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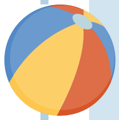
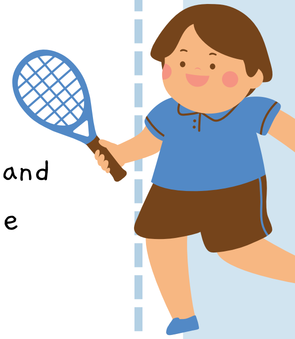
LETTER FROM THE FACILITATORS



We are excited to share the third issue of the CHILD journal that continues to highlight dedication to scientific inquiry among students in the Child Health Specialization (CHS) of the Honours Health Sciences Program at McMaster University.



Through this co-created initiative, our students ask meaningful questions about child health, critically appraise evidence-based literature, find gaps in knowledge, gather and synthesize evidence, and concisely communicate their findings. By engaging CHS alumni in the peer review process, our students are learning what it means to contribute to the world of child health in a rapidly changing world.

We hope this exploration of topics highlights that investigating child health requires multifaceted and diverse perspectives. Whether we are exploring inequities at a systems level or analyzing family and local supports, each part contributes to a more holistic understanding of the well-being of children and their families.

In publishing this issue, we continue to celebrate the power of trusting students to engage with real-world questions as academic scholars. We hope this journal models student-led, collaborative learning. As always, we invite educators and researchers alike to consider how student voices can shape the future of health scholarship.

Margaret Secord, Nevart Terzian, Alessia Greco,
Celina Antony, & Elyse Rosa
(BHSc Child Health Specialization Facilitators)

INVESTIGATING THE RELATIONSHIP BETWEEN TOURETTE SYNDROME AND SLEEP QUALITY IN CHILDREN: A REVIEW OF PATHOPHYSIOLOGY AND IMPLICATIONS FOR QUALITY OF LIFE

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ABSTRACT

Current research suggests that children with Tourette's Syndrome (TS) experience greater sleep disturbances and lower sleep quality than children without TS. Sleep plays a fundamental role in a child's mental and social-emotional development. It is hypothesized that dysfunctions in neurotransmitter activity, cortico-striato-thalamo-cortical (CSTC) circuits, and the basal ganglia are involved in the pathogenesis of tics that may influence sleep quality. Furthermore, neurotransmitters, such as norepinephrine and dopamine, are hypothesized to contribute to the pathophysiology of TS and its associated sleep disturbances. This review explores the pathophysiology of TS, tic severity, its influences on sleep quality, and the effects on quality of life (QoL) in children with the disorder. Research highlights the impact of disturbed sleep on health outcomes, demonstrating the importance of further research to address these concerns and improve QoL in children with TS. However, there is limited research on the specific mechanisms that influence sleep quality in children with TS.

INTRODUCTION

Tourette Syndrome

TS is a childhood-onset neurodevelopmental disorder characterized by tics, which are sudden, repetitive, and involuntary motor or vocal movements [1]. Tics are categorized as simple and complex. Simple tics are brief and repetitive movements or sounds affecting a limited number of muscle groups, while complex tics involve multiple muscle groups, and can involve a sequence of movements or vocalizations [1]. TS typically emerges between the ages 5-10, and tics may be more pronounced in childhood, often improving in adolescence and early adulthood [2,3]. A systematic review determined that the global prevalence of TS is 0.5% [3]. The CDC reports that 1 out of 162 children (0.6%) have TS, with 44% of children diagnosed with TS in 2016-2017 having moderate to severe tics in the United States [4].

Diagnosis

The current standard of diagnostic criteria for TS and other tic disorders, follow the criteria outlined in the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5-TR), developed by the American Psychiatric Association [5,6]. Other tic disorders described by the DSM-5-TR are Persistent Motor or Vocal Tic Disorder and Provisional Tic Disorder, whose diagnostic criteria vary from TS in terms of the duration of tics (more than 1 year for TS and Persistent Motor or Vocal Tic Disorder, versus less than 1-year Provisional Tic Disorder). Additionally, TS varies in that diagnosis requires the presence of both motor and vocal tics, whereas other tic disorders diagnose only require motor or vocal tics [5].

TS diagnosis is based on the presence of both motor and vocal tics that occur several times a day (concurrently or non-concurrently) or most days [5]. Tic frequency is not specifically quantified; the DSM-5 states that tics need to occur frequently throughout the day, typically in episodes, daily or intermittently for a duration of at least one year [5]. The tics must have emerged prior to 18 years of age, and tics must not result from medications used to treat another condition [2,5]. TS diagnoses are made by healthcare professionals who assess a child's symptoms and medical history [1,5].

Comorbidities

TS is often comorbid with other conditions, with the most prevalent being attention-deficit hyperactivity disorder (ADHD), anxiety, obsessive-compulsive disorder (OCD), depression, and insomnia [7-9].

Although there is no cure for TS, treatments and therapies can help children manage their tics, and in many cases, improve symptoms over time, particularly during adolescence and early adulthood [10-12]. Pharmacological treatments include Clonidine and Haloperidol, pimozide, and aripiprazole [12]. An emerging therapy that uses habit reversal training (HRT) is Comprehensive Behaviour Intervention of Tics (CBIT) [10]. While treating comorbidities does not eliminate tics, it can also lead to tic reduction, and improve children's relationships with family, friends in school and at home [10]. Individuals will need different treatment plans based on the comorbidities children with TS are diagnosed with [10].

Quality of Life in Tourette Syndrome

QoL refers to an individual's well-being, which encompasses physical health, mental and emotional state, social relationships, and functional abilities [13]. Research indicates that children with TS often experience lower QoL compared to their peers without TS [14-16].

Youth with TS consistently reported the lowest QoL scores compared to other groups, including those with ADHD and mobility limitations, as well as youth without chronic conditions [17]. Another study by Storch et al. indicates that children and youth with more severe tics or comorbid conditions tend to feel less welcome in social spaces, spend less time with friends, and experience more familial conflicts. Children with TS may experience ridicule or social exclusion in school settings due to an inability to complete tasks due to their tics, which may lead to isolation and a hesitance to socialize [14]. This study highlights the challenges faced by youth with TS, particularly in social relationships and academic settings, emphasizing the need for tailored support to enhance their overall well-being [14].

TS can have a significant impact on various aspects of a child's QoL, including sleep [18,19]. Research has shown that sleep is negatively affected in patients with TS, particularly children. Children with TS experience decreased total sleep time, lower sleep efficiency, and an elevated arousal index [18,20,21]. A case-control study published in 2017 reports an incidence rate of sleep disorders of 7.24% in children with TS, compared to 3.53% in children without TS [18]. Sleep disturbances and disorders are highly prevalent in children with TS and are categorized as part of the DSM-5 criteria for sleep disorders, associated with frequent awakenings, arousals, or problems with initiating and returning to sleep [7]. Another study states that 65% of 123 children and young adults, all of whom had TS, experienced sleep disturbances, which ranged from various types including increased sleep disruption and decreased sleep efficiency [22]. Sleep disturbances in children can result in worsened QoL outcomes including impairment in daytime functioning, emotional and behavioral difficulties, and compromised cognitive and academic performance [18, 23-25]. Effective management of sleep problems has been reported to improve tic control and positively impact quality of life in children with TS [7].

Understanding the impact of TS on QoL and sleep quality in children is crucial for providing comprehensive care and interventions that address their holistic well-being and improve their overall outcomes. In this review, we discuss the current literature surrounding TS and the biological mechanisms that contribute to tics. We aim to investigate the relationship between TS and sleep quality in children due to the onset of tics during childhood and youth.

