, ranging from α = .69 to .72, except Peer Problems (α = .46). The SDQ is often used in studies of autistic children (Allik et al., 2006; Chalfant et al., 2007).

The *Brief Family Distress Scale* (BFDS; Weiss & Lunsky, 2011) was used to obtain a measure of the current level of crisis parents perceive in their family. Respondents indicate, on a scale from 1 to 10, which numbered statement most accurately represents the state of their family's current distress. Statements range from 1 = *everything is fine, my family and I are not in crisis at all*, 5 = *things are*

very stressful, but we are getting by with a lot of effort, 10 = *we are currently in crisis, and it could not get any worse*. The BFDS has been used in other studies of parents of children with developmental disabilities (Hastings, 2016; McKenzie et al., 2017).

The General Functioning subscale of the *McMaster Family Assessment Device* (FAD; Epstein et al., 1983) was used as a measure of family functioning over the past 2 weeks. Responses for each of the 12 items are provided on a 4-point scale, ranging from Strongly agree to Strongly disagree. This tool has been used in parents of children with disabilities (Brown et al., 2015) and had good internal consistency ($\alpha = .82$) in the current sample.

Procedure

Recruitment. The study was approved by ethics boards at York University, the Centre for Addiction and Mental Health, and Surrey Place in Toronto, Ontario. This study was registered prior to data collection with the ISRCTN registry (97093664). See Supplemental Table 1 for the Consolidated Standards of Reporting Trials (CONSORT) Checklist. Participants were recruited from postings on websites of child autism community agencies, autism service e-newsletters, and referrals from the community between January-November 2019. Interested parents were invited to participate in a telephone screening interview to assess eligibility. For participants who met screening criteria, study procedures were explained, and written informed consent was obtained. As shown in Figure 1, 77 participants were screened, with 7 excluded for not meeting inclusion criteria, and 7 declining to participate. Power analyses using G*Power 3.1 indicated that a sample size of 55 would be sufficient to detect medium effects. No participants reported experiencing harm from participation in the trial.

Randomization. Following the baseline appointment, the first author randomized parents into either the Treatment or Waitlist group using online randomization software (Urbaniak & Plous, 2015). Participant randomization was stratified by gender, to ensure approximately equal numbers of males and females in each group. Sixty-three participants were randomized. If partners participated together, one was randomly excluded from the analysis ($n = 9$), leaving 27 in each group. Parents in the Treatment group participated in their assigned ACT workshop within 2 weeks. Waitlisted parents participated in the intervention after the follow-up time point.

Study design. Following participant screening, baseline data were collected within 2 weeks prior to the commencement of the workshop. Parents completed demographic and evaluation measures online, and goals were defined during a phone appointment. Child social communication abilities and adaptive functioning were assessed at baseline; the SCQ was administered online, and the SIB-R was completed during the phone appointment in interview format.

All outcome measures (DASS-21, PANAS, PSI-4, AAQ-II, VLQ, CFQ, BMPS, BFDS, FAD, SDQ, goal attainment) were administered online at 7 weeks post-baseline (post-refresher/ Time 3) and 17 weeks post-baseline (follow-up/Time 4). The primary outcome measure (DASS-21 Depression), as well as four additional brief measures (DASS-21 Stress, PANAS, BFDS, goal attainment) were re-administered at the mid-point of treatment (3 weeks post-randomization/Time 2). These measures were selected to obtain a brief snapshot of four areas of functioning (i.e. mental health, positive functioning, family distress, and behavior change) immediately after the first two intervention sessions/wait. Following Time 4 data collection, the Waitlist group participated in the intervention.

Community involvement

Families of autistic individuals were involved in this study. Parents of autistic people designed the intervention (Fung et al., 2018b) and co-facilitated every workshop, working in partnership with clinicians. No autistic people were involved in the research process.

Data analysis

Using SPSS version 27, we performed a series of linear mixed effects regression analyses at each post-treatment time point of the relationship between Group and each outcome measure (DASS-21 Depression, DASS-21 Stress, PANAS, PSI-4, AAQ-II, VLQ, CFQ, BMPS, BFDS, FAD, SDQ). As fixed effects, we entered Group (Treatment/ Waitlist), Time (two levels: Baseline and Time 2 or 3), and Group × Time interaction into the model. The interaction directly tested whether Treatment and Waitlist groups showed a different pattern of change in outcome measures at the different study time points. As random effects, we included an intercept for subjects to account for repeated measurements. Mixed effects models accommodate for missing values using the maximum likelihood method, and therefore, this analysis included all randomized participants ($N = 54$ [9 partners excluded]).

Cohen's *d* effect sizes were calculated using the difference in change scores between Treatment and Waitlist groups, divided by the pooled standard deviation at baseline. Effect size magnitudes were defined as: negligible (<0.20), small (0.20–0.49), medium (0.50–0.79), and large ($\geqslant 0.80$) (Cohen, 1992).

Differences between the Treatment and Waitlist groups in their goal attainment (i.e. goal achieved/not achieved) were tested using Pearson chi-square analyses. Missing goal attainment values were imputed in SPSS using

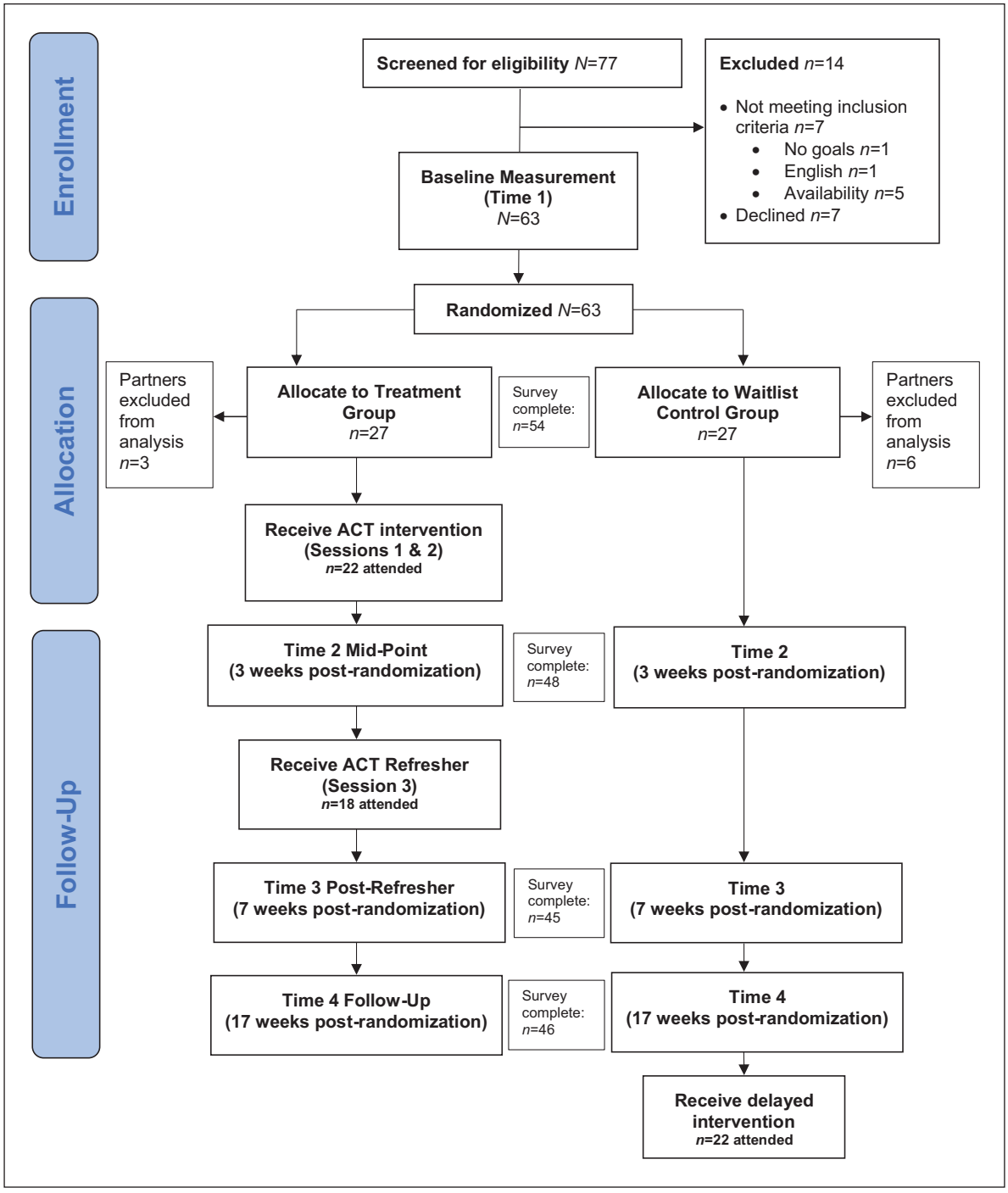

Figure 1. CONSORT flow diagram.

multiple imputation, including all demographic variables, baseline scores, and variables in the analysis as predictors. Chi-square values from 10 imputed datasets were pooled in R using the micombine.chisquare function from miceadds (Enders, 2010). This function produces a combined statistic, which is approximately F-distributed. Odds ratio was used as the effect size measure.

Two analyses included treatment completers only from the Treatment group ($n = 18$), who completed the full 3-day intervention with refresher, in addition to all Waitlist participants ($n = 27$). First, for depression, the number of participants in each group who moved from the clinical to non-clinical range was calculated at Time 2 and Time 3. The interaction between Group and improvement to the

non-clinical range was tested using Pearson chi-square analyses. Second, to assess whether improvements were maintained at 4-month follow-up (only possible for the Treatment group), linear mixed effects regression analyses were performed on treatment completers ($n=18$) with Time (two levels: Time 3 and Time 4) as the fixed effect, and subjects' intercept as a random effect.

Results

Baseline demographics

There were no significant differences in participant characteristics at baseline (see Table 1). Participants were 83% female, two thirds had completed university, and approximately one third were currently taking psychotropic medication. All 54 participants completed the survey at Time 1. At Time 2, 48 participants completed the survey (88.9%), at Time 3, 45 participants (83.3%), and at Time 4, 46 participants (85.2%).

Primary outcome

Depression. At Time 2 (mid-point), following the main 2-day workshop, parents in the Treatment group demonstrated significant reductions in depression scores ($b=-3.21$, $p=.001$), whereas these improvements were not seen for the Waitlist group ($b=0.46$, $p=.51$), with a large Time × Condition interaction (see Table 2). At Time 3 (post-refresher), the Treatment group showed sustained reductions in depression scores ($b=-2.60$, $p=.03$) and the Waitlist continued to show no reduction in depression ($b=0.08$ $p=.88$), with a medium Time × Condition effect.

In the Treatment group, 18 participants completed the full 3-day intervention, including the refresher. Improvements observed in the Treatment group were maintained at 4-month follow-up, with no differences among treatment completers between Time 3 and Time 4 depression scores ($b=-0.14$; $p=.90$; see Supplemental Table 3 for Time 4 analyses).

Improvement from clinical to non-clinical range. Out of 18 treatment completers, 12 had depression scores in the moderate or higher range at baseline. Of the 12, 10 parents entered the non-clinical range for depression by Time 2 (83%), and 8 remained non-clinical by Time 3 (67% improvement). In the Waitlist group, none of the 10 participants who were in the clinical range at baseline moved to the non-clinical range at Time 2, and 3 became non-clinical by Time 3 (30%). At Time 2, there was a significant interaction between Group and moving to the non-clinical range (χ^2 (1)=15.28, $p=<.001$), which attenuated by Time 3 (χ^2 (1)=2.93, $p=.09$). By Time 3, two Waitlist participants originally in the non-clinical range moved to a clinical score on the depression subscale, as did one Treatment group participant.

Secondary outcomes

Parent mental health and positive functioning. Values for between-group analyses for all secondary outcomes can be found in Supplemental Table 2. There was no significant Time × Condition effect for stress at Time 2 or Time 3. At Time 2, while the Treatment group did not have a significant reduction in stress from baseline, there was a trend toward reduction ($b=-1.67$, $p=.07$), which was not observed in the Waitlist group ($b=0.22$, $p=.80$). At Time 3, significant improvements in stress from baseline were observed in the Treatment group ($b=-2.58$, $p=.01$), but not for the Waitlist group ($b=-0.99$, $p=.17$). At follow-up, maintenance of the improvements in stress between baseline and Time 3 was seen, with no significant changes from Time 3 to Time 4 for treatment completers ($b=0.67$, $p=.43$).

At Time 2, there was a significant Time × Condition interaction for positive affect, with a small effect size. The Treatment group showed an increase in positive affect ($b=2.76$, $p=.02$), whereas the Waitlist group had lower positive affect at Time 2 ($b=-2.95$, $p=<.001$). At Time 3, there was no treatment effect observed for positive affect.

At Time 3, there was no significant Time × Condition interaction for parenting stress-related health, but there was a trend toward more improvement in the Treatment group ($b=-0.89$, $p=.16$) than the Waitlist group ($b=0.84$, $p=.20$). For isolation, there was no Time × Condition effect, nor any within-group change for either group.

Goal attainment. At Time 2, 81.9% of the Treatment group had met at least one of their goals, compared to 43.0% of the Waitlist group, and there was a significant association between Group and goal attainment, $F(1, 461.41)=7.47$, $p=.007$, $d=0.80$. Based on the odds ratio, the odds of achieving or exceeding one's goal was 5.99 times higher if in the Treatment group than in the Waitlist group (95% confidence interval:1.73, 20.70). This association was not present at Time 3, $F(1, 394.16)=1.45$, $p=.23$, $d=0.33$ or at Time 4, $F(1, 637.51)=0.12$, $p=.73$, $d=0.09$. At Time 3, 75.2% of the Treatment group was achieving or exceeding their goal, compared to 58.1% for the Waitlist group.

ACT process measures. There was no Time × Condition effect for any ACT process measures. Looking at within-group change for each group, improvements were observed in the Treatment group for experiential avoidance ($b=-4.22$, $p=.01$) and cognitive fusion ($b=-3.78$, $p=.001$), whereas none were observed for the Waitlist group ($b=-0.86$, $p=.56$ and $b=-2.03$, $p=.11$ respectively). There was no significant within-group change for valued living or mindful parenting at Time 3 for either group. At 4-month follow-up, maintenance of the Treatment group's improvements between baseline and Time 3 was seen for experiential avoidance. While not significant, there was also a trend toward further improvement ($b=-2.76$; $p=.07$). There

Table 1. Group differences in parent and child characteristics at baseline.

Variable	Treatment $n = 27$	Waitlist $n = 27$	t or χ^2	p
Parents				
Age *M* (*SD*)	47.11 (7.66)	49.56 (9.80)	$t = -1.02$	.31
Range	36–66	32–69		
Sex *n* (%)			$\chi^2 = 1.20$	.27
Female	24 (88.9%)	21 (77.8%)		
Male	3 (11.1%)	6 (22.2%)		
Marital status *n* (%)			$\chi^2 = 0.91$	.34
Married/common law	19 (70.4%)	22 (81.5%)		
Single/divorced/separated	8 (29.6%)	5 (18.5%)		
Current sleep/psychotropic medication			$\chi^2 = 4.21$	.12
Yes	6 (22.2%)	12 (44.4%)		
No	20 (74.1%)	12 (44.4%)		
Missing	1 (3.7%)	3 (11.1%)		
Current individual/group/family therapy			$\chi^2 = 4.60$	.10
Yes	8 (29.6%)	4 (14.8%)		
No	10 (37.0%)	16 (59.3%)		
Missing	9 (33.3%)	7 (25.9%)		
Ethnicity *n* (%)			$\chi^2 = 8.36$	.40
Chinese	2 (7.4%)	2 (7.4%)		
East Asian	0	1 (3.7%)		
Filipino	0	1 (3.7%)		
South Asian	0	2 (7.4%)		
Black	1 (3.7%)	0		
White	20 (74.1%)	19 (70.4%)		
Latin American/Hispanic	1 (3.7%)	0		
Other/Prefer not to answer	3 (11.1%)	2 (7.4%)		
Education *n* (%)			$\chi^2 = 1.89$	.26
High school graduate	1 (3.7%)	1 (3.7%)		
Graduated college	6 (22.2%)	5 (18.5%)		
Some university	2 (7.4%)	3 (11.1%)		
Graduated university	10 (37.0%)	7 (25.9%)		
Post-graduate degree	8 (29.6%)	11 (40.7%)		
Household income (CAD) *n* (%)			$\chi^2 = 3.18$	.53
$<$\$50,000	2 (7.4%)	2 (7.4%)		
\$50,000–\$99,999	8 (29.6%)	6 (22.2%)		
\$100,000–\$149,999	5 (18.5%)	5 (18.5%)		
$\geqslant$\$150,000	10 (37.0%)	8 (29.6%)		
Prefer not to answer	2 (7.4%)	6 (22.2%)		
Children				
Age *M* (*SD*)	15.07 (7.92)	14.07 (7.77)	$t = 0.47$	.64
Range	3–34	3–27		
Sex *n* (%)			$\chi^2 = 1.03$	.31
Female	7 (25.9%)	4 (14.8%)		
Male	20 (74.1%)	23 (85.2%)		
Years since autism diagnosis *M* (*SD*)	7.26 (7.80)	8.35 (7.11)	$t = -0.47$	.64
Range	0–33	1–22		
Ethnicity *n* (%)			$\chi^2 = 5.02$	.66
Indigenous	0	1 (3.7%)		
Chinese	1 (3.7%)	1 (3.7%)		
South Asian	0	2 (7.4%)		
Black	1 (3.7%)	0		
White	22 (81.5%)	21 (77.8%)		
Other/prefer not to answer	3 (11.1%)	2 (7.4%)		

(Continued)

Table 1. (Continued)

Variable	Treatment $n = 27$	Waitlist $n = 27$	t or χ^2	p
SIB-R Standard Score *M* (*SD*)	75.63 (27.75)	63.41 (36.64)	$t = 1.38$	.17
Range	12–118	0–124		
SCQ Total Score *M* (*SD*)	18.56 (6.81)	19.78 (5.73)	$t = -0.71$	.48
Range	6–32	11–29		

SD: standard deviation; *SIB-R*: Scales of Independent Behavior–Revised; *SCQ*: Social Communication Questionnaire.

Table 2. Primary outcome for treatment and waitlist groups from baseline using linear mixed effects model.