BACKGROUND ON SLEEP

Sleep quality is defined as the holistic assessment of the wellness of an individual's sleep [26-29]. It has four main attributes: sleep latency, duration, efficiency, and wake-after-sleep onset. Multiple factors can influence sleep quality including BMI, circadian rhythm, stress, anxiety, and various neurological disorders, such as TS [26,28]. Typical sleep patterns follow a 'standard' circadian rhythm–the physical, mental, and behavioural changes an organism experiences over a 24-hour cycle [29-31]. Further, light, food intake, social environment, physical activity, etc. can impact this cycle [29-31]. In humans, almost every organ and tissue has its own circadian rhythm which collectively functions in the day and night cycles of life [29,31]. The circadian rhythm is regulated by proteins encoded by many genes that coordinate wakefulness and sleepiness upon activation [29-31]. Variants or malfunctions in these genes can cause abnormalities in an individual's circadian rhythm [29-31]. Additionally, this cycle is regulated by the brain and is composed of a large group of nerve cells that form the suprachiasmatic nucleus (SCN), a structure located in the hypothalamus that controls the production of melatonin based on the amount of light received by the eyes. Melatonin levels, controlled by the pineal gland, typically rise in response to decreased exposure of light to the eyes [31,32]. This elevation in melatonin acts as the onset for the initiation of the sleep phase in the body's circadian rhythm [31,32]. A large majority of sleep disorders are said to be caused by a malfunction in the pathways involved with these proteins and/or nerve networks. Therefore, understanding the role of the body's circadian rhythm, and its regulation processes, is crucial for developing strategies to improve sleep quality [31,32].

Sleep plays a fundamental role in a child's mental and social-emotional development [19,26,33]. The literature shows that poor sleep quality can result in behavioral and emotional consequences such as irritability, fatigue, slowed responses, and daytime dysfunction [29,33]. Negative physical health outcomes can also result from poor sleep quality [26-29,33]. Multiple studies have investigated sleep disorders using the SATED scale; 1) satisfaction/quality, 2) alertness/sleepiness/napping, 3) timing, 4) efficiency/latency, 5) duration [28]. It has been concluded that an abnormality in any of these areas can be associated with many health outcomes, such as mortality, diabetes, heart disease, and/or impaired neurobehavioral performance [29,33].

A sleep disorder is a condition that disturbs typical sleep patterns [19,33]. Sleep disorders are considered a comorbidity of TS, and the sleep quality of many children with TS can be negatively impacted as a result [19,33]. For instance, difficulties such as fragmented sleep and sleep interfering behaviours may be experienced. Some examples of sleep disorder comorbidities in TS include insomnia (difficulty falling/staying asleep), excessive daytime sleepiness, disorders of arousal (e.g., sleepwalking, sleep talking, sleep terrors, enuresis), persistence of tics during sleep, and the presence of periodic limb movements during sleep [19,33]. Moreover, the increased prevalence of sleep disorders persists after controlling for other known risk factors, such as obesity and co-occurring ADHD, strengthening the correlation between TS and sleep disorders [33]. This comorbidity is important to recognize as sleep disorders may lead to poor associated health outcomes (diabetes, heart disease, impaired neurobehavioral performance, etc.), and impact QoL of children with TS [19,29,33]. Therefore, the high prevalence of sleep disorder comorbidity highlights a need to understand the causes of the correlation between TS and poor sleep quality. By investigating the mechanisms underlying sleep disturbances, the development of more effective strategies for sleep-related issues and the enhancement of overall health outcomes in children with TS may be promoted.

Pathophysiology, Treatment, and Connection to Sleep

TS is a childhood-onset neurodevelopmental disorder characterized by tics, which are sudden, repetitive, and involuntary motor or vocal movements [1]. Tics are categorized as simple and complex. Simple tics are brief and repetitive movements or sounds affecting a limited number of muscle groups, while complex tics involve multiple muscle groups, and can involve a sequence of movements or vocalizations [1]. TS typically emerges between the ages 5-10, and tics may be more pronounced in childhood, often improving in adolescence and early

Neurotransmitter Systems and Activity on REM and NREM Sleep:

Changes within various neurotransmitter systems (including dopaminergic, serotonergic, and noradrenergic systems) are associated with changes in the brain that influence polysomnographic variables that are used to identify sleep abnormalities during rapid eye movement (REM) and non-rapid eye movement (NREM) sleep [34,35]. Polysomnography tests are considered the gold standard to measure sleep architecture, as they reliably document brain activation patterns and other important homeostatic indicators of sleep quality [34]. While REM sleep consists of vivid dreaming, increased brain and heart activity, and rapid eye movement, NREM sleep consists of three stages, where stages one and two involve lighter sleep and stage three involves deep and restorative sleep [34]. Polysomnographic studies in children with TS have found that tics can occur throughout any stage of REM and NREM sleep, which may lead to sleep disruptions and poor sleep quality [34].

Dysfunctions in Neurotransmitter Activity, Basal Ganglia, and CSTC Circuits:

Evidence suggests that excitatory dopamine from the CSTC circuit is linked to the pathogenesis of TS, as excess dopaminergic activity may contribute to the manifestation of tics [36]. Furthermore, dysfunction in the dopaminergic pathways could alter sleep-wake regulation and potentially interfere with one’s ability to fall or stay asleep [34]. The dopaminergic ventral tegmental area and nucleus accumbens are dopaminergic brain structures that can modulate the mesopontine brainstem nuclei [7,36]. The mesopontine brainstem nuclei are involved in regulating wakefulness, and the transition between REM and NREM sleep [34,36]. When dopaminergic signalling is dysregulated, its modulating effects on the mesopontine brainstem nuclei can result in poor sleep quality [34]. Moreover, research suggests that unaltered serotonergic firing is associated with sleep maintenance properties, emphasizing the importance of serotonin in maintaining healthy sleep patterns [34,36]. Studies have found that children with TS or sleep disorders may exhibit altered serotonergic firing patterns that lead to decreased serotonergic activity that reduces sleep quality [21,34]. Additionally, it is hypothesized that increased noradrenergic activity is associated with the pathogenesis of tics and is considered a sign of hyperarousal that may also contribute to sleep disturbances by promoting wakefulness [21,34]. However, the specific mechanisms of how serotonin and noradrenaline influence sleep quality in TS is still being researched, as there are conflicting findings on how their activity may be altered in children with TS.

Neuroimaging studies have indicated that dysfunction in the basal ganglia and CSTC circuits seen in children with TS may cause abnormal transmission of neural signals to premotor cortices through thalamocortical activation, prompting involuntary sensory urges and the occurrence of tics [34]. It is postulated that during consciousness, the prefrontal cortex compensates by modulating misguided basal ganglia signals to suppress the occurrence of tics [34]. However, during REM and NREM sleep, the activation of the prefrontal cortex is diminished, which can result in aberrant activity in the basal ganglia and disrupt the normal functioning of CSTC circuits [34]. Thus, abnormal basal ganglia signalling and impaired CSTC circuits may lead to increased tic expression and reduced quality of sleep. Given that there are limited neuroimaging studies available that investigate the interplay of the basal ganglia and CSTC circuits on tic expression during sleep, further research is required to confirm this relationship.

Application of Pathophysiology in TS and Sleep Disorders:

To provide an application of the connection between the pathophysiology of TS and its relation to sleep disorders, this section will investigate the use of Clonidine, an alpha2 agonist that is used to treat both tic disorders and sleep disorders [37]. While Clonidine's intended use per Health Canada is primarily for the treatment of hypertension, it is still commonly utilized for children and adolescents in the management of tic symptoms as an off-label intervention [37]. Clonidine functions primarily by affecting the activity of norepinephrine, a neurotransmitter part of the sympathetic nervous system [38]. During sleep, norepinephrine originates in the upper brainstem and plays a role in our brain's arousal system that leads to wakefulness, according to the Institute of Medicine (US) Committee on Sleep Medicine and Research [39]. This norepinephrine release is associated specifically with a secondary pathway that involves flow to the hypothalamus, where it receives inputs from nerve cells that then traverse the basal forebrain, where they pick up additional inputs from cells containing acetylcholine and gamma-aminobutyric acid. Ultimately, all these inputs enter the cerebral cortex, where they diffusely activate the nerve cells and prepare them for the interpretation and analysis of incoming sensory information [39]. A review on current treatments for TS highlights that Clonidine acts on the α2 adrenergic receptors located in the locus coeruleus within the brain stem. This limits the release and turnover of norepinephrine, resulting in a reduced sympathetic outflow from the CNS and lessening the number of tics [40]. Clonidine has also been shown to impact other neurotransmitters, with dopamine being a significant player that can be affected. In a study conducted by Yoshida et al., Clonidine was shown to significantly decrease the concentration of dopamine in the nucleus accumbens of rats [41]. This decreased release may lead to a dysfunction in the role of dopamine, which is concurrent with literature on dopamine's impact on a circadian rhythm [42]. The pathophysiology of sleep disorders in children with TS remains very complex. Additional literature also points to dopaminergic dysfunction as a potential cause [7]. This has been supplemented with a more epigenetic based approach to understanding the pathophysiology of the impact of TS on sleep, with various imaging techniques such as positron emission tomography or magnetic resonance spectroscopy utilized to implicate other neurotransmitters [7].

IMPLICATIONS ON CHILD DEVELOPMENT

Quality of Life (QoL)

QoL is suggested to be negatively affected in children with TS.

In addition to the manifestation of tics in TS, it is known that additional emotional and behavioural difficulties can arise, including issues relating to impulse control as well as OCD and ADHD comorbidities [43-47]. Research suggests that poorer QoL was associated with increased tic symptom severity, ADHD diagnosis, and obsessive-compulsive behaviours [48]. In a study that compared the QoL of children aged 10 to 17 years with TS, epilepsy, and those without diagnosed medical conditions, researchers found significant differences in various QoL contextual items [49]. Specifically, the findings indicated that children with TS experienced a reduced QoL compared to those in the healthy control group. Using the Youth QoL instrument-research version scale, children with TS demonstrated hindered involvement in social activities due to TS, less involvement in house chores, reduced communication with adults about important personal issues, and lower overall levels of participation, a fundamental principle in QoL outcomes. There were also increases in reported externalizing and internalizing behavioural problems including lower self-esteem and increased impulsivity. TS patients reported greater tic severity exhibited significantly lower scores than controls for the QoL contextual items and relationship domains, signifying a positive correlation between tic severity and reduced QoL. Another study that included qualitative measures of interviews yielded similar results, coding qualitative themes across the TS group such as "Tourette syndrome can be distressing and disabling," "needing to control tics," and "struggling to fit into society's expectations of normal behaviour" [48]. Per these findings, it can be understood that tic severity plays a large role in the QoL, and emotional/behavioural challenges faced by children with TS.

Daytime Functioning

Another study examined the relationship between sleep, daytime functioning (defined by competencies at school, activities, and social interactions), and tic severity in children with TS [24]. The study utilized actigraphy and parent reported QoL measures to assess sleep quality and its effects on QoL. The aim was to assess objective sleep parameters and potential links between sleep and daytime functioning in children with and without TS. The primary sleep measure used in the study, actigraphy, is a common non-invasive method to objectively assess sleep behaviour by utilizing a wrist-worn accelerometer to record continuous movement and activity [50]. Its non-invasive nature and ability to provide long-term monitoring make actigraphy particularly suitable for pediatric sleep research. This is especially beneficial for children who might find other monitoring methods uncomfortable or disruptive, or for those who require ongoing tracking of sleep patterns, such as children with TS or other conditions affecting sleep [51]. Via parent-reported measures, it was shown that children with TS displayed significantly greater behavioural manifestations of executive difficulties including working memory, inhibition, and attention, in comparison to controls. A measure called the Flanker task was used to assess inhibitory control in the children. There were associations between the previous night's actual sleep time and next-day accuracy on this task, wherein children with shorter sleep durations and more disturbances the previous night made more errors across its incongruent trials. This finding is consistent with prior research demonstrating the relationship between nightly sleep quality and inhibition tasks and overall functioning, emphasizing the importance of sleep in the cognitive functioning of children [52-54]. For that reason, it is crucial to further investigate sleep quality in children with TS to improve daytime functioning and tic severity through sleep-targeted interventions in terms of healthcare services, educational plans, and family support systems.

DISCUSSION

Emerging evidence suggests that children with TS are at a higher risk for sleep disruptions and sleep disorders [7,18,22,23]. This can be explained by the presence of tics during sleep and challenges with sleep onset in children. Several studies suggest that altered dopaminergic, serotonergic, and noradrenergic systems are associated with sleep abnormalities; however, the specific mechanisms of these neurotransmitters in influencing sleep disturbances in children with TS are not well-understood [34,35]. Furthermore, it is hypothesized that the basal ganglia and CSTC circuits play a major role in the occurrence of tics during rapid REM and NREM sleep [34]. Considering the significance of sleep for children's daily functioning, it is essential to assess sleep in the clinical management of TS. Implementing interventions for affected patients that target sleep can potentially alleviate symptoms and improve overall health outcomes. The studies reviewed in this article demonstrate the feasibility of actigraphy as a minimally invasive objective measure in assessing sleep onset in children, which can be leveraged to a greater extent to further investigate the impact of tics on sleep quality in TS [50,51]. Tic severity typically peaks during childhood, and it is therefore important that studies investigating the impact of TS involve pediatric populations wherein symptoms are most severe, and management can prove most crucial to child development outcomes [2,3].

CONCLUSION

This review explores the neurobiology of tic severity in TS and its influences on sleep quality and QoL in children. Considering the gaps in existing literature, the studies used in this review highlight that the pathophysiology of tics observed in TS may lead to poor sleep quality and negatively influence QoL. However, further research is needed to understand the variables to enhance treatment strategies, improve health outcomes, and foster educational advancements to enhance the QoL for children with TS.

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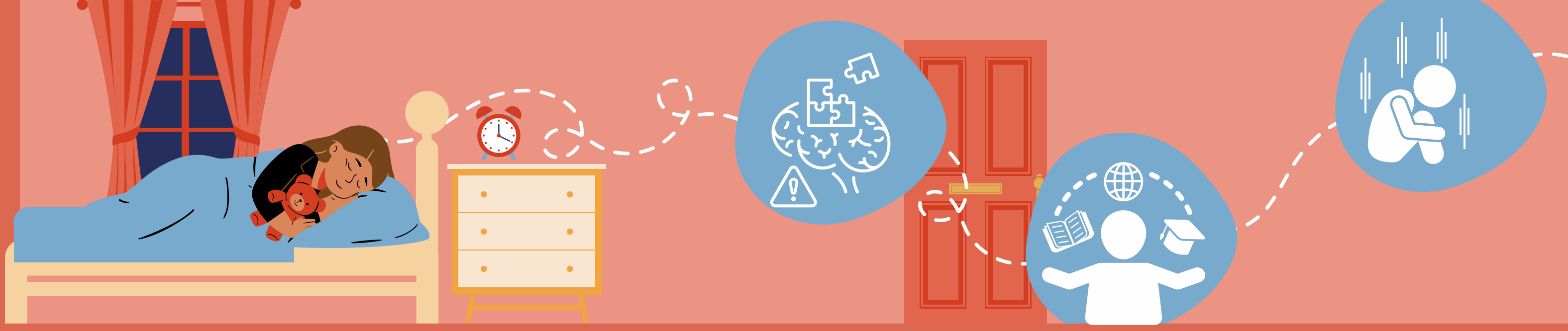

EXPLORING THE SEVERITY OF SLEEP-RELATED COGNITIVE IMPAIRMENTS AMONG CHILDREN WITH FAMILIAL MEDITERRANEAN FEVER: A NARRATIVE REVIEW

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ABSTRACT

Background: Familial Mediterranean Fever (FMF) is the most common autoinflammatory disorder across both Canada and the United States. Children with FMF are thought to experience poorer sleep quality than their healthy counterparts. Sleep is a vital component of overall well-being and a strong predictor of healthy cognitive development. As such, the importance of understanding the cognitive impairments that result from poor sleep quality among this group must not be overlooked.