Measure	EMM (*SE*)		Time × Condition Effect	p	d
	Treatment	Waitlist			
DASS-21 Depression					
Baseline	7.12 (0.95)	5.48 (0.57)			
Time 2	3.91 (0.57)	5.95 (0.87)	$F(1, 48.31) = 10.53$	.002	–0.87
Time 3	4.53 (0.97)	5.56 (0.82)	$F(1, 48.08) = 5.02$	.03	–0.64

SE: standard error; DASS-21: Depression Anxiety Stress Scale; EMM: estimated marginal mean.

was a change observed in cognitive fusion, indicating further improvements since Time 3 for the Treatment group ($b = -2.44$, $p = .04$).

Family functioning and youth mental health. Between baseline and Time 2, being in the Treatment group was associated with reductions in family distress ($b = -1.09$, $p = < .001$), not seen for the Waitlist group ($b = -0.31$, $p = .13$). The Time × Condition effect was significant with a small effect size. At Time 3, a medium Time × Condition effect was seen for family distress, with parents in the Treatment group ($b = -1.00$, $p = .001$) demonstrating greater improvement compared to the Waitlist group ($b = -0.10$, $p = .77$). At 4-month follow-up, maintenance of the improvements in family distress between baseline and Time 3 was seen, with no significant changes from Time 3 to Time 4 for treatment completers ($b = 0.05$, $p = .77$). There was no within-group change or Time × Condition effect for general family functioning or youth mental health.

Discussion

The current study used an RCT design to test the efficacy of a brief, group-based ACT intervention for mothers and fathers of autistic children, adolescents, and adults. Overall, the intervention resulted in gains for parents, with the greatest treatment effects observed for parent depression and family distress, which were maintained at the 4-month follow-up. Parents also reported short-term gains with respect to personal goal attainment, and small changes in positive affect. Although there was no significant Group × Time interaction for stress, cognitive fusion, and experiential avoidance, all these outcomes showed improvement, with small effect sizes, for the Treatment group at post-intervention, while the Waitlist group did not improve; these within-group changes were also maintained at follow-up. Parents did not report any significant improvements with respect to mindful parenting, valued living, overall family functioning, or child mental health.

Parent mental health and positive functioning

The most consistent treatment effect was seen for parent depression. These findings align with previous studies of ACT for parents of autistic people (Blackledge & Hayes, 2006; Joekar et al., 2016; Lunsky et al., 2018; Poddar et al., 2015), wherein parent self-reports of depression decreased post-intervention. In our study, 67% of Treatment condition participants with clinical levels of depression were in the normal range at Time 3, compared to only 30% of those in the Waitlist condition. While Blackledge and Hayes had some evidence for cognitive defusion as a mediator for depression improvements, and studies of ACT in other populations also show post-treatment defusion mediating changes in depression (Zettle et al., 2011), our sample showed less change in defusion than found in prior research. Fung and colleagues (2018) did report that post-ACT intervention improvements in mental health for mothers of autistic children were mediated by improvements in values-consistent action. Although our sample did not show change in their reports of how consistently they acted in accordance with their values, they did in action toward individually-set goals, which could underlie the changes in depression observed. Additional research with a larger cohort of participants would help to better elucidate the relationship between

valued living, goals, cognitive fusion, and depression in parents of autistic youth.

Conversely, no significant differences between groups for stress were reported. Parents of autistic children can experience chronic stressors, including high levels of mental health problems in their autistic youth (e.g. Simonoff et al., 2008). Indeed, in the current study, the average child mental health problems score was in the "very high" range, including specific conduct problems ranging from "raised" to "high," and the overall impact of these problems in the "very high" range. These family characteristics may require targeted interventions before pronounced differences are seen in parents' personal experience of stress.

Parents who participated in ACT had greater increases in positive affect compared to the Waitlist group at Time 2, though the effect was small and dissipated by Time 3. The link between positive affect and perceived social support in parents of children with disabilities has been repeatedly demonstrated (Findler et al., 2016; Smith et al., 2012), and it follows that positive affect would increase immediately after an intensive workshop designed to allow parents with similar experiences to connect. This may not have been maintained as time passed, without sustained social connections or ongoing engagement with the material.

Goal attainment, similarly, showed a large group difference at Time 2, which was no longer apparent at Time 3. At Time 2, the Treatment group was six times more likely to report achieving or exceeding one of their goals, with 82% of the Treatment parents and only 43% of the Waitlist parents indicating goal achievement. Recent research on goal attainment indicates that ongoing progress is most likely when goals are set just slightly out of reach, balancing challenge while preventing discouragement (Chevance et al., 2021). Further, ACT encourages parents to shift their behavior to align with their most closely held values. Since many Treatment group participants had already achieved or exceeded their highest-rated goal just after the workshop weekend, they might have benefited from re-setting their goals higher after reaching them, or based on values they identified and prioritized in the workshop. Last, as the highest-rated goal may have differed at different time points, we cannot say whether attainment of a particular goal was maintained for each participant at subsequent measurement times, only that many reported attaining at least one of their goals.

ACT psychological processes

In terms of the four ACT psychological processes, cognitive fusion and experiential avoidance showed small improvements in the Treatment group only, though not to a degree that resulted in a Group × Time interaction, and there was no change in mindful parenting or valued living. As shown in other studies of ACT interventions, some process-related changes take several months to become evident, particularly with respect to behavior change (González-Menéndez et al., 2014; Kohtala et al., 2017). Further, for mindfulness-based interventions, there is a strong relation between degree of mindfulness practice at home and amount of improvement in mindfulness and wellbeing (Carmody & Baer, 2009; Greenberg et al., 2018; Parsons et al., 2017). Published ACT manuals emphasize the importance of home practice (Hayes, 2005), and it is generally established that practice is beneficial to maximize effects of psychotherapy in general (Hoet et al., 2018). A weekly format with regular homework check-ins, or individual support, might support more practice and therefore greater change in behavior.

Family functioning and child mental health

More distal outcomes of family functioning and child mental health were also assessed. There were strong consistent treatment effects for family distress (BFDS). Although the intervention is not intended to attenuate family crises, participants in ACT are encouraged to change their relationship to their internal and external experiences. The BFDS asks parents to provide a broad appraisal of how difficult their current situation is and post-intervention, parents might have appraised their circumstances in a different light. It is also possible that reductions in distress occurred following experiences that were not sufficiently captured in our process measures. For instance, participants engaged in positive, supportive, and relaxing activities, possibly providing a sense of greater ability to cope with stressors or a temporary sense of relief, translating into shifts in their experience of distress. Further, through engaging with fellow group members in similar situations, and sharing experiences of distress, it is possible that participants received important informal social support that helped them to manage the stressors in their lives. Results do not support the hypothesis that a brief ACT workshop would result in changes in more general family functioning and child mental health. The factors affecting mental health problems are complex and multifaceted, with many unrelated to parenting behavior or parent mental health, and require more direct management to result in improvement (White et al., 2018).

Strengths and limitations

This study had many strengths. This was one of only two RCTs testing the efficacy of ACT for improving well-being in parents of autistic children, allowing us to elucidate the treatment effects of ACT while controlling for time and repeated measurements. In addition, the inclusion of a follow-up assessment ensures that longer-term program effects are captured. The heterogeneity of the sample, in

terms of including fathers, as well as parents of autistic children spanning a range of ages, socio-communication functioning, and adaptive skills, is also a strength, and improves generalizability of findings to more parents of autistic people.

There are also key limitations to our study. Participants were recruited mainly through autism service agencies, and many were already receiving supports and services for their child, which may limit generalization to families who do not have any support, or who are unwilling to potentially wait 4 months to receive the program. Twenty-two Treatment group participants completed the main intervention weekend, and four were unable to attend the refresher one month later, leaving only 18 full "treatment completers." In addition, all outcomes were assessed through subjective parent report, which could potentially influence the validity of results or limit some effects of the intervention being captured. Further, the high average level of household income and parent education, limited racial/ethnic diversity, and relatively low levels of parent mental health problems in this cohort limit the conclusions that can be drawn regarding the effect of ACT for parents with less education, with varying cultural backgrounds, or with greater need.

Conclusion

Given the higher levels of distress experienced by parents of autistic individuals compared to parents who do not have a child with a disability (Keenan et al., 2016), it is crucial to identify brief, effective and accessible supports. This study aligns with the growing body of research that suggests that ACT can benefit parents across multiple domains. Important next steps are to understand more about parents' experiences in the workshop to learn which aspects of the intervention are most useful. This might include a measure of social connection in the group. Future trials might also compare different modes of intervention delivery (i.e. weekly vs brief intensive format, in-person vs virtual delivery) as well as the effect of practice or longer-term support, on outcomes. In addition, studies may consider implementing assessments of children and families from a secondary reporter, such as a teacher or other parent, or youth self-report. Finally, it would be important to include a longer follow-up time point in additional RCTs of ACT.

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Supplemental material

Supplemental material for this article is available online.

Notes

1. We recognize that there are many viewpoints with respect to selecting language to describe autism and other disabilities. We use identity-first language to reflect the preference of many autistic adults and their family members (Botha et al., 2021; Kenny et al., 2016).
2. The term *children* will subsequently be used to refer to participating parents' autistic children, regardless of the age of the child, adolescent, or adult.

References

Abidin, R. R. (2012). *Parenting stress index* (4th ed.) (PSI-4). Psychological Assessment Resources.

Allik, H., Larsson, J. O., & Smedje, H. (2006). Health-related quality of life in parents of school-age children with Asperger syndrome or high-functioning autism. *Health and Quality of Life Outcomes*, *4*(1), Article 1.

Bitsika, V., & Sharpley, C. F. (2004). Stress, anxiety and depression among parents of children with autism spectrum disorder. *Australian Journal of Guidance and Counselling*, *14*(2), 151–161.

Blackledge, J. T., & Hayes, S. C. (2006). Using acceptance and commitment training in the support of parents of children diagnosed with autism. *Child & Family Behavior Therapy*, *28*(1), 1–18.

Bond, F. W., Hayes, S. C., Baer, R. A., Carpenter, K. M., Guenole, N., Orcutt, H. K., Waltz, T., & Zettle, R. D. (2011). Preliminary psychometric properties of the Acceptance and Action Questionnaire–II: A revised measure of psychological inflexibility and experiential avoidance. *Behavior Therapy*, *42*(4), 676–688.

Botha, M., Hanlon, J., & Williams, G. L. (2021). Does language matter? Identity-first versus person-first language use in autism research: A response to Vivanti. *Journal of Autism and Developmental Disorders*, *53*, 870–878. https://doi.org/10.1007/s10803-020-04858-w

Bovend'Eerdt, T. J., Botell, R. E., & Wade, D. T. (2009). Writing SMART rehabilitation goals and achieving goal attainment

scaling: A practical guide. *Clinical Rehabilitation*, *23*(4), 352–361.

Brown, F. L., Whittingham, K., Boyd, R. N., McKinlay, L., & Sofronoff, K. (2015). Does stepping stones triple P plus acceptance and commitment therapy improve parent, couple, and family adjustment following paediatric acquired brain injury? A randomised controlled trial. *Behaviour Research and Therapy*, *73*, 58–66.

Bruininks, R. H., Woodcock, R. W., Weatherman, R. F., & Hill, B. K. (1996). *Scales of independent behavior-revised*. Riverside.

Carmody, J., & Baer, R. A. (2009). How long does a mindfulness-based stress reduction program need to be? A review of class contact hours and effect sizes for psychological distress. *Journal of Clinical Psychology*, *65*(6), 627–638.

Chalfant, A. M., Rapee, R., & Carroll, L. (2007). Treating anxiety disorders in children with high functioning autism spectrum disorders: A controlled trial. *Journal of Autism and Developmental Disorders*, *37*(10), 1842–1857.

Chevance, G., Baretta, D., Golaszewski, N., Takemoto, M., Shrestha, S., Jain, S., Rivera, D. E., Klasnja, P., & Hekler, E. (2021). Goal setting and achievement for walking: A series of N-of-1 digital interventions. *Health Psychology*, *40*(1), 30–39.

Chiarello, L. A., Effgen, S. K., Jeffries, L. M., McCoy, S. W., & Tezanos, A. G. V. (2020). Relationship of school-based physical therapy services to student goal achievement. *Pediatric Physical Therapy*, *32*(1), 26–33.

Cohen, J. (1992). Statistical power analysis. *Current Directions in Psychological Science*, *1*(3), 98–101.

Crawford, J. R., & Henry, J. D. (2004). The Positive and Negative Affect Schedule (PANAS): Construct validity, measurement properties and normative data in a large non-clinical sample. *British Journal of Clinical Psychology*, *43*(3), 245–265.

Ekas, N. V., Lickenbrock, D. M., & Whitman, T. L. (2010). Optimism, social support, and well-being in mothers of children with autism spectrum disorder. *Journal of Autism and Developmental Disorders*, *40*(10), 1274–1284.

Enders, C. K. (2010). *Applied missing data analysis*. Guilford Press.

Epstein, N. B., Baldwin, L. M., & Bishop, D. S. (1983). The McMaster family assessment device. *Journal of Marital and Family Therapy*, *9*(2), 171–180.

Estes, A., Munson, J., Dawson, G., Koehler, E., Zhou, X. H., & Abbott, R. (2009). Parenting stress and psychological functioning among mothers of preschool children with autism and developmental delay. *Autism*, *13*(4), 375–387.

Feinberg, E., Augustyn, M., Fitzgerald, E., Sandler, J., Suarez, Z. F. C., Chen, N., Cabral, H., Beardslee, W., & Silverstein, M. (2014). Improving maternal mental health after a child's diagnosis of autism spectrum disorder: Results from a randomized clinical trial. *JAMA Pediatrics*, *168*(1), 40–46.

Findler, L., Jacoby, A. K., & Gabis, L. (2016). Subjective happiness among mothers of children with disabilities: The role of stress, attachment, guilt and social support. *Research in Developmental Disabilities*, *55*, 44–54.

Fung, K., Lake, J., Steel, L., Bryce, K., & Lunsky, Y. (2018a). ACT processes in group intervention for mothers of children with autism spectrum disorder. *Journal of Autism and Developmental Disorders*, *48*, 2740–2747.

Fung, K., Steel, L., Bryce, K., Lake, J., & Lunsky, Y. (2018b). *Using acceptance and commitment therapy for parents of children with autism* (A guide to programs for parenting children with autism spectrum disorder, intellectual disabilities or developmental disabilities: Evidence-based guidance for professionals). Kingsley.

Gillanders, D. T., Bolderston, H., Bond, F. W., Dempster, M., Flaxman, P. E., Campbell, L., Kerr, S., Tansey, L., Noel, P., Ferenbach, C., Masley, S., Roach, L., Lloyd, J., May, L., Clarke, S., & Remington, B. (2014). The development and initial validation of the cognitive fusion questionnaire. *Behavior Therapy*, *45*(1), 83–101.

González-Menéndez, A., Fernández, P., Rodríguez, F., & Villagráa, P. (2014). Long-term outcomes of acceptance and commitment therapy in drug-dependent female inmates: A randomized controlled trial. *International Journal of Clinical and Health Psychology*, *14*, 18–27.

Goodman, R. (2001). Psychometric properties of the strengths and difficulties questionnaire. *Journal of the American Academy of Child & Adolescent Psychiatry*, *40*(11), 1227–1345.

Gould, E. R., Tarbox, J., & Coyne, L. (2018). Evaluating the effects of acceptance and commitment training on the overt behavior of parents of children with autism. *Journal of Contextual Behavioral Science*, *7*, 81–88.

Greenberg, J., Braun, T. D., Schneider, M. L., Finkelstein-Fox, L., Conboy, L. A., Schifano, E. D., & Lazar, S. W. (2018). Is less more? A randomized comparison of home practice time in a mind-body program. *Behaviour Research and Therapy*, *111*, 52–56.

Hahs, A. D., Dixon, M. R., & Paliliunas, D. (2018). Randomized controlled trial of a brief acceptance and commitment training for parents of individuals diagnosed with autism spectrum disorders. *Journal of Contextual Behavioral Science*, *12*, 154–159.

Han, A., Yuen, H. K., & Jenkins, J. (2021). Acceptance and commitment therapy for family caregivers: A systematic review and meta-analysis. *Journal of Health Psychology*, *26*(1), 82–102.

Harris, R. (2006). Embracing your demons: An overview of acceptance and commitment therapy. *Psychotherapy in Australia*, *12*(4), 70–76.

Hastings, R. P. (2016). Do children with intellectual and developmental disabilities have a negative impact on other family members? The case for rejecting a negative narrative. *International Review of Research in Developmental Disabilities*, *50*, 165–194.

Hastings, R. P., Kovshoff, H., Brown, T., Ward, N. J., Espinosa, F. D., & Remington, B. (2005). Coping strategies in mothers and fathers of preschool and school-age children with autism. *Autism*, *9*(4), 377–391.

Hayes, S. A., & Watson, S. L. (2013). The impact of parenting stress: A meta-analysis of studies comparing the experience of parenting stress in parents of children with and without autism spectrum disorder. *Journal of Autism and Developmental Disorders*, *43*(3), 629–642.

Hayes, S. C. (2005). *Get out of your mind and into your life: The new acceptance and commitment therapy*. New Harbinger.

Hayes, S. C., Luoma, J. B., Bond, F. W., Masuda, A., & Lillis, J. (2006). Acceptance and commitment therapy: Model,

processes and outcomes. *Behaviour Research and Therapy*, *44*(1), 1–25.

Hoet, A. C., Burgin, C. J., Eddington, K. M., & Silvia, P. J. (2018). Reports of therapy skill use and their efficacy in daily life in the short-term treatment of depression. *Cognitive Therapy and Research*, *42*(2), 184–192.

Jennings, L. A., Ramirez, K. D., Hays, R. D., Wenger, N. S., & Reuben, D. B. (2018). Personalized goal attainment in dementia care: Measuring what persons with dementia and their caregivers want. *Journal of the American Geriatrics Society*, *66*(11), 2120–2127.

Joekar, S., Farid, A. A., Birashk, B., Gharraee, B., & Mohammadian, M. (2016). Effectiveness of acceptance and commitment therapy in the support of parents of children with high-functioning Autism. *International Journal of Humanities and Cultural Studies*, *2*(4), 2763–2772.

Jones, L., Hastings, R. P., Totsika, V., Keane, L., & Rhule, N. (2014). Child behavior problems and parental well-being in families of children with autism: The mediating role of mindfulness and acceptance. *American Journal on Intellectual and Developmental Disabilities*, *119*(2), 171–185.

Kayfitz, A. D., Gragg, M. N., & Robert Orr, R. (2010). Positive experiences of mothers and fathers of children with autism. *Journal of Applied Research in Intellectual Disabilities*, *23*(4), 337–343.

Keenan, B. M., Newman, L. K., Gray, K. M., & Rinehart, N. J. (2016). Parents of children with ASD experience more psychological distress, parenting stress, and attachment-related anxiety. *Journal of Autism and Developmental Disorders*, *46*(9), 2979–2991.

Kenny, L., Hattersley, C., Molins, B., Buckley, C., Povey, C., & Pellicano, E. (2016). Which terms should be used to describe autism? Perspectives from the UK autism community. *Autism*, *20*(4), 442–462.

Kohtala, A., Muotka, J., & Lappalainen, R. (2017). What happens after five years? The long-term effects of a four-session acceptance and commitment therapy delivered by student therapists for depressive symptoms. *Journal of Contextual Behavioral Science*, *6*, 230–238.

Lovibond, S. H., & Lovibond, P. F. (1995). *Manual for the Depression Anxiety Stress Scales*. Psychology Foundation of Australia.

Lunsky, Y., Fung, K., Lake, J., Steel, L., & Bryce, K. (2018). Evaluation of Acceptance and Commitment Therapy (ACT) for mothers of children and youth with autism spectrum disorder. *Mindfulness*, *9*(4), 1110–1116.

Lunsky, Y., Robinson, S., Reid, M., & Palucka, A. (2015). Development of a mindfulness-based coping with stress group for parents of adolescents and adults with developmental disabilities. *Mindfulness*, *6*(6), 1335–1344.

McGarrigle, L., & Rockwood, K. (2020). The responsiveness of goal attainment scaling using just one goal in controlled clinical trials: An exploratory analysis. *Journal of Patient-reported Outcomes*, *4*, 1–10.

McKenzie, K., Ouellette, -., Kuntz, H., Blinkhorn, A., & Démoré, A. (2017). Out of school and into distress: Families of young adults with intellectual and developmental disabilities in transition. *Journal of Applied Research in Intellectual Disabilities*, *30*(4), 774–781.

Meirsschaut, M., Roeyers, H., & Warreyn, P. (2010). Parenting in families with a child with autism spectrum disorder and a typically developing child: Mothers' experiences and cognitions. *Research in Autism Spectrum Disorders*, *4*(4), 661–669.

Neece, C. L. (2014). Mindfulness-based stress reduction for parents of young children with developmental delays: Implications for parental mental health and child behavior problems. *Journal of Applied Research in Intellectual Disabilities*, *27*(2), 174–186.

Parsons, C. E., Crane, C., Parsons, L. J., Fjorback, L. O., & Kuyken, W. (2017). Home practice in mindfulness-based cognitive therapy and mindfulness-based stress reduction: A systematic review and meta-analysis of participants' mindfulness practice and its association with outcomes. *Behaviour Research and Therapy*, *95*, 29–41.

Poddar, S., Sinha, V. K., & Urbi, M. (2015). Acceptance and commitment therapy on parents of children and adolescents with autism spectrum disorders. *International Journal of Educational and Psychological Researches*, *1*(3), 221–225.

Reid, C., Gill, F., Gore, N., & Brady, S. (2016). New ways of seeing and being: Evaluating an acceptance and mindfulness group for parents of young people with intellectual disabilities who display challenging behaviour. *Journal of Intellectual Disabilities*, *20*(1), 5–17.

Rutter, M., Bailey, A., & Lord, C. (2003). *The social communication questionnaire*. Western Psychological Services.

Simonoff, E., Pickles, A., Charman, T., Chandler, S., Loucas, T., & Baird, G. (2008). Psychiatric disorders in children with autism spectrum disorders: Prevalence, comorbidity, and associated factors in a population-derived sample. *Journal of the American Academy of Child and Adolescent Psychiatry*, *47*, 921–929.

Singh, N. N., Lancioni, G. E., Winton, A. S., Fisher, B. C., Wahler, R. G., Mcaleavey, K., Singh, J., & Sabaawi, M. (2006). Mindful parenting decreases aggression, noncompliance, and self-injury in children with autism. *Journal of Emotional and Behavioral Disorders*, *14*(3), 169–177.

Singh, N. N., Lancioni, G. E., Winton, A. S., Karazsia, B. T., Myers, R. E., Latham, L. L., & Singh, J. (2014). Mindfulness-based positive behavior support (MBPBS) for mothers of adolescents with autism spectrum disorder: Effects on adolescents' behavior and parental stress. *Mindfulness*, *5*(6), 646–657.

Sivberg, B. (2002). Family system and coping behaviors: A comparison between parents of children with autistic spectrum disorders and parents with non-autistic children. *Autism*, *6*(4), 397–409.

Smith, L. E., Greenberg, J. S., & Seltzer, M. M. (2012). Social support and well-being at mid-life among mothers of adolescents and adults with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, *42*(9), 1818–1826.

Thullen, M., & Bonsall, A. (2017). Co-parenting quality, parenting stress, and feeding challenges in families with a child diagnosed with autism spectrum disorder. *Journal of Autism and Developmental Disorders*, *47*(3), 878–886.

Tonge, B., Brereton, A., Kiomall, M., Mackinnon, A., King, N., & Rinehart, N. (2006). Effects on parental mental health of an education and skills training program for parents of

young children with autism: A randomized controlled trial. *Journal of the American Academy of Child & Adolescent Psychiatry*, *45*(5), 561–569.

Urbaniak, G. C., & Plous, S. (2015). Research randomizer (Version 4.0) [Computer software]. http://www.randomizer.org/

Weiss, J. A., Cappadocia, M. C., MacMullin, J. A., Viecili, M., & Lunsky, Y. (2012). The impact of child problem behaviors of children with ASD on parent mental health: The mediating role of acceptance and empowerment. *Autism*, *16*(3), 261–274.

Weiss, J. A., & Lunsky, Y. (2011). The Brief Family Distress Scale: A measure of crisis in caregivers of individuals with autism spectrum disorders. *Journal of Child and Family Studies*, *20*(4), 521–528.

White, S. W., Simmons, G. L., Gotham, K. O., Conner, C. M., Smith, I. C., Beck, K. B., & Mazefsky, C. A. (2018). Psychosocial treatments targeting anxiety and depression in adolescents and adults on the autism spectrum: Review of the latest research and recommended future directions. *Current Psychiatry Reports*, *20*(10), 1–10.

Whittingham, K., Sanders, M. R., McKinlay, L., & Boyd, R. N. (2015). Parenting intervention combined with Acceptance and Commitment Therapy: A trial with families of children with cerebral palsy. *Journal of Pediatric Psychology*, *41*(5), 531–542.

Wilson, K. G., Sandoz, E. K., Kitchens, J., & Roberts, M. (2010). The Valued Living Questionnaire: Defining and measuring valued action within a behavioral framework. *The Psychological Record*, *60*(2), 249–272.

Zettle, R. D., Rains, J. C., & Hayes, S. C. (2011). Processes of change in acceptance and commitment therapy and cognitive therapy for depression: A mediation reanalysis of Zettle and Rains. *Behavior Modification*, *35*(3), 265–283.

4. L’étude qualitative (qualitative research)

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Happiness in Parents of Children with Autism Spectrum Disorder: A Qualitative Study

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ABSTRACT
Literature reports the challenges of parenting a child with Autism Spectrum Disorder (ASD), and its impacts on emotional and psychosocial wellbeing, both generally and specifically in the Hong Kong context. Positive parenting experiences are less well defined; however, research and theory in the positive psychology field suggests that people living with adversity can find and create meaning, positivity and happiness. This study aimed to investigate Hong Kong parents' experiences and perceptions of happiness and well-being in raising their children with ASD. Eight parents (2 fathers; 6 mothers) were interviewed in-depth about their parenting-related perceptions and experiences of happiness and well-being. A thematic analysis generated four themes: A Growth Mindset, Connectedness, Self-Care, and A Better Me, each comprising several related sub-themes. Findings indicated that happiness was crucial for these parents, despite the challenges of raising a child with ASD in Hong Kong, and that they actively sought out activities to enhance their happiness, satisfaction and meaning. Results are in keeping with positive psychology theory, add specific detail relating to this group and could be extended with further research. Findings could also inform policy makers and support services in the development of welfare plans, educational resources and social support for this group of vulnerable families in future.

KEYWORDS: Autism spectrum disorder, happiness, parenting, qualitative, thematic analysis.

Autism Spectrum Disorder (ASD) is a neurodevelopmental disorder defined by deficits in socio-emotional communication and restricted or repetitive behaviors, which continue throughout the lifespan (American Psychiatric Association, 2013). ASD is estimated to affect 1 in 44 children globally (Center for Disease Control and Prevention [CDC], 2021). ASD children may experience learning difficulties, such as speech delay and language impairments (Sun et al., 2019). There may also be difficulties with social reciprocity (A. Huang et al., 2017) and adaptive functioning (Towle et al., 2014). Around half of the ASD population also meets the diagnostic criteria for Attention-Deficit Hyperactivity Disorder (ADHD; Dellapiazza, 2021; Murray, 2010). Dellapiazza (2021) noted that the concurrence and severity of ASD and ADHD in children were positively correlated with social impairments, externalizing problems (e.g., aggressive and oppositional behaviors) and internalizing behaviors (e.g., anxiety and mood

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disorders). Complex ASD symptoms seriously affect children's developmental performance, social communication, and behavior.

Parenting can bring emotional rewards (Schoen et al., 1997), life satisfaction, and a sense of meaning (Nelson et al., 2014). However, parenting a child with ASD is more complex and challenging, and recent research from a variety of geographical and cultural contexts suggests these parents often experience significant levels of stress, anxiety, and depression when compared with parents of neurotypical children (e.g., Al-Oran et al., 2021; C. Huang et al., 2013; Lai et al., 2015; Tsermentseli & Kouklari, 2021). Papadopoulos (2021) conducted a Greece-based qualitative study of mothers' experiences of raising their ASD children and reported high levels of emotional, family, and social burden. Mothers' experiences were characterized by feelings of vulnerability, frustration about the child's future, the fear of stigmatization which reduced their confidence in engaging with the social world, and a sense of isolation from their wider family due to the caregiving responsibilities for their ASD child(ren). Parents suffer from self-blame, low satisfaction in the parent-child relationship, difficulty in managing their ASD child's behaviors (Lai et al., 2015), and worry about peer bullying (Cappadocia et al., 2011; Forrest et al., 2019). Aktan et al. (2020) found that burnout was negatively related to life satisfaction and quality of life in parents of children with disabilities.

Focusing on the Hong Kong context, the number of students diagnosed with ASD has increased rapidly in the past 10 years (Education Bureau of the Government of the Hong Kong Special Administrative Region, 2019). More than 10,000 ASD students are studying in local mainstream schools, with numbers increasing every year. A survey conducted by the Education University of Hong Kong (The Education University of Hong Kong [EdUHK], 2016) revealed the condition of parents of ASD children in Hong Kong. The survey focused on investigating the discrimination situation and well-being of parents of ASD children. The findings reported that 90% of participants claimed to encounter discrimination and that 60% were suffering from depressive symptoms. A concerning 21.3% of participants reported having thoughts of suicide or self-harm (EdUHK, 2016). The social stigma of ASD in Hong Kong proved very stressful to these parents and may be linked to traditional Chinese culture and values.

Chinese society is considered a collectivist culture that advocates conformity and activities that benefit the group over individual interests; it highly values group cooperation, social support, self-discipline, and respect to others (Crisp, 2020; Trafimow et al., 1991). The cultural norm for parenting is raising a child who can achieve great success in every aspect of their life (Tait et al., 2015). An old Chinese idiom, 'Nurturing children to become dragons' (望子成龍) summarizes Chinese parents' expectations of parenting and of their children. Many parents set high expectations for their children and try to mold them into dragons, the symbol of mastery or the elite (Ng et al., 2014). A child with ASD does not conform to these hopes and expectations, resulting in parental self-stigma and self-blame (Mak & Kwok, 2010; Tait et al., 2015). Ng et al. (2014) noted that Hong Kong Chinese parents (especially mothers) tended to link their own worth and value to their children's performance more strongly than parents of other cultural backgrounds. Children's academic and behavioral performances became parents' report cards, reflecting the success—or otherwise—of the parents (Chao, 1994). The social stigma was particularly strong in more traditional Chinese families in Hong Kong, with 'abnormal' children regarded as a family scandal (Tait et al., 2015), resulting in embarrassment, shame, fear of disclosing the ASD diagnosis, and difficulties in accepting their children. High levels of avoidance-style coping and parental anxiety have been found in this population (Voliovitch, 2021). Parents of ASD children in Hong Kong also experience a lack of professional support from the existing health system (Yi et al., 2020). Yi et al. (2020) described delays in screening and diagnosis, causing frustration and confusion and a lack of professional mental health support for parents. Parents reported feeling hopeless, abandoned, and distressed at the diagnostic and post-diagnostic stages and facing enormous internal and external pressures

which greatly impacted their mental health and also affected their children's intervention and improvement (Reed & Osborne, 2012).

Existing research, both globally and within Hong Kong, clearly articulates the complex challenges and relational burdens associated with parenting a child with ASD. There is a relative lack of research capturing a more balanced, nuanced picture of the experience. However, a few isolated studies have considered the positive, happiness-related experiences of this group. For example, Rafferty et al. (2020) interviewed fathers with ASD children, describing experiences of love, pride, hope, joy, empathy, gratitude, and personal growth. These accounts are reminiscent of the positive focus and findings within the positive psychology field, which expands our understanding of individuals' encounters with adversity beyond distress into meaning, sense-making, positivity, and happiness (Ivtzan et al., 2015).

According to some, finding happiness is one of the most important goals for human beings (Frey & Stutzer, 2010; Veenhoven & Ehrhardt, 1995), and people's choices, priorities, and behaviors can be determined by the goal of happiness (Huta, 2016). Happiness has been a topic of discussion within the positive psychology field for several decades (Boniwell et al., 2019; Seligman, 2004), and measures of happiness have been developed (Boniwell et al., 2019; Boyle et al., 2014); however, as a concept, happiness is not easy to define. Huta (2016) summarized happiness as well-being, including both hedonic and eudemonic experiences. Hedonic experiences involve enjoyment, pleasure, and comfort, and focus on meeting personal desires and a subjective experience of well-being in the moment. Eudaimonia is a broader, abstract, and more psychological concept involving, for example, meaning, purpose, value, personal growth, ethics, and autonomy. Pioneer of positive psychology, Seligman (2004) advocated authentic happiness which referred to a pleasurable life, an engaged life, and a meaningful life. In 2011, Seligman described the 'flourish' concept, the notion that people could act deliberately to achieve a flourishing life characterized by five elements: (1) positive emotions, (2) engagement, (3) relationships, (4) meanings, and (5) accomplishment. Seligman (2011) suggested that people could achieve, review, and evaluate happiness using his PERMA model.

Positive psychology has been criticized from existential and humanist perspectives (Yakushko & Blodgett, 2021), with a common criticism being its oversimplification of the dimension of emotion. It is argued that, in the real world, emotion cannot be polarized into positive or negative but is often mixed (Boniwell et al., 2019). Held (2002, 2004) rejects the simplification and polarization which views positivity as good and negativity as bad, arguing that it is impractical to ask people to reject negativity in life and work to foster positive thoughts and attitudes in pursuit of happiness, health, and wisdom. Furthermore, Held (2002) felt this could develop into a tyranny of positive thinking which blamed someone who failed to display optimism and virtue. A deeper discussion about the "dark side" of life emerged in the second wave of positive psychology (Ivtzan et al., 2015) which suggested that negative can be positive. This perspective suggests that embracing the negativity of life and accepting distress permits learning, the development of agile mindsets, skills for success and life fulfillment. Ivtzan et al. (2015) recognized that people who confront adversity in life underwent positive changes and transformational growth, similar to Calhoun and Tedeschi's (2004) concept of Post-Traumatic Growth (PTG).

Research suggests that having a child diagnosed with ASD is often interpreted by parents as a life crisis or trauma (Carmassi, 2021; Casey et al., 2012). Waizbard-Bartov et al. (2018) investigated crisis-related growth—developed from Tedeschi and Calhoun's (2004) concept of PTG—in parents with ASD children. They concluded that parents had experienced growth in areas that included mastery, personal strength, new perspectives on the value of life, ethics and spirituality, interaction with others, and using their newly acquired parenting knowledge in professional and political contexts. Other studies have reported similar findings (Ooi et al., 2016; Zhang et al., 2015). Two Chinese research teams (Liu & To, 2020; Zhang et

al., 2015) found that PTG was evident in the population of Chinese parents with ASD children with positive impacts on their ability to cope with stress. Waizbard-Bartov et al. (2018) investigated personal growth in parents with ASD children and concluded that parents had crisis-related growth in the forms of enhanced mastery of personal strength; constructing new perspectives on the value of life; ethics; interpersonal interaction and professional or political application of parenting knowledge. However, these studies found that the PTG Spiritual Change dimension, which focuses on spiritual beliefs and living with gratitude, was not evident among Chinese parents. Waizbard-Bartovet al. (2018) suggested that cultural influences, such as parents' perception that having an 'abnormal child' was shameful, might act to restrict personal growth in this area.

Existing evidence about parents of ASD children in Hong Kong paints a bleak picture. There is scope to develop a richer, more balanced, and nuanced representation of these parents' experiences by focusing in greater detail on parents' happiness and well-being experiences. These insights will help raise awareness and enrich understanding of this group's lived experience. They may also help in addressing the stigma of ASD and in informing the development by researchers and policymakers of supportive or educational interventions for parents and families. Much of the evidence that exists is quantitative, applying measures of happiness that may influence, prime, or restrict participant responses (Husser & Fernandez, 2017). Therefore, the current study aimed to explore qualitatively and in-depth the happiness and well-being experiences of parents of ASD children in Hong Kong.

Method

Study Design

The research employed a qualitative research design guided by critical realist ontological and epistemological assumptions (Bhaskar, 1978), which recognize that reality has stable and enduring features that exist independently of human perception but that individuals have different experiences of the real. Individual accounts reflect those experiential differences (Fiske & Taylor, 1991) and are appropriately accessed via semi-structured, in-depth qualitative interviews.

Participants

Parents were considered eligible if they had children aged 10-20 with an ASD diagnosis, were of Chinese ethnicity, had raised their children and lived in Hong Kong for at least five consecutive years, and could communicate in Cantonese. Participants who met the study criteria were recruited by an ASD children's trainer who worked in a local private training provider in Hong Kong. The trainer introduced the study, and interested parents were sent a detailed information sheet, and provided contact details for the researcher, who explained the information and answered questions over the phone. Signed consent forms were required.