Objective: This narrative review aims to synthesize relevant research findings to determine the extent to which cognitive development is influenced by poor sleep quality in children with FMF. This information can inform future intervention strategies to improve sleep quality and cognitive developmental outcomes in children ages 8-15.

Results: Attacks of FMF are highly related to negative sleep experiences. Inadequate sleep deprives the childhood brain from undergoing necessary structural and functional developments. Sleep deprivation is thought to inhibit neuroplasticity, hinder academic outcomes, and give rise to a range of comorbidities. Unresolved, this issue can give rise to a myriad of developmental problems via disruptions in cognitive functioning, behaviour, and emotional well-being.

Conclusions: This review suggests that children with FMF experience a wide range of sleep disturbances. To protect the cognitive health of children with this disorder, clinical assessment and support through behaviour or pharmacological therapy is crucial. By recognizing the relevance of sleep quality on cognitive development, healthcare providers and health policy experts can best meet the healthcare needs of children with FMF.

INTRODUCTION

Familial Mediterranean Fever (FMF) is a hereditary autoinflammatory disorder characterized by recurrent episodes of fever and serositis in the chest, abdomen and joints [1]. FMF is a recurrent fever syndrome that primarily affects the paediatric population with approximately 65 and 90 percent of patients experiencing their first attack before the ages of 10 and 20, respectively [2]. It is the most common monogenic autoinflammatory disorder in Canada and the United States, with children of Turkish, Armenian, Middle Eastern, North African Jewish, and Arab descent being the most frequently affected ethnic groups [3].

FMF attacks within the paediatric population present with a range of symptoms, including fever, abdominal pain, chest pain, joint pain, and erysipelas-like skin lesions. These episodes typically last between one and three days and often resolve spontaneously, with patients remaining asymptomatic between attacks [2]. FMF is most frequently attributed to a mutation in the Mediterranean fever (MEFV) gene and is typically inherited in an autosomal recessive fashion [4]. A mutation in this gene leads to a misfolded pyrin protein and dysregulated inflammatory response, resulting in the characteristic episodes of fever and inflammation seen in FMF patients [4]. The triggers of FMF attacks remain elusive; however, research has shown notable positive associations between the likelihood of an FMF attack and various factors such as infection, physical or emotional stress, and menstruation.

Quality sleep is vital to a child's proper functioning and well-being [5]. Greater sleep duration and sleep efficiency are associated with high sleep quality, while a greater number of sleep disturbances, and sleep-onset delay are associated with poorer sleep quality [6]. Sleep quality impacts all domains of a child's health, including socioemotional and cognitive development [7]. Sleep quality is also associated with several behavioural outcomes in children, including temperament, emotional regulation and motivation [8], and impacts several cognitive functions, including memory consolidation, concentration and problem-solving [9]. Healthy sleep habits are essential for promoting optimal cognitive development in children. Consistent bedtimes, a calming bedtime routine, and a comfortable sleep environment are key components of healthy sleep habits [10].

A study conducted by Makay et al. suggests that children with FMF may be at greater risk of experiencing poor sleep quality compared to children without FMF [11]. Pain during FMF episodes, anxiety about future attacks, and disruptions in daily routines due to FMF symptoms could potentially contribute to poorer sleep quality, posing a threat to a child's normal development [11].

Understanding the impact of FMF on children's sleep quality and subsequent cognitive development is crucial for developing targeted interventions to mitigate potential adverse outcomes. This review aims to explore the extent to which sleep quality impacts cognitive development in children with FMF aged 8-15. Adolescence is typically marked by the beginning of puberty, which begins between the ages of 8 and 14 years for girls, and 9 and 15 years for boys. Puberty is characterized by significant biological and social transformations which impact both health and behaviour, including sleep patterns [12]. By understanding the intricate interplay between FMF, sleep quality, and cognitive development in children of this age, this narrative review aims to inform interventions that can optimize sleep health and enhance cognitive functioning in children. Addressing sleep-related issues in children with FMF holds the potential to improve overall quality of life, academic performance, and long-term health outcomes [11].

METHODS

A targeted search of the databases PubMed, Web of Science, and OVID was conducted. Key search terms included "Familial Mediterranean Fever", "FMF", "sleep disturbances", and "cognitive development", among others. Boolean operators were strategically utilised to ensure that all relevant studies were presented. While our primary focus was on literature significant to Canada and the United States, we prioritized studies from all countries to capture a broader understanding of the topic. [LA1] The inclusion of primary studies and articles that focused on a paediatric population (ages 8-15), were emphasized.

RESULTS

Impacts on Sleep

Mechanisms of Impact:

Children with FMF are known to experience more sleep disturbances than their healthy counterparts [11]. While this relationship has been proposed within existing literature, there is a paucity of information about the exact mechanism causing this effect. The leading hypothesis suggests that pain resulting from symptoms caused by this disorder could contribute to sleep disruptions. FMF is in part characterised by the recurrent presentation of serositis, which can induce chest and abdominal pain [11,13]. Other common presentations include myalgia—muscle pain—in response to physical exertion [14]. Recurrent pain has long been associated with sleep impairments among children and adolescents and may help to explain the common sleep disturbances faced within this group [15]. Pain experienced by FMF patients could itself be related to sleep quality, as sleep disturbances have been known to exacerbate the perception of pain [16]. However, more research is warranted to investigate this proposed model.

Sleep Disruptions:

Sleep quality is influenced by an amalgamation of factors within a child's life. Familial lifestyle choices, sociodemographics, environmental factors and significant life events all contribute to sleep quality in important ways [17]. Hence, the irregular presentation of FMF attacks may unexpectedly force changes in sleep habits among those affected [17]. Previous reports have suggested that FMF impacts sleep quality in several important ways [18]. Increased bedtime resistance, sleep-onset delay, sleep anxiety, night wakings, parasomnias, and sleep-disordered breathing have all been noted among children with FMF [11,19]. These effects are worsened in children with FMF who experience a greater number of attacks [11]. Interestingly, sleep is also worsened by the recency of a child's last attack, with patients who experienced an attack in the last month demonstrating greater sleep impairments [19].

Psychological Associations:

Issues with sleep quality can pose a considerable mental burden on children with FMF. Poor sleep-related outcomes are known to contribute to a wide array of behavioural and mood problems among adolescents [20]. Adolescent patients with FMF who experience sleep-related problems scored significantly higher on the Revised Child Anxiety and Depression Scale - Child Version than other children with FMF [19]. Behaviourally, children with sleep deprivation demonstrate increased emotional problems, difficult behaviours, and poorer temperament, among a myriad of cognitive functioning issues [21]. In particular, increased bedtime resistance among children with FMF demonstrates an existing behavioural problem that can adversely affect quality of life [11,19,22]. To prevent or address these issues, sleep-related fatigue among children with FMF should be closely evaluated and addressed by healthcare professionals [17].

Managing Symptoms:

Unfortunately, there does not currently exist a "gold standard tool" for evaluating the severity of fatigue in children with FMF [18]. There does, however, exist a treatment which has shown efficacy in helping to prevent it. It is suggested that Colchicine, the first-line therapeutic defence for FMF, helps to reduce the symptoms of FMF, facilitating improved sleep outcomes in child and adolescent patients [23,24]. Its therapeutic success serves as an important reminder that healthcare practitioners should be continually assessing sleep problems among paediatric FMF patients. Ultimately, the management and control of this disorder's symptoms are needed to address the impact of FMF on sleep quality [17]. Otherwise, children with FMF may display behavioural and neurocognitive impairments.