Table 1 provides participant details. Eight parents participated, 6 mothers and 2 fathers. Age ranged from 36 to 58, with a mean of 47 years. All parents were of Chinese ethnicity and spoke Cantonese as their first language, and all were raised and educated in Hong Kong. All participants were married but one was in the process of divorce. All had high school or above level of education. Most were described themselves as middle socio-economic status.

The average age of the participants' children (all male) was 15.5 years; one participant had twin boys with ASD. All children were diagnosed with ASD by private psychiatrists or psychiatrists from the Hospital Authority (public health system) in Hong Kong. Children in this research had mild to moderate ASD symptoms; seven out of nine boys with a speech delay at

an early age. Four children had an additional diagnosis (e.g., Attention Deficit and Hyperactive Disorder, Global Developmental Delay and Oppositional Defiant Disorder).

Table 1
Participant Demographics

No	Pseudonym	Gender	Age	Highest education level	No. of ASD children	Age of ASD children	Children's ASD severity
P1	Ann	Female	50s	Secondary	1	18	Mild
P2	Belle	Female	40s	Tertiary	2, twins	12	Mild
P3	Chris	Male	50s	Secondary	1	20	Moderate
P4	Diana	Female	50s	Secondary	1	18	Moderate
P5	Ethen	Male	40s	Tertiary	1	11	Mild
P6	Faye	Female	40s	Secondary	1	11	Moderate
P7	Grace	Female	30s	Secondary	1	12	Mild
P8	Heather	Female	40s	Tertiary	1	10	Mild

Data Collection

Each parent participated in semi-structured interviews via Zoom (an online meeting platform) between May and July 2021, which were audio-recorded for transcription with consent. The interviewer (first author) was a psychology graduate student fluent in both Cantonese and English. Interviews lasted between 35 and 70 minutes.

Clarke and Braun (2013) defined an interview as a professional conversation aimed at gathering participants' experiences and perspectives. The interview schedule was designed to help parents consider and describe their personal, individual definition and experiences of happiness and its meaning and function in parenting their ASD child(ren).

In response to Husser et al. (2017)'s observation that questions about happiness can be priming and limiting, we ensured our interview questions were open-ended. The interview guide was pre-prepared, but it was expected that unanticipated issues might arise. Spontaneous and unplanned questions related to the topic were asked, as determined by participants' responses and to follow the train of their thoughts (Clarke & Braun, 2013).

Interview questions were first piloted with three parents with neurotypical children. Changes were made to the wording of some questions based on feedback, and additional retrospective questions were added, for example, how parents felt about hearing about their child's diagnosis and changes in thoughts, feelings, and well-being along their parental journey. In the resulting final schedule, interview questions were designed first to address the participant's general background, and then their understanding of the notion of happiness. Subsequent questions were based on relevant theoretical and empirical literature about emotions, relationships, parental roles, and journeys, and how they derived happiness through their experiences.

Member Checking

Interviews were conducted in Cantonese, the participants' first language. Audio recordings were transcribed verbatim, pseudonymized, and translated into English by the first author. The second author proofread the English transcripts and corrected grammatical errors. Transcripts in Cantonese were sent to participants, who were asked to check and approve the contents. They were offered the opportunity to add or make amendments. None made amendments, but one participant provided further detailed comments.

Ethical Considerations

Ethical approval was granted by the University Ethics Committee (AM/RKT/PSP-PF-2/2014-15). Processes of approach, recruitment, informed consent, and data protection complied fully with the ethical principles of respect, competence, responsibility, and integrity, as set out by The British Psychological Society (BPS, 2021).

Data Analysis

Transcripts were analyzed using reflexive thematic analysis (Braun & Clarke, 2021; Clarke & Braun, 2013), an inductive process of identifying themes and patterns of meaning across a dataset from the bottom up. The data analysis process followed Braun and Clarke's (2021) and Clarke and Braun's (2013) six steps, including familiarization with the data, coding, searching for themes, reviewing themes, defining, and naming themes, and writing up. The first author read transcripts several times to familiarize and immerse herself in the data. Complete coding involved a review of the entire dataset to identify data relevant to the research aims, during which the researcher moved through and beyond description and summary to conceptual analysis, which is crucial for reflexive thematic analysis. In this process, both explicit and implicit meanings were considered, which involves the researcher in conceptualizing and actively interpreting the meaning of participants' responses. Searching for themes was a process of searching for patterns, identifying similarities and overlaps between codes, creating groupings, developing themes, and considering any ungrouped codes. Reviewing themes was a careful process of refining themes, which meant reviewing, checking, merging, and splitting initial groupings to make better sense of the data and organizing concepts. The aim was to capture all relevant data into the most meaningful and coherent thematic groupings. Defining and naming themes involved creating definitions that clearly characterized and demonstrated the distinctiveness of each theme, naming themes in order to best represent the story of that theme, with sub-themes to reflect nuances. Writing up involved representing the participants' experiences and setting out the themes, sub-themes, data extracts and analytic process in a logical and accessible way for the reader.

Reflexive thematic analysis involves an interpretive process guided by the researchers' stance, disciplinary knowledge, epistemology, and lens (Clarke & Braun, 2013). It acknowledges the active engagement between the researcher and their data in order to develop themes conceptualized from codes. The first author is a Hong Kong University academic in the field of nursing; she conducted the first three steps of the analysis process independently, then considered the resulting initial themes with the second author, following which steps four and five included a collaborative process of discussion and refinement. The second author is a UK University academic, chartered psychologist, and qualitative researcher in the fields of social, health, and counseling psychology; she is also an integrative psychotherapist with a practice underpinned by relational, existential-humanistic principles.

Results

Four themes were generated: a growth mindset, connectedness, self-care, and a better me. These are presented with sub-themes in Table 2.

Table 2
Themes and Their Associated Sub-Themes Derived from Parents' Happiness Experiences

Themes	Sub-themes
1. **Growth Mindset**	- Redefining the Meaning of Happiness - Using Knowledge to Combat Anxiety - Never Compare with Others
2. **Connectedness**	- Life Satisfaction - Stress Coping Strategies - Fueling Up with Recognition
3. **Self-Care**	- Relaxation - 'Me' Time - Self Compassion
4. **A Better Me**	- Embracing Suffering - Making Sacrifices - Gratitude - Living with Purpose

Theme 1: A Growth Mindset

The Growth Mindset theme represented data from all eight parents and reflects the belief that personal development was possible even in the most difficult times. Most of these parents went through a difficult period when they received their children's ASD diagnosis. They spoke about the overwhelming impact of this news, with some characterizing it as the biggest crisis of their lives. Many described responses of shock, denial, depression, and anxiety based on a lack of knowledge about the disorder and anxiety about their children's future. For most participants, the hardest moment in parenting was during and immediately after the diagnostic period.

> *I wanted the world to stop; time to stop. I did wish they would stop growing up. If they grew up, there would be even bigger difficulties we'd have to face in life.* (Belle, speaking of her 12-year-old twin sons)

Beyond the initial shock and distress, parents went on to respond positively and with resilience to the challenges posed by this significant event in their lives. This response seemed to reflect a growth mindset, based on Dweck (2006)'s concept of quality. The growth mindset allowed individuals to flourish and achieve despite challenges through personal resilience, determination, and a willingness to learn and grow. Three sub-themes reflected the responses which characterized these parents' growth mindset: (1) Redefining the meaning of happiness, (2) being knowledgeable to combat anxiety, and (3) never comparing with others.

Re-defining the Meaning of Happiness

This was a commonly recurring theme in participants' accounts. Re-defining happiness involved changing the source of happiness from 'self' to 'self and children.' The change was evident when parents were asked about their happiness experiences before and after becoming a parent. Before becoming a parent, parents experienced happiness through personal successes in the form of career achievements and fulfillment of personal desires, for example, through leisure activities. With parenthood, participants found happiness through their children. The assertion, "The child's happiness is my happiness" was a frequent response of parents to the question, "What is the definition of happiness to you now?" When they saw their children living

happily, becoming more independent in handling their schoolwork and daily life, these were authentic happiness experiences for parents.

> *Before I became a dad (apparently forced smile), happiness, haha, er, er, happiness at that time would be related to my career, job satisfaction, er, er, achieving something. But now, after becoming a dad, I spend more time with my family, especially with my son - and my wife. Especially with my son, I will think about how to be with him and help him. Mainly, my happiness is related to my son's improvement, such as what I have given to him, and his improvement is my reward.* (Chris)

Parents expressed the sense that they were now clearer about what was important to them in life. Parenthood, and the challenges they had faced as parents of neurodiverse children with ASD, had offered them a deeper understanding of the purpose and meanings of life. After parents had accepted their children's condition and faced some of the challenges, as observed by Heather, they found new perspectives on life goals and happiness. Heather mentioned: "If someone has never faced any difficulties in their life - so if you interviewed me 20 years ago, I may not have been able to give you a real answer about happiness."

Redefining the meaning of happiness was the building block which enabled parents to see their children differently and enjoy their company more. As described by two other mothers, authentic happiness came from witnessing their children grow. The meaning of happiness changed when their identities changed. This change was accompanied by an increase in enjoyment and a decrease in negative emotions, characteristic of parents' growth mindsets.

> *Before being a mom, my happiness came from being with my friends. Being with them gave me so much pleasure and happy moments. We spent time together, eating, drinking, playing and having fun. We sometimes got drunk. (Laugh) [...] After being a mom, the happiest moment is seeing my son grow up, no matter if he gets sick or learns new things at school. I get satisfaction from seeing him learning.* (Grace)

> *Just like other Hong Kong girls, I loved to go on dates with my friends. We played together, shopped, and did some traveling. After becoming a mom, I changed completely. Witnessing the efforts my son and I make is what makes me happy. This is really wonderful and precious.* (Diana)

Using Knowledge to Combat Anxiety

Parents sometimes experienced a sense of hopelessness about their children's future. Parents were anxious that their children's behavioral, emotional and academic performance would always be inferior to others and were unsure how to help. However, parents also confronted the situation; they accepted their children's condition and started to think about taking action. By doing this, parents gained some control over the situation and strengthened their abilities to work with it. Actions included gaining knowledge about ASD traits and learning how to train and communicate with their children using special techniques. This process involved searching and finding varied resources, such as online or face-to-face courses provided by local health systems and private ASD children's training centers.

> *As we were unfamiliar with this disease, I looked for relevant resources and information to learn more. On the contrary, at that time, I had feelings of sadness but 'the sadness' motivated me to find more information. [...] I also geared myself up by taking on a lot of courses, especially about psychology and counseling.* (Belle)

With their new-found knowledge, parents' anxiety and worries gradually shrank over time. Parents mentioned that they began to take a more balanced approach to think about and evaluate their family's situation and future needs. With this mindset change, parents strove for a better future for their children.

> *When I gained more knowledge, the catastrophic feelings I'd had about my son disappeared little by little over time. I no longer catastrophized the situation. Maybe by taking this action, I found that my son was not 'incurable'. I used to think that his problems would last for the rest of his life but in reality, it is not like that. After fixing the catastrophizing issue, I could find the way out.* (Heather)

Never Comparing with Others

All parents strongly stated that they had given up comparing their ASD children with neurotypical peers. They perceived that comparing their children with others was pointless and, instead, believed that parents should be aware of their children's limitations while taking satisfaction from small gains and signs of progress. They appreciated examples of their children's competence and mastery rather than focusing on academic and performance achievements. Many parents reported being surprised that their children's performance exceeded their expectations, which brought them great joy and satisfaction. Chris explained, "When my son reaches standards that I didn't expect, I feel very happy. For example, my son's learning journey, we rarely need to push him to study. He is a self-disciplined person."

Furthermore, parents valued their children's positive character traits, such as being studious and willing to listen to parents' advice and showing self-discipline, kindness, and honesty. All parents believed that they experienced happiness no less than parents of neurotypical children and that every child has strengths and weaknesses, whether neurotypical or neurodiverse. Parents considered that it was their responsibility to discover their children's strong points and avoid blaming them for any limitations, as explained by Chris and Grace.

> *As parents, we should always see and help children to use their strengths but not patronize them. [...] Parents with normally developed children may place higher expectations on their children. [...] But for us, we know our kid's limitations, he may not live as a normal person, and we need to adjust our expectations.* (Chris)

> *First, you have to accept that your children have deficits. Second, you need to adjust your mindset. If you do not do that, how can you ask others to accept your child? You need to learn how to help and support your children. You need to understand your child's condition well so as to teach your child to understand themselves too. [...] It is important that you and your child accept the ASD condition. This is the key to being happy.* (Grace)

Theme 2: Connectedness

Connectedness reflected parents' thoughts about engaging with the social world and the functions it served for them. Data suggested that they tended to categorize their social contacts and networks into three tiers: intimate, external, and professional. Each of these played a different role in maintaining happiness. Three sub-themes reflected these functions: (1) life satisfaction, which reflected the function of the intimate and close family; (2) Stress coping strategies, which reflected the benefits of connecting with friends, work colleagues, church, and support groups; and (3) fueling up with recognition, reflecting the support offered by connecting with health professionals, teachers, and therapists.

Life Satisfaction

Intimate sources of connection and support included parents' immediate family relationships with children and spouses. For the parents in this research, their immediate families were held in the highest regard, had the greatest importance, and were seen as the core of their lives. For some, parenthood fulfilled their childhood dreams. These parents were greatly influenced by the high importance placed on Chinese traditional values of consanguinity. They felt that raising a child was the extension of their lives to the next generation, therefore, simply being a parent was a source of life satisfaction.

> *I have loved children since I was a small girl. [...] Taking care of children and being a housewife to take care of my own children are my dream jobs. [...] I feel that I wanted to have a child to extend my life in the future.* (Grace)

Contributing to their family members, seeing them happy, and spending time with them were vital to parents' sense of purpose and personal contentment.

> *My family is my first priority and I see them all the time. My family refers to my wife and my children. If they are happy, I will be happy too. [...] Their happiness is the source of my happiness.* (Ethen)

Like some others in this sample, Ethen found both happiness in his children and a way to pass on what he had learned, his core values and life experience, which was a source of real satisfaction to him:

> *We (he and his son) discussed a lot of things and people around us when we were alone. We talked about people around us and judged whether their behavior was appropriate. This is a good approach - talking to him and sharing my way of thinking with him would make him more accepting of what I suggest.* (Ethen)

Parents expressed contentment when seeing their children happy and making gains. Parents received love, a sense of status and respect from their children, and experienced satisfaction and happiness through being valued by their family members. Fathers also appreciated their wives and mentioned wanting and valuing time alone with them. They cherished the intimate relationship with their wives which acted as a powerful source of support and satisfaction for them. Chris said, "I have just jogged with my wife, and we saw the sunset, this is also a kind of happiness that you do not really need to chase after."

Stress Coping Strategies

Parents' external networks referred to people they connected with outside their immediate families, such as friends, the workplace, church, and parent support groups. Parents indicated that these networks were the core of their interpersonal relationships before they became parents. However, the importance of these forms of friendship now varied among parents. Some preferred to keep their worries within the intimate family and resisted sharing these externally. Others continued to value their external network, which allowed them to connect to the world. Speaking with others, getting information, and hearing their views and experiences offered new ideas to help them cope with stress. Although these differences might reflect personality trait differences (e.g., along the introversion–extraversion spectrum), in this study, differences seemed gender-related or linked to church membership.

Mothers typically said that when under stress, they spoke with members of their external network. Sharing was an important stress moderator for mothers as they enjoyed sharing their worries and benefited from advice from their peers. In return, they showed their love and care to their friends in order to maintain the relationship.

> *When my son does something that makes me really angry or if I encounter family problems, when these unhappy events happen, I will talk to them (her friends). Even though they may not comfort me, they listen to me, and we exchange our opinions. This makes me feel better straight away. I do not need to face the problems alone. I do not need to hide; I do not like to hide my feelings.* (Ann)

However, this aspect was different for fathers in this study. Fathers knew that friends could provide new perspectives and information on the issues that they were facing, but preferred to share their worries with their wives, seldom proactively talking about serious issues with their external networks. Though fathers rarely shared worries beyond the immediate family, they reported finding it acceptable to share with peers in parent support groups, whom they knew would understand their ASD children's difficulties. Support groups helped them release stress, share, and compare their experiences with others in similar positions, and changed their perspectives on their own children's situation in a positive direction. Chris added, "During training, I met some other parents and kids, we shared our experiences and our kids' situation. This changed my mindset, and I became less pessimistic. Escaped from my (pessimistic) world."

Some parents had reduced both the effort to maintain their pre-parenthood external networks and the amount of time they spent with them. For example, the only non-Christian participant claimed that she had almost no external contacts outside of her immediate family. However, for some of the parents describing themselves as Christian, the fellowship group at church and its weekly gatherings were an important external source of contact and support, including during the COVID-19 pandemic restrictions. Belle explained, "I did online worship so I did not need to physically go to the church. This gave me more opportunities to connect with other people, communicate and interact […] I think this is one of the things makes me happy."

Fueling Up with Recognition

Parents had many opportunities to meet the professionals working in their children's learning and health services, such as teachers, therapists, psychologists, and doctors. The relationship between parents and helping professionals was defined as a professional network. Most parents initially struggled to adapt and accept their ASD children's traits and diagnosis, but these parents' stories demonstrated that stress and frustration characterized their whole

parenting journey. Parents whose children showed a serious developmental delay early in life expressed not wanting to miss any opportunity to ensure they received the support they needed at the "golden" period for training, that is, before six years old. They saw themselves as fully responsible for their children's future and determined to do their best for them. Parents invested a great deal of time, money, and hope in engaging with various forms of training with their children.

> *We took him for a lot of training, a lot of intensive training every day. He had attention deficits; his eye-motor coordination was weak. The training helped him a lot, we took him for horse riding, piano lesson and table tennis. Therefore, it was really busy.* (Chris)

Parents received strong support from the helping professionals in the hospitals, training centers, and schools. The relationship between parents and helping professionals was essential to parents. Parents valued this relationship as they needed professional advice to raise their children and monitor their development progress. Not only did professionals support their children, but their involvement was also rewarding and reassuring for parents:

> *My son's therapists gave me and my son an award to praise our efforts in the training program. They said that I was a hardworking mom and willing to listen to their explanations. I received professional advice from them and this helped me to demystify the situation.* (Heather)

The extract from Heather's interview reflected the importance of professional recognition of her work and achievements. This validation was a very positive experience that contributed greatly towards her well-being, reinforced her involvement in their children's program, and increased her willingness to face challenges and difficulties.

Theme 3: Self-Care

Activities in which parents actively engaged to maintain their happiness and well-being along the stressful parenting journey formed the Self-Care theme. Three sub-themes within this theme were: (1) relaxation, (2) 'me time,' and (3) self-compassion.

Relaxation

Parents spoke about feeling relaxed and happy with their children. They also enjoyed being with them when undertaking shared activities such as traveling, watching football matches, and enjoying the beauty of nature.

> *Most of the happy moments are about play. We watched football, basketball matches, we hiked. My son loves sports, er, as he has ASD, he has the privilege to go to the theme park for free. So, we always go there to spend time there. We have the same hobbies, so, always, I go out with him after work, we play together and spend time together. These are unforgettable moments to me.* (Chris)

> *For example, we cannot go traveling (to other countries) now. Maybe we will go to the countryside together and have experiences that we share together. We also discover new things; for example, we (with her*

> *children) enjoy having a day trip to the local island. [...] these experiences are really valuable to me.* (Heather)

'Me Time'

All parents agreed that they needed 'me time,' which was time for being alone. Parents took pleasure in many different activities when alone, including watching TV and movies, enjoying good food, reading books, and meditation. Parents tried to squeeze some leisure time into their routine every day, which offered them a brief escape andan opportunity to destress. Heather said, "I like being alone in a quiet place. I like to think deeply about things for a long time. I like to go to the seaside. I feel relaxed when I hear the waves." Similarly, Ethen added, "Even though the movie is not that appealing, in that hour I can escape from reality. On the other hand, it is too much of a luxury for me to spend a lot of time on a hobby."

As Ethen explained, however, participants found that their opportunities for personal hobbies were few, given the needs of their families. Some had previously enjoyed hiking and outdoor activities, but as these activities were considered too time-consuming, they had given them up since the birth of their child(ren).

Self-Compassion

Self-compassion was related to nurturing oneself with kindness and gentleness (Neff et al., 2003). Parents described engaging in concrete efforts to treat themselves better and support their positive emotional well-being. One mother said she would dress-up and have a manicure because enhancing her appearance was a way of showing love and caring for herself. Other deeper self-compassion activities included using self-talk to bring comfort when under stress, avoiding self-blame when making mistakes, and accepting and reframing their difficult situations, as demonstrated in the following extracts. Chris said, "I told myself, 'This is not the end of the story; many things are still uncertain." Grace added,

> *They (Her husband's family) thought my son wasjust stupid. They did not pay attention and were not willing to understand more about my son and ASD. They just thought he was stupid just like his father. His father was a slow learner. [...] I talk to myself: it is no use being angry or thinking too much about something that has distressed you. I tell myself to allow myself some space to adapt to the fact. I will do something else. I talk to myself: when I am unhappy, I would cry. This is an emotion.* (Grace)

Theme 4: A Better Me

This theme reflected participants' inner strength, which allowed them to maintain a good life and find a "better me." Under this theme, there were four sub-themes: (1) embracing suffering, (2) making sacrifices, (3) gratitude, and (4) living with purpose.

Embracing Suffering

Experiencing and overcoming difficulties was considered a meaningful experience that contributed to the happy experiences of parents. Heather explained:

> *I encountered some obstacles in my life. The obstacles were very difficult to overcome, difficulties I could never have imagined. When I reached the lowest point in my life, I then bounced back. I finally understood that this is the feeling of happiness.*

In the current study, seven out of eight participants were Christians. Though there were a lot of difficulties and painful experiences when raising their ASD children, parents' interpretations of these experiences were influenced by their Christian faith. For example, they believe that suffering in life is not purely negative but has meaning.

> *I think I have humbled myself to follow God's will. All the sufferings are arranged by God and he wants to test me. He wants me to learn the God of love through life experience. Therefore, I always try my best to live according to God's commandments when making decisions in my life. Through this, I find peace of mind.* (Faye)

Many participants believed that their faith had brought them hope in difficult times and that God had helped them overcome their difficulties. They also perceived their unique parenting experiences as tools they could use to help others. They participated in voluntary work to share their experiences and help other less experienced parents and gained happiness through helping others. Participants also expressed that parenting their special needs children had given them a deeper life experience, greater empathy, and understanding of others' feelings. Diana added, "I learned to 'play it by ear', I find God is so good to me. [...] Be kind to others, help others, keep doing these things can sustain happiness."

Making Sacrifices

Most parents considered parenthood an exceptional and extraordinary opportunity to experience and find the meaning of life. A key meaning of parenthood was to make sacrifices based on responsibility or love. Parents were willing to sacrifice based on the responsibility of parenthood. When parents were asked whether they created happiness for themselves, half responded that they did not really think in that way. Instead, mindful of their new responsibilities, they put aside their own desires and put others ahead of themselves.

> *If I visualize the situation now, I am at the center of this picture. Other subjects in this picture are drawn from the center, the center is me. After I became a mom, there were more people in this picture. These people were brought by me, I need to be responsible for my choices. Therefore, I need to respect this picture.* (Heather)

The love parents experienced through the parent-child relationship led them to make sacrifices for their children. Examples of love and sacrifice could be found in the accounts of several mothers whose experience of pregnancy and parenthood had been complicated by suffering from depression. They all agreed that without love, they could not go through those difficult times.

> *I think I can bear witness to love. (sobbing…) I never imagined being a mom could be so harsh. (sigh…) When I look back on the time of my pregnancy, I had expected that it would be tough but it was way beyond what I could have possibly imagined. [...] I felt very uncomfortable, extremely uncomfortable. I remembered I suffered from depression*

> *during pregnancy. [...] I never imagined that I would have such a great love for overcoming all these difficulties. [...] I discovered that being a mom means sacrificing so much and the love I feel is so great. This is what I feel about being a mom, a mom is really great.* (Belle)

Gratitude

All parents expressed that gratitude was very important for maintaining a positive mindset. Many explained this concept using the same metaphor, 'The glass is half full or half empty.' They preferred to consider what they possessed rather than what they did not. The simple things in their everyday lives brought them peace and life satisfaction because they were grateful for what they had.

Ann said, "I am satisfied with my life, I have a place to live, food to eat. That's enough."

> *Positive thinking is like the metaphor of the half-full glass of water. You can keep yourself positive by being grateful all the time because I am a religious person. Always being kind to others is very important, this builds up the interaction among people.* (Diana)

Living with Purpose

Many parents defined happiness as living with purpose. They explained that they enjoyed achieving or, even better, exceeding the goals and purpose they had set for themselves and others. For example, purpose included achieving career goals, raising children, or adapting to a new life after immigration. These participants made plans to help them achieve their goals, bringing them life satisfaction and happiness.

> *Happiness is related to having a purpose and goal. It is hard to be happy without a purpose and a goal, regardless of whether I have an ASD child or not. During the process of raising my own child, I need to find my purpose and goals as a parent.* (Heather)

Discussion

This study aimed to gather the lived happiness experiences and meanings of Hong Kong parents of children with ASD. The experiences, perceptions, and meaning of happiness for our participants formed four themes: a growth mindset, connectedness, self-care, and a better me. Parents considered happiness to be a crucial element in their life, so, despite the challenges of raising a child with ASD in Hong Kong, they actively sought it out. They engaged actively in activities to enhance the sense of happiness, satisfaction, or meaning in their life.

Consistent with existing literature (C. Huang et al., 2013; Lai et al., 2015; Papadopoulos, 2021; Tsermentseli & Kouklari, 2021), parents in the current study spoke of their anxiety about their children's future and developmental delay during the diagnostic period. Most spoke of a 'dark period' following their children's diagnosis with ASD, which was a traumatic experience for most and led them to depressive symptoms and catastrophizing thought patterns. Some parents reported themselves as suffering from depressive symptoms, which also aligns with the existing evidence that parents of ASD children have poor psychological outcomes, including self-stigma, distress, and low levels of happiness (Al-Oran et al., 2020; Aktan et al., 2020; EdUHK, 2016). One mother described experiencing significant stigma within her wider family, in keeping with previous research demonstrating the social stigmatization of ASD children and families (Tait et al., 2015). In fact, most parents in our research did not explicitly raise the issue

of stigma; however, the preference of some to reduce sharing their worries outside the immediate family may implicitly reflect this. It is also possible that these parents achieved a degree of protective personal resilience and growth (Calhoun & Tedeschi, 2004).

Despite, and perhaps because of, their challenges and difficulties, our parents had gone on to experience PTG or personal growth and development, leading them to achieve the authentic happiness they described and maintain a positive well-being. Waizbard-Bartov et al. (2018) noted that when parents of ASD children become conscious that the original, traditional parenting schema does not fit their own family's situation, they need to alter their mindset in order to be resilient in the face of the crisis that having a child with ASD represents. Our findings suggested that many of these parents had indeed undergone that mindset change and developed some resilience to the various challenges they had faced. The parental adjustments evident in the current study were resonant with the adjustments described by Calhoun and Tedeschi (2004) in their model of PTG, including an appreciation for life, changes in perception of what is important, meaningful relationships with others, sense of personal strength, new opportunities and spiritual or existential development.

The current research also extended Waizbard-Bartov et al.'s (2018) hypothesis that the notion of PTG is adaptable to the cultural context in its capacity to change core values and fixed parental roles. For instance, these Hong Kong parents were rooted in traditional Chinese culture and values, which set demanding parental roles and high expectations of children (Chao, 1994; Chen, 2015; Ng et al., 2014; Tait et al., 2015). Reconstructing and reorganizing the parental schema brought these parents a new perspective on and expectations for their children. Most became less concerned with or influenced by the pressure to raise an elite child. Besides, parents' openness led them to be more appreciative of their children's abilities outside academic achievement, focusing instead on their kindness, conduct, determination, and filial piety (孝順). This shift in perception could buffer the effects of self-stigma and social stigma on raising children with disabilities in Chinese society.

The findings were also reminiscent of concepts encapsulated within the PERMA model (Seligman, 2011). One concept within the PERMA model was positive emotion, which was evident in various themes in the current study. In the Connectedness theme, parents spoke about the positive contributions of others to their positive emotions. In the *self-care* theme, parents found ways to destress and show themselves compassion. In the *a better me*, the gratitude sub-theme, parents spoke about feeling grateful for what they had and resisting focusing on what was missing, leading to greater satisfaction with their children and their life. Similarly, Timmons et al. (2017) found that mothers of ASD children who proactively embraced thankfulness and gratitude experienced enhanced child-parent relationships and appreciation of their children. This is in keeping with literature that demonstrates an association between positive emotions in parenting and parental fulfillment, gratitude, and pride (Nelson et al., 2014; Rafferty et al., 2020). Positive emotion also enhances adaptive coping (Lai et al., 2015), helping parents to accept difficulties or limitations in their ASD children's social functioning, communication (G. Lee, 2009; Ling et al., 2010; Rao & Beidel, 2009) learning (L. Lee et al., 2007) and behavior (Lecavalier et al., 2006; C. Huang et al., 2013). Positive emotion, therefore, plays a significant role in mitigating parental stress.

A second relevant PERMA concept was meaning (Seligman, 2011). The notion of meaning was significant to these parents and was reflected in the *a better me* theme. Parents saw meaning in the distress and difficulties they had encountered in the parenting journey with their children with ASD. The meaning of these experiences lay in accepting suffering, growing stronger as people, developing greater empathy, and helping others. Wang et al. (2007) found that recognizing meaning and purpose in life reduces stress and suicidal behavior. Although parents spoke about the importance of meaning, and there was the sense that this contributed to their overall happiness, quantitative research would be required to support a clear or causal link.

This study has provided novel insights into the experiences and meaning of happiness among parents of children with ASD in Hong Kong. The small sample and qualitative design mean that the findings may not be generalizable to other parents. Additional quantitative research with larger samples could be conducted to investigate possible links which emerged. For example, there seemed to be differences between male and female participants in preferences for and impact on the happiness of sharing outside the family and for spending time with spouse. For the male participants, the couple's intimacy was an invaluable source of happiness, but female participants rarely mentioned their partner's importance to them and the family. Numerous quantitative studies (Higgins et al., 2005; Hoseinnejad et al., 2020) have found that couples with ASD children have lower levels of marital happiness and relationship satisfaction compared with control group couples. The importance of couple satisfaction as a predictor of family functioning (Gau et al., 2012; Greenlee et al., 2022; Pedro et al., 2015) and quality of life (Benson, 2014; Brisini & Solomon, 2020) is well established. Johnson and Piercy's (2017) qualitative study specifically explored the couple intimacy experiences of parents with ASD children and led to the development of a new process model of how couples raising children with ASD negotiate intimacy. This model highlighted the cognitive and relational shifts couples had to make in negotiating couple intimacy. These shifts included changing expectations, consenting to find moments for intimacy, making conscious efforts towards intimacy, and sharing childcare to facilitate Me and couple time. Relationship intimacy mitigated stress and enhanced family functioning. Recent studies on fathers' perspectives in ASD families consistently report a lack of couple intimacy-related and 'me time,' as well as fathers taking on the traditional breadwinner role and taking less of a parenting role (e.g., Cook et al., 2005; Lewington et al., 2021; Lien et al., 2021; Rafferty et al., 2020). Similarly, in our Self-Care theme, parents described having very limited time for themselves, giving up previous hobbies, or feeling guilty about personal interests, and in the Connectedness theme, some parents had not maintained their earlier close friendships. Future research on gender differences associated with marital satisfaction, couple intimacy, and happiness in Hong Kong families with ASD children would help develop our understanding of psychological well-being within this group.

The research also has implications. Because of the apparent relevance of the PERMA model to parenting experiences within this participant group, the model could be considered an educational tool to support parents in focusing on or developing skills and attitudes to boost their well-being. Its impact on well-being could be measured using, for example, the Satisfaction with Life Scale (Diener et al., 1985) or the General Health Questionnaire (Goldberg et al., 1997). These findings could be helpful to researchers and policymakers in developing welfare policy and social support for these vulnerable families and parents.

This is the first study to capture the happiness experiences of Chinese ethnic parents of children with ASD in Hong Kong. It adds to the small existing body of knowledge and research in the area and highlights the relevance of existing theoretical frameworks, such as the PERMA model (Seligman, 2011), PTG, and crisis-related growth (Calhoun & Tedeschi, 2004; Waizard-Bartov et al., 2018) in parents of ASD children. Current interventions to support children within the local health system (Yi et al., 2020) could be accompanied by parental support and education, helping parents to enhance their psychological well-being, and through family-related counseling based on theoretical concepts from the PERMA model and PTG ideas. This could fill the current policy and support gap and address the unmet psychological support needs for this minority group.

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References

Aktan, O., Orakcı, Ş., & Durnalı, M. (2020). Investigation of the relationship between burnout, life satisfaction and quality of life in parents of children with disabilities. *European Journal of Special Needs Education*, *35*(5), 679–695. https://doi.org/10.1080/08856257.2020.1748429

Al-Oran Haytham Mohammad, & Khuan Lee. (2021). Predictors of parenting stress in parents of children diagnosed with autism spectrum disorder: A scoping review. *The Egyptian Journal of Neurology, Psychiatry and Neurosurgery*, *57*(1), Article 103. https://doi.org/10.1186/s41983-021-00345-w

American Psychiatric Association (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). DSM-5.

Benson, P. R. (2014). Coping and psychological adjustment among mothers of children with ASD: An accelerated longitudinal study. *Journal of Autism and Developmental Disorders*, *44*(8), 1793–1807. https://doi.org/10.1007/s10803-014-2079-9

Bhaskar, R. (1978). *A realist theory of science*. Harvester Press.

Boniwell, I., Tunariu, A. D., & Hefferon, K. (2019). *Positive psychology: Theory, research and applications* (2nd ed.). Open University Press.

Boyle, G., Saklofske, D. H., & Matthews, G. (2014). *Measures of personality and social psychological constructs* Elsevier Science & Technology. https://doi.org/10.1016/C2010-0-68427-6

Braun, V., & Clarke, V. (2021). One size fits all? What counts as quality practice in (reflexive) thematic analysis? *Qualitative Research in Psychology*, *18*(3), 328–352. https://doi.org/10.1080/14780887.2020.1769238

Brisini, K. S. C., & Solomon, D. H. (2020). Children's transitions and relational turbulence in marriage: Can transition processing communication help? *Journal of Family Communication*, *20*(1), 82–96. https://doi.org/10.1080/15267431.2019.1662421

Calhoun, L. G., & Tedeschi, R. G. (2004). The foundations of oosttraumatic growth: New considerations. *Psychological Inquiry*, *15*(1), 93–102. https://doi.org/10.1207/s15327965pli1501_03

Cappadocia, M. C., Weiss, J. A., & Pepler, D. (2011). Bullying experiences among children and youth with autism spectrum disorders. *Journal of Autism and Developmental Disorders; J Autism Dev Disord, 42*(2), 266–277. https://doi.org/10.1007/s10803-011-1241-x

Carmassi, C., Dell'oste, V., Foghi, C., Bertelloni, C. A., Conti, E., Calderoni, S., Battini, R., & Dell'osso, L. (2021). Post-traumatic stress reactions in caregivers of children and adolescents/young adults with severe diseases: A systematic review of risk and protective factors. *International Journal of Environmental Research and Public Health*, *18*(1), 1–20. https://doi.org/10.3390/ijerph18010189

Casey, L. B., Zanksas, S., Meindl, J. N., Parra, G. R., Cogdal, P., & Powell, K. (2012). Parental symptoms of posttraumatic stress following a child's diagnosis of autism spectrum disorder: A pilot study. *Research in Autism Spectrum Disorders*, *6*(3), 1186-1193. https://doi.org/10.1016/j.rasd.2012.03.008

Centers for Disease Control and Prevention. (2021). *Data and statistics on autism spectrum disorder*. https://www.cdc.gov/ncbddd/autism/data.html

Chao, R. (1994). Beyond parental control and authoritarian parenting style: Understanding Chinese parenting through the cultural notion of training. *Child Development, 65*(4), 1111–1119. https://doi.org/10.1111/j.1467-8624.1994.tb00806.x

Chen, W.-W. (2015). The relations between perceived parenting styles and academic achievement in Hong Kong: The mediating role of students' goal orientations. *Learning and Individual Differences, 37,* 48–54. https://doi.org/10.1016/j.lindif.2014.11.021

Clarke, V., & Braun, V. (2013). *Successful qualitative research: A practical guide for beginners*. SAGE Publications.

Cook, J. L., Jones, R. M., Dick, A. J., & Singh, A.. (2005). Revisiting men's role in father involvement: The importance of personal expectations. *Fathering (Harriman, Tenn.), 3*(2), 165–178. https://doi.org/10.3149/fth.0302.165

Crisp (2020). *Essential social psychology* (4th edition.). SAGE Publications.