Cognitive Development

Adequate sleep is a core component of cognitive function in adolescents [25]. During this critical period in development, sleep is a strong predictor of typical brain development, neurocognitive outcomes, and mental health [26-28]. This puts children with FMF at a significant risk for cognitive impairment given that they experience more frequent sleep disturbances [11]. As such, we have chosen to describe three outcomes heavily influenced by sleep. Given the gravity of sleep disturbances that children with FMF have in comparison to their typical counterparts, they are particularly vulnerable to deficits in neuroplasticity, lesser academic outcomes, and comorbidities [11,26,32,34]. Though this connection is understudied, further research should be done to establish a better understanding of the relationship between children with FMF and sleep-related cognitive impairments.

Neuroplasticity:

Neuroplasticity or neural plasticity, as defined by the National Institutes of Health, refers to the process by which the brain undergoes functional and structural changes, allowing the nervous system to respond to intrinsic or extrinsic stimuli by reorganising its structure, functions, or connections [29]. Sufficient sleep is crucial for neuroplasticity, especially in the refining of neural connections through the process of synaptic pruning [26]. This process streamlines neural circuits, facilitating increased cognitive skills [30], such as memory consolidation and integration [31]. However, sleep deprivation and poor sleep quality, both of which are prominent in children with FMF, pose a significant challenge to neuroplasticity. It can lead to a reduction in spine density and disrupt neuronal connectivity in the hippocampal CA1 region, a region of the hippocampus heavily involved with memory formation and retrieval of hippocampal-dependent memories [32]. Notably, Rapid Eye Movement (REM) sleep—a sleep cycle phase where brain activity, breathing, heart rate, and blood pressure increase, and the eyes move rapidly while closed—is incredibly important for neural plasticity [32,33]. REM is thought to play an important role in brain development and memory consolidation; this understanding is supported by the heightened formation and elimination of synapses in mice models [26]. Studies have also concluded that deprivation of REM sleep can cause the inhibition of hippocampal synaptic plasticity by affecting the amount of apical and basal dendritic spines present, as well as hippocampal long-term potentiation [32]. The intricate relationship between sleep and neuroplasticity highlights the essential role of sufficient sleep in refining neural connections and promotion of REM as well as underscores that sleep deprivation poses significant challenges to these processes. These complex relationships highlights the importance of addressing sleep-related challenges in FMF to support optimal brain development in affected children.

Academic Outcomes:

The foundation for understanding the association between sleep and academic outcomes stems from neurocognitive skills and their pivotal role in achieving academic success. Research has shown that sleep fragmentation, late bedtimes, and disrupted sleep significantly impact a child's learning ability, school performance and neurobehavioral functioning [27]. To succeed in the classroom, students are required to complete complex tasks which require creativity, abstract thought and integration [27]. Sleep disruptions weaken these cognitive skills by decreasing motivation, engagement, concentration, reasoning and problem-solving [34]. As elucidated earlier, these implications extend to structural and functional changes in the brain associated with memory– without sufficient sleep, the brain cannot undergo the structural changes required for memory consolidation [26,32] which hinders a child's ability to recall learned information. Furthermore, sleep deprivation has been shown to impact concentration negatively [35]. A study The foundation for understanding the association between sleep and academic outcomes stems from neurocognitive skills and their pivotal role in achieving academic success. Research has shown that sleep fragmentation, late bedtimes, and disrupted sleep significantly impact a child's learning ability, school performance and neurobehavioral functioning [27]. To succeed in the classroom, students are required to complete complex tasks which require creativity, abstract thought and integration [27]. Sleep disruptions weaken these cognitive skills by decreasing motivation, engagement, concentration, reasoning and problem-solving [34]. As elucidated earlier, these implications extend to structural and functional changes in the brain associated with memory– without sufficient sleep, the brain cannot undergo the structural changes required for memory consolidation [26,32] which hinders a child's ability to recall learned information. Furthermore, sleep deprivation has been shown to impact concentration negatively [35]. A study that assessed the relationship between quality of sleep and learning concentration in children found that 58.5% of children with low sleep quality had a middle learning concentration level and 30.2% had a low concentration level, with a p-value indicating a statistically significant correlation [36]. Moreover, research has shown that children with FMF miss significantly more school than their peers, which may hinder their ability to keep up with coursework and negatively impact their academic performance [37]. Ultimately, the negative effect of insufficient sleep on attention, learning and memory has been deemed to be one of the primary causes of children's failure to thrive in school settings [38], obstructing their ability to achieve academic success.

Comorbidities:

Adequate sleep plays a pivotal role in resetting our brains and protecting overall cognitive health [39]. On the other hand, insufficient sleep can give rise to a range of comorbidities. Namely, sleep deprivation places children at a heightened risk for developing mental health disorders such as anxiety, depression, attention deficit and hyperactivity disorder (ADHD) [28,40]. Research suggests a bidirectional correlation between lack of sleep and mental health disorders, demonstrating high rates of sleep problems among children with psychiatric conditions and reports of youth complaining of insomnia meeting the criteria for a mental health diagnosis [41]. Notably, lack of sleep has been found to further exacerbate mental illness. For example, depressed youth who suffer from insomnia and hypersomnia experience longer depressive episodes and more severe symptomatology [41]. Interestingly, studies reveal that almost 50% of children with ADHD also suffer from sleep disturbances, reinforcing the relationship between sleep quality and mental illness [41]. Prioritising sleep inchildren aged 8-15 is crucial to maintain overall cognitive health but also serves to prevent the development and progression of mental health disorders in youth. This relationship is particularly relevant for children with FMF, as research has found that they are more likely to suffer from anxiety and depression [42]. Given their increased susceptibility to both sleep disturbances and mental health disorders, prioritizing sleep in children with FMF may also play a role in mitigating the development of comorbidities.

Therapeutic Interventions

In addressing sleep disturbances, much of the existing literature focuses on the diagnosis and management of insomnia in the general population. The typical treatment process of insomnia begins with a comprehensive assessment of causes and triggers, followed by an exploration of interventions, including sleep hygiene routines, behavioural therapy, and pharmacological treatment [41]. Given the effectiveness of these interventions for treating insomnia in the broader population, exploring their potential benefits for children with FMF who suffer from sleep disturbances could be valuable. However, there is limited research on treatment options specifically tailored to paediatric FMF patients experiencing sleep issues, highlighting a gap in targeted interventions for these children.

Sleep Hygiene:

Educating children with FMF and their caregivers on sleep hygiene routines was noted to be an important start to treatment, as children with FMF are more prone to sleep disturbances that can impact their overall well-being. [43,44]. Important aspects of sleep hygiene include consistency in sleep and wake-up times, dietary factors such as caffeine intake, minimization of overstimulation prior to established bedtimes, and environmental factors such as ventilation and temperature in a child’s bedroom [43].

Behaviour Therapy:

Furthermore, the goal of behavioural therapy would be to remove negative associations that may result in sleep disturbances like insomnia, which are prevalent among children with FMF [43]. Some behavioural techniques include cognitive restructuring techniques, positive routines, and stimulus control; their benefits include improvement in sleep quality and quantity [43,44]. Given that children with FMF frequently experience disrupted sleep, techniques like Cognitive Behavioural Therapy for Insomnia (CBT-I), were noted to be more effective than other insomnia therapies and beneficial in mitigating sleep-related challenges [45].

Pharmacological Therapy:

In the treatment of sleep disturbances, behavioural therapy should be considered a first-line treatment [44]. Medication would typically be introduced when behavioural therapies are ineffective, or when children and their caregivers struggle to adapt to them [43,44]. Rather than pursuing medication on its own, a combination therapy of both pharmacologic and behavioural intervention has been noted to be an effective strategy [44]. To determine suitable pharmacological therapies, the following guidelines must be considered: the therapy should target symptoms and be developmentally appropriate for the child, the primary sleep disorder should be addressed prior to pursuing insomnia medication, and benefits should outweigh potential side effects [43]. Some examples of pharmacological therapies include melatonin, iron, and antihistamines [43]. In the context of treatment in patients with FMF, an important consideration is the potential of medications negatively interacting with or being involved in the fever’s pathways. For instance, a study conducted by Musabak et al. found that melatonin may be involved in the pathogenesis of FMF; this may affect the safety of the medication in care [46]. As such, further understanding of the safety and efficacy of medications would be required before prescribing pharmacological therapy to treat FMF.