Dellapiazza, F., Audras-Torrent, L., Michelon, C., & Baghdadli, A. (2021). Clinical characteristics of children with ASD and comorbid ADHD: Association with social impairment and externalizing and internalizing behaviours. *Research in Developmental Disabilities, 113,* 103930–103930. https://doi.org/10.1016/j.ridd.2021.103930

Diener, E., Emmons, R. A., Larsen, R. J., & Griffin, S. (1985). The Satisfaction with Life Scale. *Journal of Personality Assessment, 49*(1), 71–75. https://doi.org/10.1207/s15327752jpa4901_13

Dweck, C. S. (2006). *Mindset: The new psychology of success: Vol. February 2012*. Random House Publishing Group.

Education Bureau of the Government of the Hong Kong Special Administrative Region. (2019). *EDB to enhance support for students with autism spectrum disorders*. https://www.info.gov.hk/gia/general/201910/03/P2019100300291.htm?fontSize=1

Fiske, S. T., & Taylor, S. E. (1991). *Social cognition* (2nd ed.). McGraw-Hill.

Forrest, D. L., Kroeger, R. A., & Stroope, S. (2019). Autism spectrum disorder symptoms and bullying victimization among children with autism in the United States. *Journal of Autism and Developmental Disorders*, *50*(2), 560–571. https://doi.org/10.1007/s10803-019-04282-9

Frey, B. S., & Stutzer, A. (2010). Happiness and public choice. *Public Choice*, *144*(3/4), 557–573. https://doi.org/10.1007/s11127-010-9681-y

Gau, Chou, M.-C., Chiang, H.-L., Lee, J.-C., Wong, C.-C., Chou, W.-J., & Wu, Y.-Y. (2012). Parental adjustment, marital relationship, and family function in families of children with autism. *Research in Autism Spectrum Disorders, 6*(1), 263–270. https://doi.org/10.1016/j.rasd.2011.05.007

Goldberg, D. P., Gater, R., Sartorius, N., Ustun, T. B., Piccinelli, M., Gureje, O., & Rutter, C. (1997). The validity of two versions of the GHQ in the WHO study of mental illness in general health care. *Psychological Medicine, 27*(1), 191–197. https://doi.org/10.1017/S0033291796004242

Greenlee, J. L., Piro-Gambetti, B., Putney, J., Papp, L. M., & Hartley, S. L. (2022). Marital satisfaction, parenting styles, and child outcomes in families of autistic children. *Family Process, 61*(2), 941–961. https://doi.org/10.1111/famp.12708

Held, B. S. (2002). The tyranny of the positive attitude in America: Observation and speculation. *Journal of Clinical Psychology*, *58*(9), 965–991. https://doi.org/10.1002/jclp.10093

Held, B. S. (2004). The negative side of positive psychology. *The Journal of Humanistic Psychology*, *44*(1), 9–46. https://doi.org/10.1177/0022167803259645

Higgins, D. J., Bailey, S. R., & Pearce, J. C. (2005). Factors associated with functioning style and coping strategies of families with a child with an autism spectrum disorder. *Autism: the International Journal of Research and Practice*, *9*(2), 125–137. https://doi.org/10.1177/1362361305051403

Hoseinnejad, H., Chopaniyan, F., Sarvi Moghanlo, O., Rostami, M., & Dadkhah, A. (2020). Marital satisfaction and happiness in parents with autistic and normal children. *Iranian Rehabilitation Journal*, *18*(1), 49–56. https://doi.org/10.32598/irj.18.1.655.1

Huang, A. X., Hughes, T. L., Sutton, L. R., Lawrence, M., Chen, X., Ji, Z., & Zeleke, W. (2017). Understanding the self in individuals with Autism Spectrum Disorders (ASD): A review of literature. *Frontiers in Psychology*, *8*, 1422–1422. https://doi.org/10.3389/fpsyg.2017.01422

Huang, C.-Y., Yen, H.-C., Tseng, M.-H., Tung, L.-C., Chen, Y.-D., & Chen, K.-L. (2013). Impacts of autistic behaviors, emotional and behavioral problems on parenting stress in caregivers of children with autism. *Journal of Autism and Developmental Disorders*, *44*(6), 1383–1390. https://doi.org/10.1007/s10803-013-2000-y

Husser, J. A., & Fernandez, K. E. (2017). We are happier than we realize: Underestimation and conflation in measuring happiness. *Journal of Happiness Studies*, *19*(2), 587–606. https://doi.org/10.1007/s10902-016-9831-0

Huta, V. (2016). An overview of hedonic and eudaimonic well-being concepts. In L. Reinecke & M. B. Oliver (Eds.), *The Routledge handbook of media use and well-being: International perspectives on theory and research on positive media effects* (pp. 14–33). Routledge. https://doi.org/10.4324/9781315714752

Ivtzan, I., Lomas, T., Hefferon, K., & Worth, P. (2015). *Second wave positive psychology: Embracing the dark side of life*. Routledge. https://doi.org/10.4324/9781315740010

Johnson, J., & Piercy, F. P. (2017). Exploring partner intimacy among couples raising children on the autism spectrum: A grounded theory investigation. *Journal of Marital and Family Therapy*, *43*(4), 644–661. https://doi.org/10.1111/jmft.12247

Lai, W. W., Goh, T. J., Oei, T. P. S., & Sung, M. (2015). Coping and well-being in parents of children with Autism Spectrum Disorders (ASD). *Journal of Autism and Developmental Disorders*, *45*(8), 2582–2593. https://doi.org/10.1007/s10803-015-2430-9

Lecavalier, L., Leone, S., & Wiltz, J. (2006). The impact of behaviour problems on caregiver stress in young people with autism spectrum disorders. *Journal of Intellectual Disability Research*, *50*(3), 172–183. https://doi.org/10.1111/j.1365-2788.2005.00732.x

Lee, G. K. (2009). Parents of children with high functioning autism: How well do they cope and adjust? *Journal of Developmental and Physical Disabilities*, *21*(2), 93–114. https://doi.org/10.1007/s10882-008-9128-2

Lee, L.-C., Harrington, R. A., Louie, B. B., & Newschaffer, C. J. (2007). Children with autism: Quality of life and parental concerns. *Journal of Autism and Developmental Disorders*, *38*(6), 1147–1160. https://doi.org/10.1007/s10803-007-0491-0

Lewington, L., Lee, J., & Sebar, B. (2021). "I'm not just a babysitter": Masculinity and men's experiences of first-time fatherhood. *Men and Masculinities*, *24*(4), 571–589. https://doi.org/10.1177/1097184X21993884

Lien, K., Lashewicz, B., Mitchell, J., & Boettcher, N. (2021). Blending traditional and nurturing fathering: Fathers of children with autism managing work and family. *Family Relations*, *70*(1), 264–281. https://doi.org/10.1111/fare.12472

Ling, C. Y. M., Mak, W. W. S., & Cheng, J. N. S. (2010). Attribution model of stigma towards children with autism in Hong Kong. *Journal of Applied Research in Intellectual Disabilities*, *23*(3), 237–249. https://doi.org/10.1111/j.1468-3148.2008.00456.x

Liu, X., & To, S. (2020). Personal growth experience among parents of children with autism participating in intervention. *Journal of Autism and Developmental Disorders*, *51*(6), 1883–1893. https://doi.org/10.1007/s10803-020-04681-3

Mak, W. W. S., & Kwok, Y. T. Y. (2010). Internalization of stigma for parents of children with autism spectrum disorder in Hong Kong. *Social Science & Medicine, 70*(12), 2045–2051. https://doi.org/10.1016/j.socscimed.2010.02.023

Murray, M. J. (2010). Attention-deficit/hyperactivity disorder in the context of autism spectrum disorders. *Current Psychiatry Reports*, *12*(5), 382–388. https://doi.org/10.1007/s11920-010-0145-3

Neff, K. D. (2003). The development and validation of a scale to measure self-compassion. *Self and Identity, 2*(3), 223–250. https://doi.org/10.1080/15298860390209035

Nelson, S. K., Kushlev, K., & Lyubomirsky, S. (2014). The pains and pleasures of parenting: When, why, and how is parenthood associated with more or less well-being? *Psychological Bulletin, 140*(3), 846–895. https://doi.org/10.1037/a0035444

Ng, F. F.-Y., Pomerantz, E. M., & Deng, C. (2014). Why are chinese mothers more controlling than american mothers? "My child is my report card." *Child Development*, *85*(1), 355–369. https://doi.org/10.1111/cdev.12102

Ooi, K. L., Ong, Y. S., Jacob, S. A., & Khan, T. M. (2016). A meta-synthesis on parenting a child with autism. *Neuropsychiatric Disease and Treatment*, *2016*(1), 745–762.

Papadopoulos, D. (2021). Mothers' experiences and challenges raising a child with autism spectrum disorder: A qualitative study. *Brain Sciences*, *11*(3), 1–16. https://doi.org/10.3390/brainsci11030309

Pedro, M. F., Ribeiro, T., & Shelton, K. H. (2015). Romantic attachment and family functioning: The mediating role of marital satisfaction. *Journal of Child and Family Studies*, *24*(11), 3482–3495. https://doi.org/10.1007/s10826-015- 0150-6

Rafferty, D., Tidman, L., & Ekas, N. V. (2020). Parenting experiences of fathers of children with autism spectrum disorder with or without intellectual disability. *Journal of Intellectual Disability Research, 64*(6), 463–474. https://doi.org/10.1111/jir.12728

Rao, P. A., & Beidel, D. C. (2009). The impact of children with high-functioning autism on parental stress, sibling adjustment, and family functioning. *Behavior Modification, 33*(4), 437–451. https://doi.org/10.1177/0145445509336427

Reed, P., & Osborne, L. A. (2012). Diagnostic practice and its impacts on parental health and child behaviour problems in autism spectrum disorders. *Archives of Disease in Childhood*, *97*(10), 927–931. https://doi.org/10.1136/archdischild-2012-301761

Schoen, R., Kim, Y. J., Nathanson, C. A., Fields, J., & Astone, N. M. (1997). Why do americans want children? *Population and Development Review*, *23*(2), 333–358. https://doi.org/10.2307/2137548

Seligman, M. E. P. (2004). *Authentic happiness: Using the new positive psychology to realize your potential for lasting fulfillment*. Atria..

Seligman, M. E. P. (2011). *Flourish: A visionary new understanding of happiness and well-being*. Free Press.

Sun, I. Y. , Cortez, A. C. M. , & Fernandes, F. D. M. (2019). Learning disabilities in children with autism. In S. Misciagna (Ed.), *Learning disabilities: Neurological bases, clinical features and strategies of intervention* (pp. 12-21). IntechOpen. https://doi.org/10.5772/intechopen.89234

Tait, K., Fung, F., Hu, A., Sweller, N., & Wang, W. (2015). Understanding Hong Kong Chinese families' experiences of an autism/ASD diagnosis. *Journal of Autism and Developmental Disorders*, *46*(4), 1164–1183. https://doi.org/10.1007/s10803-015-2650-z

Tedeschi, R. G., & Calhoun, L. G. (1996). The posttraumatic growth inventory: Measuring the positive legacy of trauma. *Journal of Traumatic Stress*, *9*(3), 455–471. https://doi.org/10.1002/jts.2490090305

The Education University of Hong Kong. (2016, March 31). *Survey: Most parents of children with autism report discriminatory experiences*. https://www.eduhk.hk/fehd/en/highlights.php?id=1197

Timmons, L., Ekas, N. V., & Johnson, P. (2017). Thankful thinking: A thematic analysis of gratitude letters by mothers of children with autism spectrum disorder. *Research in Autism Spectrum Disorders*, *34*, 19–27. https://doi.org/10.1016/j.rasd.2016.11.009

Towle, P. O., Vacanti-Shova, K., Shah, S., & Higgins-D'alessandro, A. (2013). School-aged functioning of children diagnosed with autism spectrum disorder before age three: Parent-reported diagnostic, adaptive, medication, and school placement outcomes. *Journal of Autism and Developmental Disorders*, *44*(6), 1357–1372. https://doi.org/10.1007/s10803-013-1997-2

Trafimow, D., Triandis, H. C., & Goto, S. G. (1991). Some tests of the distinction between the private self and the collective self. *Journal of Personality and Social Psychology*, *60*(5), 649–655. https://doi.org/10.1037/0022-3514.60.5.649

Tsermentseli, S., & Kouklari, E.-C. (2021). Impact of child factors on parenting stress of mothers of children with autism spectrum disorder and intellectual disability: A UK school-based study. *Early Child Development and Care*, *191*(10), 1555–1566. https://doi.org/10.1080/03004430.2019.1658090

Veenhoven, R., & Ehrhardt, J. (1995). The cross-national pattern of happiness: Test of predictions implied in three theories of happiness. *Social Indicators Research*, *34*(1), 33–68. https://doi.org/10.1007/BF01078967

Voliovitch, Y., Leventhal, J. M., Fenick, A. M., Gupta, A. R., Feinberg, E., Hickey, E. J., Shabanova, V., & Weitzman, C. (2021). Parenting stress and its associated components prior to an autism spectrum disorder (ASD) diagnostic evaluation. *Journal of Autism and Developmental Disorders*, *51*(10), 3432–3442. https://doi.org/10.1007/s10803-020-04804-w

Waizbard-Bartov, E., Yehonatan-Schori, M., & Golan, O. (2018). Personal growth experiences of parents to children with autism spectrum disorder. *Journal of Autism and Developmental Disorders*, *49*(4), 1330–1341. https://doi.org/10.1007/s10803-018-3784-6

Wang, M.-C., Richard Lightsey, O., Pietruszka, T., Uruk, A. C., & Wells, A. G. (2007). Purpose in life and reasons for living as mediators of the relationship between stress, coping, and suicidal behavior. *The Journal of Positive Psychology*, *2*(3), 195–204. https://doi.org/10.1080/17439760701228920

Yakushko, O., & Blodgett, E. (2021). Negative reflections about positive psychology: On constraining the field to a focus on happiness and personal achievement. *The Journal of Humanistic Psychology*, *61*(1), 104–131. https://doi.org/10.1177/0022167818794551

Yi, H., Siu, Q. K. Y., Ngan, O. M. Y., & Chan, D. F. Y. (2020). Parents' experiences of screening, diagnosis, and intervention for children with autism spectrum disorder. *American Journal of Orthopsychiatry*, *90*(3), 297–311. https://doi.org/10.1037/ort0000433

Zhang, W., Yan, T. T., Barriball, K. L., While, A. E., & Liu, X. H. (2015). Post-traumatic growth in mothers of children with autism: A phenomenological study. *Autism: The International Journal of Research and Practice*, *19*(1), 29–37. https://doi.org/10.1177/1362361313509732

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Is Sedentary Behavior Associated With Executive Function in Children and Adolescents? A Systematic Review

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Background: Prolonged time on sedentary behavior, especially screen-based sitting time, is associated with unfavorable health indicators in children and adolescents. However, the effects of sedentary behavior on cognitive function remain to be elucidated.

Objective: The purpose of this systematic review was to synthesize the evidence on the associations of sedentary behavior with executive function in children and adolescents.

Methods: Four electronic databases (i.e., PubMed, Web of Science, PsycINFO, and SPORTDiscus) were searched for studies examining the associations between sedentary behavior and executive function in children and adolescents. Study quality was assessed by the NIH Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies.

Results: A total of 1,151 records were initially identified through database searches and other searches. Twelve cross-sectional and four longitudinal studies met the inclusion criteria. Of the 16 studies, seven studies found significant negative associations between sedentary behavior and executive function, and two studies presented positive associations. Eight studies measured sedentary time using accelerometers and showed varied associations between objectively measured sedentary time and executive function. Nine studies measured screen-based sedentary behavior, of which five studies found negative associations of sedentary time with executive function.

Conclusion: The available evidence on the associations between sedentary behavior and executive function is not conclusive in children and adolescents. However, screen-based sedentary behavior may be negatively associated with executive function.

Keywords: sedentary behavior, screen time, executive function, children, adolescents

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INTRODUCTION

Sedentary behavior is a distinct construct from physical activity, referring to any waking behaviors with an energy expenditure of <1.5 metabolic equivalent units (METs) while in a sitting, reclining, or lying posture (1). Common sedentary behaviors include prolonged sitting, screen-based behaviors (e.g., TV viewing, computer/tablet using, video gaming), etc. Time spent on sedentary behaviors can be self/parent-reported or be objectively monitored by wearable devices

such as accelerometers. Currently, sedentary time remains high in children and adolescents, and the trend continues to increase over the past few decades in some countries. According to the Global School-based Student Health Survey among 97 countries, about 25% of boys and girls aged 13–15 years old reported sitting longer than 3 h per day, in addition to sitting at school and for homework (2). From 2007 to 2016, the estimated total sitting time increased from 7 h per day to 8.2 h per day among adolescents in the United States (3). In China, an increasing trend of the prevalence of screen-based viewing time was also observed in school-age children (4).

Accumulating evidence showed that sedentary behavior, especially prolonged TV viewing, have been linked with increased risk of a variety of chronic diseases, such as obesity (5), type 2 diabetes (6, 7), cardiovascular diseases (8), and certain types of cancer (9). In children and adolescents, sedentary behaviors have also been linked with unfavorable health indicators, such as lower physical fitness (10), higher fatness (11), clustered cardiometabolic risk scores (12), and lower self-esteem (13). Even worse, emerging evidence has shown that excessive sedentary behaviors are associated with mental illness and poorer cognitive function (14, 15). However, the findings on the relationship between sedentary behavior and cognitive function are mixed. A systematic review included eight studies examining the associations of sedentary behavior with cognitive function in adults older than 40 years (15). It concluded that greater amounts of sedentary behaviors were associated with poorer cognitive function over the lifespan. A more recent systematic review including 13 cross-sectional and longitudinal studies suggested inconsistent evidence on the direction of the association of sedentary behavior with cognitive function in older adults with a mean age of 65+ years (16). Another systematic review in young children (≤5 years) found that different types of sedentary behavior may exert different influences on cognitive development (17). Screen time, particularly TV viewing, was either not associated with or negatively associated with cognitive skills. However, no existing studies have critically reviewed the literature of the association between sedentary behavior and executive function in children and adolescents.

Previous studies have shown that physical activity and fitness have beneficial effects on cognitive function in children and adolescents (18, 19). The effects are disproportionately larger for executive function (20). Executive function refers to a set of top-down mental processes needed for goal-directed behaviors, such as inhibitory control, working memory, cognitive flexibility, planning (21). Executive function is critical for school readiness, academic performance, and future career success (21, 22). However, evidence on the effects of sedentary behavior on executive function remains conflicting in children and adolescents. Some studies showed that sedentary behavior is not associated with executive function in childhood (23–25), whereas other studies found negative (26, 27) or positive associations (28). No existing systematic reviews have addressed this research gap. In addition, a preview review suggested a type-specific association between sedentary behavior and health indicators in children and adolescents (29). Another gap in the literature is the lack of the associations between type-specific sedentary behaviors and executive function.