Additional Supports:

In addition to therapeutic interventions, Corkum et al. noted an association between later start times in school with a higher number of students meeting sleep duration recommendations and a lower number of students experiencing morning fatigue [47]. Though no mandate was shared to change start times in Canada, informal reports across some school boards have shared success in implementing the aforementioned strategy [47].

DISCUSSION

Existing literature suggests that children diagnosed with FMF are at greater risk of experiencing poor sleep quality [11]. This poor sleep quality manifests as and is defined through sleep disturbances, increased bedtime resistance, sleep-onset delay, sleep anxiety, night wakings, parasomnias, and sleep-disordered breathing [11,19]. This narrative review demonstrates a correlation between FMF-associated poor sleep quality and cognitive development in children ages 8-15. A lack of sufficient sleep caused by bedtime resistance and other psychological obstacles associated with FMF poses significant challenges to the optimal development of neuroplasticity in children. Furthermore, sleep disruptions also weaken cognitive skills students require to succeed in the classroom, including attention, learning and memory [38]. Lastly, as a result of insufficient sleep, children with FMF are at a heightened risk of developing mental health disorders.

Although this narrative review presents promising findings, further research needs to be conducted to also synthesise findings regarding the impact of poor sleep quality caused by FMF on neuroplasticity, learning in academic settings, and other associated comorbidities in children aged 8-15 diagnosed with FMF. Conducting future research would help devise additional treatment and community supports for children with FMF. Given that FMF is the most common monogenic autoinflammatory disorder, new research has the potential to positively impact many lives. Most existing treatments for children with this diagnosis aid them in dealing with more well-known symptoms of their disorder. Furthermore, research has not been conducted on how to support children with FMF experiencing poor sleep quality as a result of their diagnosis and not due to a separate condition, like insomnia. Further research into these connections is essential to address the unique challenges faced by children with FMF, particularly as they suffer with the under-recognized symptom of poor sleep quality. Such efforts would pave the way for tailored support systems to enhance their quality of life

CONCLUSION

The impact of FMF on sleep is multifaceted and warrants careful consideration from paediatric healthcare providers. Research suggests that FMF not only disrupts sleep patterns directly through pain and discomfort associated with the condition but also indirectly through its effects on psychological well-being and cognitive development. Children with FMF often experience a range of sleep disturbances, including bedtime resistance, sleep-onset delay, and night wakings, which can significantly affect their quality of life. Poor sleep quality among FMF patients has been linked to increased levels of anxiety and depression, as well as cognitive deficits which may impede academic performance. Addressing sleep problems in children with FMF requires a comprehensive approach, including strategies such as sleep hygiene education, behavioural therapy, and, when necessary, pharmacological intervention. Additional investigation is warranted to confirm the relationship between FMF symptoms and sleep quality, and its influence on cognitive development in children and adolescents.

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UNDERSTANDING SEX-BASED DIFFERENCES IN CHILDHOOD CONDUCT DISORDER

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ABSTRACT

Externalizing symptoms are behaviours that violate major social norms, such as aggression, violation of rules, or deceitfulness. Conduct disorder (CD) is a mental disorder defined by patterns of externalizing behaviour. Recent studies find a significant discrepancy in the rates of childhood diagnosis between sexes, with a higher prevalence for boys compared to girls. Consequently, it has been suggested that current diagnostic criteria may not fully capture the nuanced manifestation of CD in girls. This paper aims to explore theories of sex differences in CD and implications for diagnostic criteria as described in the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition, Text Revision (DSM-5-TR). Through a review of the literature, this study examines childhood sex-specific differences in the symptoms, subtypes, comorbidities, and neurobiological correlates of CD. Theories discussed include the gender paradox, delayed onset pathway, and familial perspective theories. Based on sex-specific findings, implications for screening and diagnosis are discussed. In addition, the suggestion of advocating for further research on modifications in DSM criteria and use of sex-specific risk assessment tools is included. Research in this area has the potential to challenge misconceptions surrounding sex, gender, and externalizing behaviour with the goal of improving outcomes for Canadian youth with CD.

INTRODUCTION

According to the American Psychiatric Association, conduct disorder (CD) is a mental disorder characterized by self- and informant-reported continuous externalizing behaviour patterns that violate major age-appropriate social norms and rules, as well as rights of others [1]. This is in contrast to internalizing symptoms, which constitute internal anxiety and mood disorders, for example. The externalizing behaviours typical of CD—which can emerge in early childhood or adolescence—are grouped into four categories in Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition, Text Revision (DSM-5-TR): aggressive conduct that causes or threatens physical harm to other people or animals, nonaggressive conduct that causes property loss or damage, deceitfulness or theft, and serious violations of rules. Children with CD may also receive the specifier “with limited prosocial emotions” for exhibiting a lack of remorse or guilt, callous-unemotional (CU) traits, being unconcerned with performance, or shallow affect.[1] Children with CD may also receive the specifier “with limited prosocial emotions” for exhibiting a lack of remorse or guilt, callous-unemotional (CU) traits, being unconcerned with performance, or shallow affect [1].

Prevalence estimates of CD vary, ranging from 2-10% globally [1]. A recent review reported that the pooled global prevalence of CD is 3.0%, with boys being 2.6 times more likely to be diagnosed compared to girls [2]. In Ontario, recent estimates from the Ontario Child Health Study suggest that in children aged 12-16, the prevalence of CD is 2.52%, with higher estimates for boys (3.08%) than girls (1.92%) [3]. Taken together, these data illustrate the sex-based differences in the diagnosis of CD. Some authors have noted that the diagnostic criteria of the DSM-5-TR is not reflective of CD as it presents in women and girls [4].

The purpose of this paper is to explore theories as to why sex differences exist, synthesizing recent findings on differences in the symptoms, subtypes, comorbidities, and neurobiological correlates of CD, and discussing implications of these findings for current diagnostic procedures in Canada using the DSM-5-TR and early screening methods. This research recognizes that there are construct differences in sex and gender, and that various studies may assess different aspects of identity. Throughout this article, “male” and “female”, “girls” and "boys" are used interchangeably to refer to the sex of the individual. The conflated constructs of sex and gender in psychological research is a limitation that must be acknowledged, and all evidence should be considered with the knowledge that nuances of socially-conceptualized gender and individuals beyond the binary are not captured [5].

RESEARCH QUESTION

How can DSM diagnostic procedures and early screening methods be adjusted to consider potential sex differences in Canadian children and youth with CD?

To answer this research question, we will investigate the following:

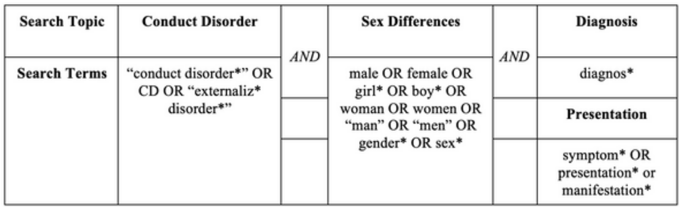
1. Are there sex differences in the symptoms, subtypes, comorbidities, and neurobiological correlates of CD?
2. What are the major hypotheses that account for these sex differences?
3. To what extent do current DSM diagnostic procedures and early screening methods account for sex differences in CD?

RATIONALE

Sex differences in the manifestation of CD is an emerging topic in research; however, established knowledge and current literature has several limitations. Far less is known about the causes of CD in females due to the majority of early mechanistic studies being restricted to male patients [6]. Furthermore, an insufficient number of female subjects have been included in research examining the sex differences in such antisocial behaviour despite it recently being established that there are epidemiological differences in prevalence [6]. There are conflicting theories surrounding these sex differences and the neurodevelopmental origins of CD [6]. The evidence surrounding current theories should be considered, considering its potential implications for current diagnostic and classification systems. Progress in this area can work to dismantle stereotypes surrounding sex, gender and externalizing behaviour, fostering more nuanced approaches to assessment and treatment, and preventing the underdiagnosis of children who may benefit from care [7].

METHODS

This paper primarily uses evidence from recent peer-reviewed journal articles and sources located on Google Scholar, Web of Science, PsychINFO, and PubMed/Medline. The table below describes the keywords employed.



Search Topic	**Conduct Disorder**	*AND*	**Sex Differences**	*AND*	**Diagnosis**
Search Terms	"conduct disorder*" OR CD OR "externaliz* disorder*"		male OR female OR girl* OR boy* OR woman OR women OR "man" OR "men" OR gender* OR sex*		diagnos*
					Presentation
					symptom* OR presentation* or manifestation*

In addition to traditional database searches, forward and backward citation searches were also conducted on identified articles.

Evidence was limited to English and French publications. Given the little amount of literature in this area, there was no limitation on the type of study. Further, any study discussing externalizing disorders in general was included if there was an explicit mention of CD as one of the conditions studied.

Data was synthesized in a qualitative manner.

DIFFERENCES IN THE CLINICAL MANIFESTATIONS OF CONDUCT DISORDER ACROSS GENDERS

Theories

There are several theories that seek to explain and describe gender differences in the clinical manifestation of CD, many of them still pending investigation.

Gender Paradox and Differential Threshold:
The gender paradox theory refers to the fact that although the prevalence of girls with CD is lower than boys, girls typically present with more severe behaviour problems and more co-occurring symptoms and disorders [8]. Related to this concept is the differential threshold of female CD theory, which suggests that the current diagnostic criteria for CD are not sensitive to the nuances in female presentation [1,9].

Delayed Onset Pathway:
The delayed onset pathway theory claims that the onset of CD in girls is usually during adolescence (AOCD), and thus typically later than boys who have an average childhood onset of CD (COCD) [10].

Familial Perspective:
Finally, one ongoing discussion is the familial perspective [11]. One review exploring family functioning and processes and associations with CD, found that one possible explanation for sex differences in CD presentation is that parents tend to emphasize prosocial behaviours and control of anger in girls, treating them with less aggression and more warmth compared to boys [11]. Girls are also more closely monitored by parents, potentially acting as a protective factor against disruptive behaviour. However, in girls who display a high number of externalizing behaviours, reciprocal associations between child disruptive behaviour and parenting behaviour explains why symptoms may be more severe in girls; this is the idea

that the child's temperament and caregiver's behavioural responses feed into each other, and over time, both reinforce and worsen the externalizing behaviours [11]. Additionally, with regards to adolescent-onset disruptive behaviour, there may be a risk period during puberty for girls. Parental monitoring decreases during this transition, which is related to an increase in interpersonal conflicts and externalizing behaviour. Girls' friendships, characterized by greater intimacy, may influence their behaviour, and involvement with deviant peers might be more negative for girls than boys.

Comorbities

The DSM-5-TR outlines various comorbidities and complications of CD that span from childhood to adulthood. These include learning disabilities, diminished academic achievement, attention-deficit/hyperactivity disorder (ADHD), mood disorders, anxiety disorders, psychotic disorders, somatic symptom disorders, impulse-control disorders, and post-traumatic stress disorder (PTSD) [1,12]. Children diagnosed with CD may progress into adulthood with an elevated likelihood of developing antisocial personality disorders [13]. Furthermore, the DSM-5-TR states that behavioural challenges associated with the disorder may result in children and youth with CD encountering issues related to substance use and criminal convictions [1]. Individuals with CD also face an elevated risk of suicidality and poor life satisfaction [1]. It is noted that CD is associated with higher rates of suicide attempts even after adjustment for comorbid mood, anxiety, and substance use disorders [1]. It is worth noting that the DSM outlines differences across genders in CD comorbidities, specifically when it comes to substance use. While the association between CD and substance use is well-established, higher rates are observed in adolescent females [1]. The reason behind this gender difference is theorized to be due to females reaching puberty at an earlier age than males [14]. The 2006 Minnesota Twin Family Study suggests that genetic influences on CD are moderated by the timing of menarche in girls. Early puberty/menarche in girls has been linked to higher rates of CD symptoms, an earlier initiation of the disorder, greater frequency of substance use, and earlier sexual activity [14]. In contrast, girls with delayed menarche appear to be protected from the well-documented increase in externalizing behaviour during adolescence [14]. As puberty in boys and girls involves different social and biological phenomena, it remains unclear whether these findings would generalize to boys, and future research should seek to evaluate the impact of early pubertal timing on CD longitudinally in boys [14].

There is evidence that females with CD are also more likely to have comorbid externalizing and internalizing disorders compared to their male counterparts, who generally display heightened rates of externalizing disorders alone [15]. Furthermore, early depressive symptoms are a stronger risk factor for delinquent behaviour in females than in males. It was found that girls exhibit both delinquency as a predictor for subsequent depression and depression as a predictor for increased subsequent delinquency. In males, only early delinquency predicted subsequent depression across adolescence [15].

Further, females with CD are more likely to exhibit comorbid depression than males, a finding consistent with the gender paradox hypothesis [16].

Timing Differences

Research indicates that childhood onset of CD is less common in girls than in boys, with a higher prevalence of onset during adolescence [17]. Only 1-2% of girls follow the childhood onset trajectory of CD, compared to 5-10% of boys. Sexdifference ratios ranging between 10-15:1 for boys to girls with CD have been reported [18-20]. It is important to note only 1-2% of females from population-based or birth cohort samples were followed from childhood through adulthood,complicating prevalence estimates. This is a significant gap because childhood onset conduct disorder (COCD) has been associated with serious adjustment and antisocial problems in young adulthood, suggesting that young girls with COCD may be underdiagnosed and lack necessary support relative to other subtypes [21]. Despite an influential model describing two distinct trajectories of CD onset and the observation of increased adolescent onset conduct disorder (AOCD) in girls, these girls tend to resemble COCD boys on personality traits, such as poor impulse control and callous-unemotional (CU) traits [22]. This highlights that CU trait symptomatology may not be as uncommon in AOCD in girls and may not be restricted to COCD.

Behavioural/Symptomatic Differences

Beyond differences in comorbidities and diagnostic timing, there are several differences in the symptoms of CD presented by girls compared to boys. A recent preliminary study found that males with CD made significantly more risky choices than male controls, while females did not differ significantly from their typical female counterparts regarding such risk- and reward-seeking behaviours [23]. This finding suggests that sex differences exist in reward-related decision-making for youth with CD and that these differences are more pronounced in males with CD. These differences in risk-taking behaviour may also relate to the earlier discussion of internalizing versus externalizing symptoms in boys versus girls. Girls with CD are more likely to have comorbid internalizing disorders compared to their male counterparts, who generally display heightened rates of externalizing characteristics alone [15].

Regarding physical aggression, while boys in community samples exhibit more physical aggression than girls, clinical samples reveal that both girls and boys with highly impaired CD show equally high levels of physical aggression [15]. This unexplained discrepancy in findings underscores the need for further research to establish the prevalence of physical aggression in females with CD.

However, Females with CD also show higher relational aggression scores than males [24]. The established research indicating that females with CD are more likely to engage in relational aggression has significant clinical implications [25]. For example, callous-unemotional (CU) traits and low affective empathy have been positively correlated with relational aggression and antisocial behaviour [24]. Even after controlling for more severe and early-onset conduct symptoms, children and youth with CU traits exhibit more antisocial symptoms in adulthood, such as adult arrests and antisocial personality symptoms, than those without [26,27]. Furthermore, in females, cognitive empathy is negatively associated with relational aggression [24]. Currently, research has predominantly focused on male-related CU traits; however, girls remain an understudied group in the examination of these traits, which evidently has implications for behaviour and delinquency [28, 29].