Therefore, the current systematic review is aimed to synthesize the literature on the association of sedentary behavior (both objectively measured sedentary time and self/parent-reported screen-based behaviors) with executive function in children and adolescents.

METHODS

This systematic review was performed following the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement (30).

Search Strategy

Two authors (SL and JG) independently searched PubMed, Web of Science, PsycINFO, and SPORTDiscus from inception to April 2021. The combinations of the following three groups of retrieval items were used: (1) sedentary behavi*, screen time, sitting time, sedentary time, TV viewing, video gam*, computer use; (2) executive function, cognitive control, working memory, inhibitory control, cognitive flexibility, planning; (3) children, adolescents. The specific search strategy was slightly adjusted according to the search builder of each database.

Study Selection

Two authors (SL and JG) screened all the retrieved titles and abstracts to exclude duplicate or irrelevant studies. The two authors screened the full text of the remaining studies after removing duplicate and apparently irrelevant studies. Any disagreements about the study selection were discussed with a third author (TH) until a consensus was reached. The two review authors also searched the bibliographies of all included articles to ensure that all relevant studies were captured. Only the longitudinal result was extracted for the cohort studies that conducted both cross-sectional and longitudinal analyses in the same population.

Inclusion and Exclusion Criteria

The studies must meet the following inclusion criteria to be included: (1) studies with cross-sectional or longitudinal design examined the associations between sedentary behavior and executive function; (2) sedentary behavior was self/parent-reported (e.g., prolonged sitting, TV viewing, computer use, video gaming) or objectively monitored by wearable devices (e.g., accelerometers); (3) executive function was objectively assessed (paradigms including Flanker task, Stroop color-word test, N-back task, Tower of London task, Trail making task, etc.); (4) the participants were apparently healthy children and adolescents aged 5–17 years; (5) studies must be published in peer-reviewed journals; (6) English full text must be available.

Studies were excluded if the sedentary behaviors were not clearly classified or measured. Studies focusing on specific screen-based or non-screen contents (e.g., violent films, educational programs), screen-based active behavior (e.g., active video gaming), or specific learning behavior (e.g., reading, puzzles) were excluded. Studies were also excluded if the executive function was parent- or teacher-reported.

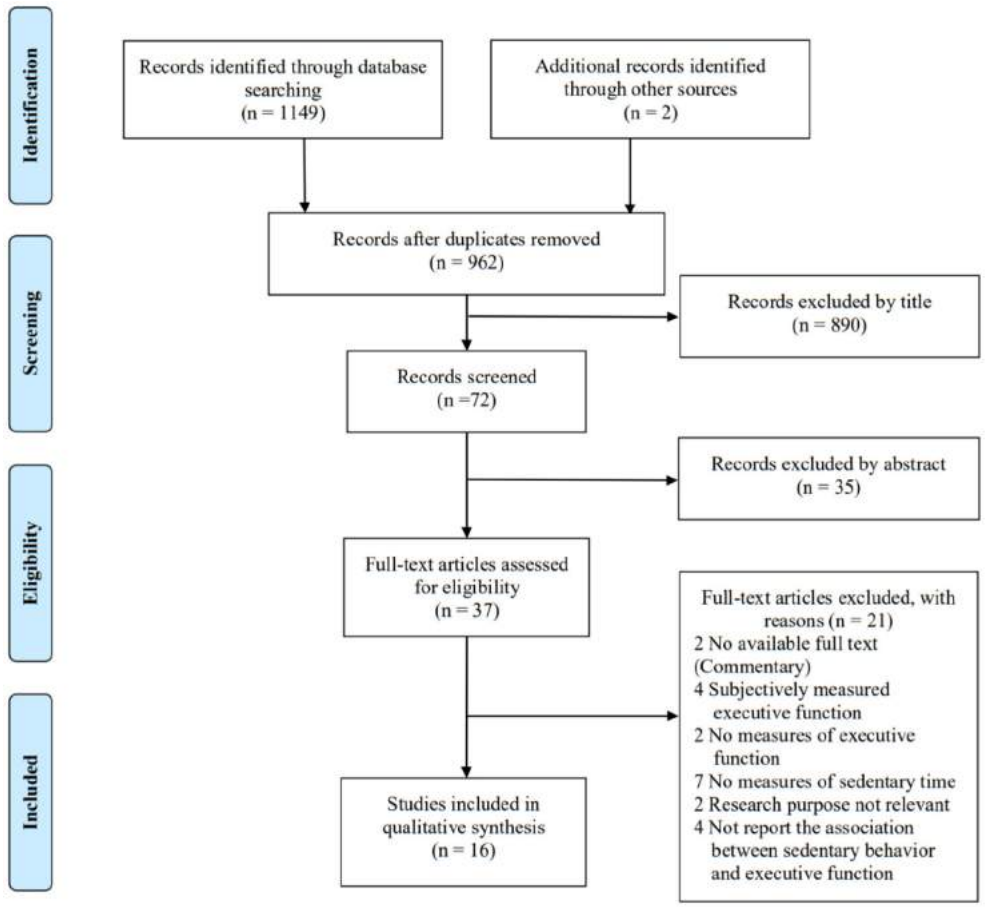

FIGURE 1 | PRISMA flow diagram of study selection.

Data Extraction

Data collection was conducted independently by two authors (SL and JG). Publication year, country, study design, sample size, covariates, measurement of sedentary behavior, assessment of executive function, and results were extracted from each included study and recorded.

Methodological Quality

The two authors assessed the quality of studies by the NIH Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies (31). This tool includes 14 items, and the reviewer could select "yes," "no," "cannot determine," or "not reported" on each item. The score for longitudinal studies ranges from 0 (the lowest quality) to 14 (the highest quality). For cross-sectional studies, three items are not applicable (items 10, 12, 13). The classifications of methodological quality are rated as "strong" (≥80%), "good" (70–79%), "fair" (60–69%), or "poor" (<60%) based on the percentage scores which are calculated as the number of "yes" responses divided by the total number of applicable items (32, 33). All discrepancies between reviewers were resolved through discussion among the reviewers or with a third reviewer if needed. The items of assessment tool are listed in **Supplementary Table 1**.

RESULTS

Search Results and Study Characteristics

A total of 1,149 records were identified through database searches, and additional two records were identified through reference list searches (see **Figure 1**). After removing duplicate records, 962 records remained. Following the screening of titles and abstracts, 37 articles were obtained for further full text review. Sixteen articles met the inclusion criteria after detailed assessment of the full-text, including 12 cross-sectional studies and four longitudinal studies.

Sedentary behaviors were measured by subjective assessment (self-reported or parent-reported) and objective assessment (accelerometer). Eight studies objectively measured sedentary time using accelerometers (25, 27, 28, 34–38). Nine studies surveyed a variety of screen-based behaviors as proxies of sedentary behaviors (i.e., total screen time, TV viewing, computer gaming, other computer use, general computer use, etc.) (23, 24, 26, 37, 39–43).

Across these 16 studies, a total of 21 cognitive tasks were used, measuring various aspects of executive function, including inhibitory control, working memory, cognitive flexibility, and planning. Study sample sizes ranged from 77 to 1,001. The participants were aged 5–17 years old. The countries of study locations were Norway, China, South Africa, Canada, Spain, United States, United Kingdom, Finland, and the Netherlands.

Study Findings

Of the included 16 studies with objectively measured sedentary time or screen-based sedentary behavior, seven studies (44%) found a negative association between sedentary behavior and executive function (23, 24, 26, 27, 40, 41, 43), while two studies (13%) found a positive association between sedentary bahavior and executive function (28, 34).

Objectively Measured Sedentary Time and Executive Function

Of the eight studies with objectively measured sedentary time (25, 27, 28, 34–38), one study (13%) found that more sedentary time was associated with poorer inhibitory control (27). Two of the eight studies (25%) demonstrated a positive association of sedentary time with one or more aspects of executive functions (inhibitory control, working memory, and planning) (28, 34), including one longitudinal study (28). Six studies (75%) observed no associations between objectively measured sedentary time and certain aspects of executive functions (25, 27, 35–38).

Screen-Based Sedentary Behavior and Executive Function

Nine studies investigated the associations of screen-based sedentary behaviors (i.e., total screen time, TV viewing, computer/video gaming, other computer use) with executive function (23, 24, 26, 37, 39–43). Of the nine studies, five studies (56%) found negative associations between screen-based sedentary behavior and certain aspects of executive function (26, 37, 40, 41, 43). Of note, one of them employed longitudinal study design (26). Eight studies (89%) observed no associations between screen-based sedentary behavior and certain aspects of executive functions (23, 24, 26, 37, 39–42).

Of the nine studies, three studies assessed the total screen time (26, 37, 39). Two of these studies (66%) showed that total screen time was not associated with executive function (working memory, cognitive flexibility) (37, 39). Only one study (33%) observed a negative association between total screen time and N-back performance in girls (26). Seven studies examined the associations between TV viewing and executive function (23, 24, 26, 37, 40, 41, 43). Of them, two studies (29%) found that more time on TV viewing was associated with poorer executive function (40, 43), and five studies (71%) did not find any associations between TV viewing and executive function (23, 24, 26, 37, 41). Three studies examined the association between general computer usages with executive function (40–42). One study found a positive association (40) and one study found a negative association (41). Two studies examined the associations between computer/video gaming and executive function (26, 37). Both of them found that spending more time on computer/video gaming was related to worse working memory.

Methodological Quality of Included Studies

The average score of cross-sectional studies was 6.67 (**Table 1**). The average score of longitudinal studies was 10. Detailed scores of quality assessment are also available in **Supplementary Table 1**.

DISCUSSION

The present study was aimed to critically review the evidence on the association between sedentary behavior and executive function in children and adolescents. Out of the 16 studies, seven studies (44%) found a negative association between sedentary behavior and executive function, while two studies (13%) presented positive associations. Eight studies measured sedentary time using an accelerometer, and showed varied associations of objectively measured sedentary time with executive function. Nine studies measured screen-based sedentary behavior, of which five studies (56%) found negative associations of sedentary time with executive function.

Eight of the included studies objectively measured sedentary time. The current review presented mixed results regarding the associations between objectively measured sedentary time and executive function in children and adolescents. It is impossible to conclude of the direction of the association between objectively measured sedentary time and executive function. Our findings are inconsistent with a systematic review in older adults, which indicated that shorter objectively assessed sedentary time was associated with better global cognitive function (44). Although the accelerometer-based measurements provided an objectively assessed sedentary time, they cannot distinguish the types of sedentary behavior. Children and adolescents may engage in cognitively active sedentary behavior, such as reading, and learning, benefiting cognitive development (45). A study further supports this idea. Brain connectivity was positively correlated with reading time and negatively correlated with screen-based media time (46). Therefore, when it comes to the associations between sedentary behavior and executive function in childhood, the types of sedentary behavior should be considered.

In this systematic review, nine included studies surveyed screen-based sedentary behavior, which provided some evidence on the association between type-specific sedentary behavior and executive function in children and adolescents. The majority of evidence suggests that screen-based sedentary time has either no effects or a detrimental effect on executive function in children and adolescents. Recent evidence found that the deleterious effects of sedentary behavior on cardio-metabolic health are most notable for screen-based behaviors (47). Regarding mental health, a study showed that only leisure screen-based sedentary behaviors are linked to worse perceived stress and anxiety (48). In the current study, the negative associations of sedentary behavior with executive function are mainly observed in the included studies that measured screen-based sedentary behaviors (i.e., TV viewing, computer use, video games, total screen time). These findings are in line with the systematic review in early childhood.

TABLE 1 | Characteristics and results table for included studies.

References, Country	Sample (1) *N* (% girls) (2) Age (years)	Measurement of sedentary behavior	Measurement of executive function	Adjusted covariates	Results
Cross-sectional studies					
Aadland et al. (34) Norway	(1) 697 (51%) (2) 10.2 ± 0.3	Accelerometer -Sedentary time	Stroop color-word test -Inhibitory control Digit span test -Working memory Verbal fluency & Trail making test -Cognitive flexibility	Age; body fat; pubertal status; birth weight; SES	Time on sedentary behavior was positively associated with working memory in girls, but with inhibitory control and cognitive flexibility in boys.
Chetty-Mhlanga et al. (39) South Africa	(1) 1,001 (53%) (2) 11 ± 1.7	Self-reported -Total screen time	Spatial working memory test (CANTAB) -Working Memory Multi-tasking test (CANTAB) -Cognitive flexibility	Age; sex; area; head injury; smoke; alcohol; drugs; farm residence; SES; mobile phone ownership; mother employment; mother education; home language; household size; government grant; repeated grade	Total screen time was not associated with working memory and cognitive flexibility.
Fairclough et al. (38) United Kingdom	(1) 359 (51%) (2) 11.5 ± 1.4	Accelerometer -Sedentary time	Spatial working memory test (CANTAB) -Working memory Multi-tasking test (CANTAB) -Inhibitory control Intra-Extra dimensional set shift task (CANTAB) -Cognitive flexibility	Age; sex; BMI z-score; IMD decile	Sedentary time was not associated with inhibitory control and cognitive flexibility.
Mora-Gonzalez et al. (25) Spain	(1) 79 (45%) (2) 10.2 ± 1.1	Accelerometer -Sedentary time	Delayed non-matched-to-sample task -Working memory	Sex; age; wave of participation; peak height velocity; BMI; parent education; IQ	Sedentary time was not associated with working memory.
Mora-Gonzalez et al. (35) Spain	(1) 100 (42%) (2) 10.1 ± 1.1	Accelerometer -Sedentary time	Stroop color-word test -Inhibitory control Zoo map task -Planning Design Fluency test and Trail making task -Cognitive flexibility	Sex; peak height velocity; BMI; wave of participation; parent education; IQ; MVPA	Sedentary time was not associated with inhibitory control, planning, and cognitive flexibility.
Mora-Gonzalez et al. (36) Spain	(1) 84 (44%) (2) 10.1±1.1	Accelerometer -Sedentary time	Flanker task -Inhibitory control	Sex; peak height velocity; BMI; parent education; IQ	Sedentary time was not associated with inhibitory control.
Ribner et al. (43) United States	(1) 807 (50%) (2) 5.7 ± 0.3	Parent-reported -TV viewing	Hearts and flowers task Dimensional change card sort Flanker task -Working memory -Cognitive flexibility -Inhibitory control	Age; sex; performance on Raven's progressive matrices	TV viewing was negatively associated with composite executive function.
Rosenqvist et al. (40) United States	(1) 381 (55%) (2) 8.4 ± 2.3	Parent-reported -TV viewing -General computer use	NEPSY-II -Inhibitory control	Age; sex; maternal education; other media variables	General computer use was not associated with inhibitory control. Negative association was observed between TV viewing and inhibitory control.
Syvaoja et al. (37) Finland	(1) 224 (57%) (2) 12.2 ± 0.6	Self-reported -TV viewing -Computer/video gaming -Computer use (other than playing) Accelerometer -Sedentary time	Spatial span test -Working memory Intra-Extra dimensional set shift task -Cognitive flexibility	Parental education; remedial education; gender; MVPA	Objective sedentary time, total screen time or TV viewing were not associated with any measures of executive functions. Computer/video game playing was negatively associated with working memory, but not with cognitive flexibility. Computer use was negatively associated with cognitive flexibility.

(Continued)

TABLE 1 | Continued

References, Country	Sample (1) *N* (% girls) (2) Age (years)	Measurement of sedentary behavior	Measurement of executive function	Adjusted covariates	Results
van der Niet et al. (27) Netherland	(1) 77 (55%) (2) 8.9 ± 1.0	Accelerometer -Sedentary time	Stroop color-word test -Inhibitory control Visual memory span test -Working memory Trail making task -Cognitive flexibility Tower of London task -Planning	Sex; age; SES	More time spent in sedentary behavior was associated with worse inhibitory control, but not to other aspects of executive functions.
Verburgh et al. (41) Netherland	(1) 168 (0%) (2) 8–12	Self-reported -TV viewing -General Computer use	Stop signal task -Inhibitory control Digit span task -Working memory Flanker Task -Executive attention	Age; BMI; IQ	General computer use was negatively associated with inhibitory control, but not with working memory and cognitive flexibility. TV viewing was not associated with any aspects of executive functions.
Xu et al. (42) United Kingdom and China	(1) 371 (47%) (2) 12.2 ± 1.0	Self-reported -General computer use	Stop signal task -Inhibitory control Figure matching task -Cognitive flexibility Spatial span task -Working memory Tower of Hanoi task -Planning	Age; general cognitive ability; family SES	General computer use was not associated with any aspects of executive functions.
Longitudinal studies					
Dubuc et al. (26) Canada	(1) 187 (62%) (2) baseline age: 13.1 ± 1.0 Follow-up age: 16.1	Self-reported -Total screen time -TV viewing -Computer/video gaming -Computer use (other than game playing)	Flanker task -Inhibitory control N-back task -Working memory	Age; pubertal status; socioeconomic status; ethnicity	In female students, changes in total screen time and time on video games were negatively associated with changes in N-back accuracy. In male students, changes in screen time was not associated with performance on Flanker task and N-back task.
López-Vicente et al. (24) Spain	(1) 307 (51%) (2) baseline age: 6 follow-up age: 14	Parent reported -TV viewing -Other sedentary behaviors	N-back task -Working memory	Age; sex; maternal education	TV viewing was not associated with working memory. Other sedentary behaviors were negatively associated with working memory.
O'Connor et al. (23) Spain	(1) 278 (49%) baseline age: 6, 9 (2) follow-up age: 14	Parent-reported -TV viewing	N-back task -Working memory	Age; sex; BMI; parental education; parental social class	TV viewing was not associated with working memory.
Wickel, (28) United States	(1) 699 (48%) (2) baseline age: 9 follow-up age: 15	Accelerometer -Sedentary time	Weinberger adjustment inventory -Inhibitory control Operation span task -Working memory Tower of London task -Planning	Ethnicity; change in PA; BMI z-score; SES	The increase in sedentary time from 9 to 15 years predicted higher inhibitory control, working memory, and planning.

BMI, body mass index; CANTAB, Cambridge Neuropsychological Test Automated Battery; IMD, Indices of Multiple Deprivation; IQ, intelligence quotient; PA, physical activity; MVPA, moderate to vigorous physical activity; SES: socioeconomic status.

Specifically, the systematic review concluded that screen time was either not associated with or had detrimental associations with cognitive function in young children (17).

The biological plausibility for the observed negative association of screen-based sedentary behavior with executive function is not clear. There might be several potential explanations. First, most digital screens are backlit and emit blue light wavelengths. It can suppress melatonin secretion to influence sleep quality (49, 50), which may, in turns, affect brain health (51, 52). Second, sedentary behavior may increase the risk of some aspects of mental problems, such as depression (14), which may negatively influence cognitive development (53). Third, recent neuroimaging studies have linked screen-based sedentary behavior with brain structure and integrity, which further supports a detrimental effect of screen-based sedentary behavior. A study indicated that

prolonged time on TV viewing was associated with lower gray matter volume in six brain regions in children (54). Increased screen-based media use was also associated with lower microstructural integrity of brain white matter in preschool-aged children (55).

Most of the included studies were of low to moderate quality. Of the 16 studies, four studies employed longitudinal study design, and only one study was rated as strong quality. Therefore, more studies with stronger design are warranted to further ascertain the effects of sedentary behavior on cognitive function in childhood. In addition, previous studies have suggested that physical activity and exercise were positively associated with executive function in children and adolescents (18, 56). Sedentary behavior may also correlate with physical activity considering the 24-h movement continuum. However, of the 16 studies, most studies did not consider physical activity as a potential covariate. The results may have been subjected to residual confounding. Future studies should consider physical activity as covariates or investigate the combined effects of sedentary behavior and physical activity.

Although the conflicting results exist, this systematic review provided preliminary evidence which supports a negative association between screen-based sedentary behavior and executive function in children and adolescents. Therefore, from the perspective of children's physical health and cognitive development, families, schools, and policymakers should consider interventions for reducing and limiting screen-based sedentary behavior in childhood.

To the best of our knowledge, this study is the first to systematically review the associations between sedentary behavior and executive function in children and adolescents. However, this study also has its limitations. First, a meta-analysis cannot be carried out due to the heterogeneities in study design and outcome measurements of the included studies. Second, all of the included studies were observational in design, and there was no intervention study. Therefore, the causal relationship between sedentary behavior and executive function cannot be inferred. Third, the searching language was limited to English, which increases the risk of omitting important studies published in other languages.

CONCLUSION

The study suggests that the associations between sedentary behavior and executive function are not conclusive in children and adolescents. However, time on screen-based sedentary behavior tends to be negatively associated with executive function.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Material**, further inquiries can be directed to the corresponding author/s.

AUTHOR CONTRIBUTIONS

SL and TH: conceptualization, writing, and original draft preparation. SL, TH, KZ, and JG: methodology and validation. SL and JG: formal analysis. SL, MS, and JG: resources. SL, JG, and MS: data curation. SL, KZ, and TH: writing—review and editing. TH: supervision and project administration. All authors have read and agreed to the published version of the manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2022.832845/full#supplementary-material

REFERENCES

1. Tremblay MS, Aubert S, Barnes JD, Saunders TJ, Carson V, Latimer-Cheung AE, et al. Sedentary behavior research network (SBRN) - terminology consensus project process and outcome. *Int J Behav Nutr Phys Act.* (2017) 14:75. doi: 10.1186/s12966-017-0525-8
2. van Sluijs EMF, Ekelund U, Crochemore-Silva I, Guthold R, Ha A, Lubans D, et al. Physical activity behaviours in adolescence: current evidence and opportunities for intervention. *Lancet.* (2021) 398:429–42. doi: 10.1016/s0140-6736(21)01259-9
3. Yang L, Cao C, Kantor ED, Nguyen LH, Zheng X, Park Y, et al. Trends in sedentary behavior among the us population, 2001–2016. *JAMA.* (2019) 321:1587–97. doi: 10.1001/jama.2019.3636
4. Cai YJ, Zhu XH, Wu XP. Overweight, obesity, and screen-time viewing among Chinese school-aged children: national prevalence estimates from the 2016 physical activity and fitness in china-the youth study. *J Sport Health Sci.* (2017) 6:404–9. doi: 10.1016/j.jshs.2017.09.002
5. DeMattia L, Lemont L, Meurer L. Do interventions to limit sedentary behaviours change behaviour and reduce childhood obesity? A critical review of the literature. *Obes Rev.* (2007) 8:69–81. doi: 10.1111/j.1467-789X.2006.00259.x
6. Hamilton MT, Healy GN, Dunstan DW, Zderic TW, Owen N. Too little exercise and too much sitting: inactivity physiology and the need for new recommendations on sedentary behavior. *Curr Cardiovasc Risk Rep.* (2008) 2:292–8. doi: 10.1007/s12170-008-0054-8
7. Hamilton MT, Hamilton DG, Zderic TW. Role of low energy expenditure and sitting in obesity, metabolic syndrome, type 2 diabetes, and cardiovascular disease. *Diabetes.* (2007) 56:2655–67. doi: 10.2337/db07-0882
8. Ford ES, Caspersen CJ. Sedentary behaviour and cardiovascular disease: a review of prospective studies. *Int J Epidemiol.* (2012) 41:1338–53. doi: 10.1093/ije/dys078
9. Lynch BM. Sedentary behavior and cancer: a systematic review of the literature and proposed biological mechanisms. *Cancer Epidemiol Biomarkers Prev.* (2010) 19:2691–709. doi: 10.1158/1055-9965.EPI-10-0815
10. Gray C, Gibbons R, Larouche R, Sandseter EB, Bienenstock A, Brussoni M, et al. What is the relationship between outdoor time and physical activity, sedentary behaviour, and physical fitness in children? A systematic review.

Int J Environ Res Public Health. (2015) 12:6455–74. doi: 10.3390/ijerph120606455
11. Must A, Tybor DJ. Physical activity and sedentary behavior: a review of longitudinal studies of weight and adiposity in youth. *Int J Obes.* (2005) 29:S84–96. doi: 10.1038/sj.ijo.0803064
12. Atkin AJ, Ekelund U, Moller NC, Froberg K, Sardinha LB, Andersen LB, et al. Sedentary time in children: influence of accelerometer processing on health relations. *Med Sci Sport Exer.* (2013) 45:1097–104. doi: 10.1249/MSS.0b013e318282190e
13. Suchert V, Hanewinkel R, Isensee B. Sedentary behavior and indicators of mental health in school-aged children and adolescents: a systematic review. *Prev Med.* (2015) 76:48–57. doi: 10.1016/j.ypmed.2015.03.026
14. Liu M, Wu L, Yao S. Dose-response association of screen time-based sedentary behaviour in children and adolescents and depression: a meta-analysis of observational studies. *Br J Sports Med.* (2016) 50:1252–8. doi: 10.1136/bjsports-2015-095084
15. Falck RS, Davis JC, Liu-Ambrose T. What is the association between sedentary behaviour and cognitive function? A systematic review. *Br J Sports Med.* (2017) 51:800–11. doi: 10.1136/bjsports-2015-095551
16. Olanrewaju O, Stockwell S, Stubbs B, Smith L. Sedentary behaviours, cognitive function, and possible mechanisms in older adults: a systematic review. *Aging Clin Exp Res.* (2020) 32:969–84. doi: 10.1007/s40520-019-01457-3
17. Carson V, Kuzik N, Hunter S, Wiebe SA, Spence JC, Friedman A, et al. Systematic review of sedentary behavior and cognitive development in early childhood. *Prev Med.* (2015) 78:115–22. doi: 10.1016/j.ypmed.2015.07.016
18. Xue Y, Yang YX, Huang T. Effects of chronic exercise interventions on executive function among children and adolescents: a systematic review with meta-analysis. *Br J Sports Med.* (2019) 53:1397–404. doi: 10.1136/bjsports-2018-099825
19. Donnelly JE, Hillman CH, Castelli D, Etnier JL, Lee S, Tomporowski P, et al. Physical activity, fitness, cognitive function, and academic achievement in children: a systematic review. *Med Sci Sport Exer.* (2016) 48:1223–4. doi: 10.1249/Mss.0000000000000966
20. Chaddock L, Pontifex MB, Hillman CH, Kramer AF. A review of the relation of aerobic fitness and physical activity to brain structure and function in children. *J Int Neuropsych Soc.* (2011) 17:975–85. doi: 10.1017/S1355617711000567
21. Diamond A. Executive functions. *Annu Rev Psychol.* (2013) 64:135–68. doi: 10.1146/annurev-psych-113011-143750
22. Magalhaes S, Carneiro L, Limpo T, Filipe M. Executive functions predict literacy and mathematics achievements: the unique contribution of cognitive flexibility in grades 2, 4, and 6. *Child Neuropsychol.* (2020) 26:934–52. doi: 10.1080/09297049.2020.1740188
23. O'Connor G, Pinero Casas M, Basagana X, Vicente ML, Davand P, Torrent M, et al. Television viewing duration during childhood and long- association with adolescent neuropsychological outcomes. *Prev Med Rep.* (2016) 4:447–52. doi: 10.1016/j.pmedr.2016.08.013
24. López-Vicente M, Garcia-Aymerich J, Torrent-Pallicer J, Forns J, Ibarluzea J, Lertxundi N, et al. Are early physical activity and sedentary behaviors related to working memory at 7 and 14 years of age? *J Pediatr.* (2017) 188:35–41. doi: 10.1016/j.jpeds.2017.05.079
25. Mora-Gonzalez J, Esteban-Cornejo I, Cadenas-Sanchez C, Migueles JH, Rodriguez-Ayllon M, Molina-García P, et al. Fitness, physical activity, working memory, and neuroelectric activity in children with overweight/obesity. *Scand J Med Sci Sports.* (2019) 29:1352–63. doi: 10.1111/sms.13456
26. Dubuc MM, Aubertin-Leheudre M, Karelis AD. Lifestyle habits predict academic performance in high school students: the adolescent student academic performance longitudinal study (ASAP). *Int J Environ Res Public Health.* (2019) 17:243. doi: 10.3390/ijerph17010243
27. van der Niet AG, Smith J, Scherder EJA, Oosterlaan J, Hartman E, Visscher C. Associations between daily physical activity and executive functioning in primary school-aged children. *J Sci Med Sport.* (2015) 18:673–7. doi: 10.1016/j.jsams.2014.09.006
28. Wickel EE. Sedentary time, physical activity, and executive function in a longitudinal study of youth. *J Phys Act Health.* (2017) 14:222–8. doi: 10.1123/jpah.2016-0200
29. Carson V, Hunter S, Kuzik N, Gray CE, Poitras VJ, Chaput JP, et al. Systematic review of sedentary behaviour and health indicators in school-aged children and youth: an update. *Appl Physiol Nutr Metab.* (2016) 41:240–65. doi: 10.1139/apnm-2015-0630
30. Moher D, Liberati A, Tetzlaff J, Altman DG, Group P. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *PLoS Med.* (2009) 6:e1000097. doi: 10.1371/journal.pmed.1000097
31. National Heart, Lung, and Blood Institute. *Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies*. National Heart, Lung, and Blood Institute. Available online at: https://www.nhlbi.nih.gov/health-topics/study-quality-assessment-tools (accessed May 5, 2014).
32. Sangsawang B, Wacharasin C, Sangsawang N. Interventions for the prevention of postpartum depression in adolescent mothers: a systematic review. *Arch Womens Ment Health.* (2019) 22:215–28. doi: 10.1007/s00737-018-0901-7
33. Musshafen LA, Tyrone RS, Abdelaziz A, Sims-Gomillia CE, Pongetti LS, Teng F, et al. Associations between sleep and academic performance in US adolescents: a systematic review and meta-analysis. *Sleep Med.* (2021) 83:71–82. doi: 10.1016/j.sleep.2021.04.015
34. Aadland KN, Moe VF, Aadland E, Anderssen SA, Resaland GK, Ommundsen Y. Relationships between physical activity, sedentary time, aerobic fitness, motor skills and executive function and academic performance in children. *Ment Health Phys Act.* (2017) 12:10–8. doi: 10.1016/j.mhpa.2017.01.001
35. Mora-Gonzalez J, Esteban-Cornejo I, Cadenas-Sanchez C, Migueles JH, Molina-Garcia P, Rodriguez-Ayllon M, et al. Physical fitness, physical activity, and the executive function in children with overweight and obesity. *J Pediatr.* (2019) 208:50–6. doi: 10.1016/j.jpeds.2018.12.028
36. Mora-Gonzalez J, Esteban-Cornejo I, Solis-Urra P, Migueles JH, Cadenas-Sanchez C, Molina-Garcia P, et al. Fitness, physical activity, sedentary time, inhibitory control, and neuroelectric activity in children with overweight or obesity: the activebrains project. *Psychophysiology.* (2020) 57:e1359. doi: 10.1111/psyp.13579
37. Syvaoja HJ, Tammelin TH, Ahonen T, Kankaanpaa A, Kantomaa MT. The associations of objectively measured physical activity and sedentary time with cognitive functions in school-aged children. *PLoS ONE.* (2014) 9:e103559. doi: 10.1371/journal.pone.0103559
38. Fairclough SJ, Tyler R, Dainty JR, Dumuid D, Richardson C, Shepstone L, et al. Cross-sectional associations between 24-hour activity behaviours and mental health indicators in children and adolescents: a compositional data analysis. *J Sports Sci.* (2021) 39:1–13. doi: 10.1080/02640414.2021.1890351
39. Chetty-Mhlanga S, Fuhrimann S, Eeftens M, Basera W, Hartinger S, Dalvie MA, et al. Different aspects of electronic media use, symptoms and neurocognitive outcomes of children and adolescents in the rural western cape region of south africa. *Environ Res.* (2020) 184:e109315. doi: 10.1016/j.envres.2020.109315
40. Rosenqvist J, Lahti-Nuuttila P, Holdnack J, Kemp SL, Laasonen M. Relationship of tv watching, computer use, and reading to children's neurocognitive functions. *J Appl Dev Psychol.* (2016) 46:11–21. doi: 10.1016/j.appdev.2016.04.006
41. Verburgh L, Scherder EJ, Van Lange PA, Oosterlaan J. Do elite and amateur soccer players outperform non-athletes on neurocognitive functioning? A study among 8-12 year old children. *PLoS ONE.* (2016) 11:e0165741. doi: 10.1371/journal.pone.0165741
42. Xu C, Ellefson MR, Ng FF, Wang Q, Hughes C. An east-west contrast in executive function: measurement invariance of computerized tasks in school-aged children and adolescents. *J Exp Child Psychol.* (2020) 199:e104929. doi: 10.1016/j.jecp.2020.104929
43. Ribner A, Fitzpatrick C, Blair C. Family socioeconomic status moderates associations between television viewing and school readiness skills. *J Dev Behav Pediatr.* (2017) 38:233–9. doi: 10.1097/dbp.0000000000000425
44. Rojer AGM, Ramsey KA, Amaral Gomes ES, D'Andrea L, Chen C, Szoeke C, et al. Objectively assessed physical activity and sedentary behavior and global cognitive function in older adults: a systematic review. *Mech Ageing Dev.* (2021) 198:e111524. doi: 10.1016/j.mad.2021.111524
45. Sweetser P, Johnson D, Ozdowska A, Wyeth P. Active versus passive screen time for young children. *Aust J Early Childhood.* (2012) 37:94–8. doi: 10.1177/183693911203700413
46. Horowitz-Kraus T, Hutton JS. Brain connectivity in children is increased by the time they spend reading books and decreased by the length of exposure to screen-based media. *Acta Paediatr.* (2018) 107:685–93. doi: 10.1111/apa.14176

47. Barnett TA, Kelly AS, Young DR, Perry CK, Pratt CA, Edwards NM, et al. Sedentary behaviors in today's youth: approaches to the prevention and management of childhood obesity: a scientific statement from the american heart association. *Circulation*. (2018) 138:E142–E59. doi: 10.1161/Cir.0000000000000591
48. Felez-Nobrega M, Bort-Roig J, Briones L, Sanchez-Niubo A, Koyanagi A, Puigoriol E, et al. Self-reported and Activpal$^{(TM)}$-monitored physical activity and sedentary behaviour in college students: not all sitting behaviours are linked to perceived stress and anxiety. *J Sports Sci.* (2020) 38:1566–74. doi: 10.1080/02640414.2020.1748359
49. Brainard GC, Hanifin JP, Greeson JM, Byrne B, Glickman G, Gerner E, et al. Action spectrum for melatonin regulation in humans: evidence for a novel circadian photoreceptor. *J Neurosci.* (2001) 21:6405–12. doi: 10.1162/08989290152541494
50. Thapan K, Arendt J, Skene DJ. An action spectrum for melatonin suppression: evidence for a novel non-rod, non-cone photoreceptor system in humans. *J Physiol.* (2001) 535:261–7. doi: 10.1111/j.1469-7793.2001.t01-1-00261.x
51. Cajochen C, Frey S, Anders D, Spati J, Bues M, Pross A, et al. Evening exposure to a light-emitting diodes (led)-backlit computer screen affects circadian physiology and cognitive performance. *J Appl Physiol.* (2011) 110:1432–8. doi: 10.1152/japplphysiol.00165.2011
52. Aribisala BS, Riha RL, Hernandez MV, Maniega SM, Cox S, Radakovic R, et al. Sleep and brain morphological changes in the eighth decade of life. *Sleep Med.* (2020) 65:152–8. doi: 10.1016/j.sleep.2019.07.015
53. Wagner S, Muller C, Helmreich I, Huss M, Tadic A. A meta-analysis of cognitive functions in children and adolescents with major depressive disorder. *Eur Child Adolesc Psychiatry.* (2015) 24:5–19. doi: 10.1007/s00787-014-0559-2
54. Zavala-Crichton JP, Esteban-Cornejo I, Solis-Urra P, Mora-Gonzalez J, Cadenas-Sanchez C, Rodriguez-Ayllon M, et al. Association of sedentary behavior with brain structure and intelligence in children with overweight or obesity: the activebrains project. *J Clin Med.* (2020) 9:1101. doi: 10.3390/jcm9041101
55. Hutton JS, Dudley J, Horowitz-Kraus T, DeWitt TG. Holland SK. Associations between screen-based media use and brain white matter integrity in preschool-aged children. *JAMA Pediatr.* (2020) 174:509. doi: 10.1001/jamapediatrics.2020.0480
56. Liu SJ Yu Q, Li ZM, Cunha PM, Zhang Y, Kong ZW, et al. Effects of acute and chronic exercises on executive function in children and adolescents: a systemic review and meta-analysis. *Front Psychol.* (2020) 11:3482. doi: 10.3389/fpsyg.2020.554915

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