Studies have shown that gender differences do exist in the association between CD and risky sexual behaviour. The association is stronger in female youth, with research finding that female juvenile offenders report a higher likelihood of risky sexual behaviour including being less likely to use condoms and having higher rates of STIs than males [30,31]. This underscores the need for gender-specific interventions and preventative supports for youth with CD.

Research has shown that boys exhibit more overt conduct problems such as status and property offenses, while girls are more prone to covert conduct problems such as shoplifting and fraud [32,33]. The lower rates of CD diagnosis in females may potentially be due to underdiagnosis and a lack of recognition of these covert problems which are often less identified. Conflicting evidence has been presented, showing that the gap between male and female rates of delinquent behaviour has narrowed over the last few decades. In particular, violent crimes may predict a greater incidence of CD diagnoses for girls [34]. This highlights the importance of further sex and gender-specific research in this field and underscores its clinical significance.

Research has found that aggressive boys with CD demonstrate reduced autonomic functioning, while aggressive girls with conduct problems exhibit greater functioning. This suggests that there may be different etiological mechanisms underlying psychopathy in males and females [35]. However, limitations to this research have been highlighted due to the overwhelming proportion of male to female participants in most studies, making it difficult to draw definitive conclusions about autonomic activity in both genders.

Another difference that has been highlighted in research involves emotion processing skills, which can act as a framework for explaining the sex differences in CD [36]. Research has shown that girls mostly outperform boys on scales of social cognition, which encompasses emotional processing tasks due to earlier maturation of brain systems involved in emotional responsibility and regulation [36]. Due to displaying these greater emotional functioning skills, they appear to be better equipped for challenges involving socialization. Furthermore, traditional gender roles do encourage more prosocial behaviour in girls, which may lead to the difference in presentation and manifestation of symptoms of CD.

Neurobiological Differences

There are several neurological and biological differences that may also help explain the differences in CD presentation.

One study using diffusion MRI to examine differences in white matter integrity in children with CD found that abnormalities of the superior longitudinal fasciculus were positively correlated with levels of CD symptoms, which has previously been associated with cognitive deficits in children [37]. Further, the uncinate fasciculus (UF) has a role linking the amygdala and the ventromedial prefrontal cortex; this study found that reduced radial diffusivity in the left UF correlated with increased CD symptoms, especially in girls. This article concluded that abnormalities in brain structure may contribute to the emergence of CD in childhood, playing a particularly important role in girls. CD was associated with cortical thinning and higher gyrification in the ventromedial prefrontal cortex in both sexes. Males with CD showed lower, and females with CD showed higher, supramarginal gyrus cortical thickness compared with controls. Relative to controls, males with CD showed higher gyrification and surface area in superior frontal gyrus, whereas the opposite pattern was seen in females [36].

There are various studies that have investigated differences in the genetic basis of CD. One particular gene of interest is the gene that produces monoamine oxidase-A (MAO-A), an enzyme that catalyzes the breakdown of monoamine neurotransmitters (serotonin, dopamine, and norepinephrine) [38]. This gene, MAO-A is located on the X-chromosome and makes male carriers hemizygous and more at risk to consequences of mutations and other variants. A recent meta-analysis confirmed that high expression variants of MAO-O could increase the risk for antisocial behaviour [38].

Various studies have explored sex hormones as a potential mechanism behind the development of CD. Testosterone and dehydroepiandrosterone (DHEA, the precursor of testosterone) have been suggested to be positively correlated with aggressive behaviour across genders, but particularly male adolescents [39]. The implications of this are not yet known, however, girls with CD have been found to have lower cortisol to DHEA ratios, higher levels of free testosterone, and lower levels of SHBG, indicating a possible hormonal imbalance [40].

This initial evidence that the pathophysiological basis of

CD may be partly sex-specific highlights the need to consider sex in future studies and suggests that males and females may require different treatments, as current physiological treatments (i.e., pharmaceutical agents) are not sex-informed and limited to treating individual symptoms rather than the disorder as a whole.

IMPLICATIONS FOR DIAGNOSTIC PROCEDURES

Rationale for Considering and Modifying the Female-Specific Diagnostic Protocols

The underlying reason for the lower prevalence of CD among females remains unknown [4]. Hypotheses include true sex-related differences in CD, a sex bias against females in the diagnostic criteria; however, both hypotheses are likely to contribute to the overall lower prevalence.

The DSM-5-TR partially includes clinical manifestations in girls. Gender-typical behaviours, including running away, oppositional behaviour, and non-confrontational aggression, are mentioned as typical manifestations in girls with CD. However, the DSM-5-TR does not include gender-specific criteria, and the diagnostic criteria are developed from studies with primarily male samples [1]. Criteria centered around behaviours more common in boys may fail to accurately diagnose CD in girls [5]. Ambiguity regarding the sensitivity and specificity of the diagnostic criteria has raised concerns among clinicians about diagnosis and treatment in female patients.

In order to better account for sex differences in CD presentation among females, several changes to the DSM-5-TR have been suggested [41-46]. The current reliance on criteria developed and validated primarily on boys may contribute to underdiagnosis or misdiagnosis in girls. Three proposed changes to the DSM for CD are discussed below. All three have mixed evidence on their specificity and sensitivity for CD diagnosis in girls and require more research before incorporation into future versions of the DSM.

Firstly, the DSM could incorporate gender-typical behaviours associated with CD in girls. Girls may express relational aggression, defined as hurting others through their social position, more often than boys with conduct problems [41]. It has been proposed that criteria for relational aggression be added to the DSM (5). However, there is mixed evidence on the overlap between high relational aggression scores and meeting the threshold for CD in girls [42]. More research is needed to examine the validity of this symptom for CD diagnosis.

Secondly, it has been proposed that the symptom threshold for CD in girls be changed from three to two [43]. There is mixed evidence to support this threshold change, with some studies reporting increased sensitivity in diagnosis without a loss in specificity [43], while some report that the loss of impairment in girls with three symptoms is much higher than those with only two symptoms, thereby not supporting a change in diagnosis threshold [42].

Finally, it has been proposed that the age subsets of CD should be changed for girls; the childhood-onset may not be needed, or the adolescent-onset should be defined as the "absence of any criteria prior to age 13" rather than age 10 [44]. However, there are mixed findings on whether girls with CD truly have a later age of onset [9,44], or if their symptoms become more intense with age but have a similar onset age as boys [42,45,46]. This highlights a need for further research following females from birth to adulthood to establish and confirm the proportion of females experiencing a childhood-onset trajectory. Furthermore, it highlights that modifications and alternatives to the two-trajectory model may be needed to support the observations in development of CD in girls.

Evaluation of the Early Assessment Risk List Version 3 (EARL-V3)

In addition to changing diagnostic procedures, increasing early screening of girls displaying symptoms of CD can help improve detection of CD. The EARL-V3 is one such screening tool that should be rigorously evaluated. The EARL-V3 is a structured professional judgement instrument for children ages 6-12 showing antisocial behaviour that is designed to specifically take a gender-sensitive approach to risk assessment [47]. It has been shown to have moderate-to-high interrater reliability and concurrent validity with other scales that predict rule-breaking and aggressive behaviour [48].The EARL-V3 should be further validated using longitudinal studies with diverse cohorts of girls, including those with conduct problems and those without, to ascertain its effectiveness in distinguishing between normative behaviours and early signs of CD in girls. As the tool is relatively new, having been released in 2021, more studies are needed to validate it although initial results are promising.

CONCLUSION

In conclusion, addressing the lower prevalence of CD among girls requires a multifaceted approach. Further research, including longitudinal studies and comprehensive assessments of gender-specific symptoms, is imperative to enhance the understanding of CD in girls and improve diagnostic accuracy for better long-term prognosis. Proposed modifications for the DSM-5-TR can encompass gender-specific diagnostic criteria, particularly focusing on adolescent-onset subtypes for girls. Additionally, the evaluation of gender-specific risk assessment tools, like EARL-21G and EARL-20B, is crucial to ensure their reliability and validity in screening for CD outcomes.

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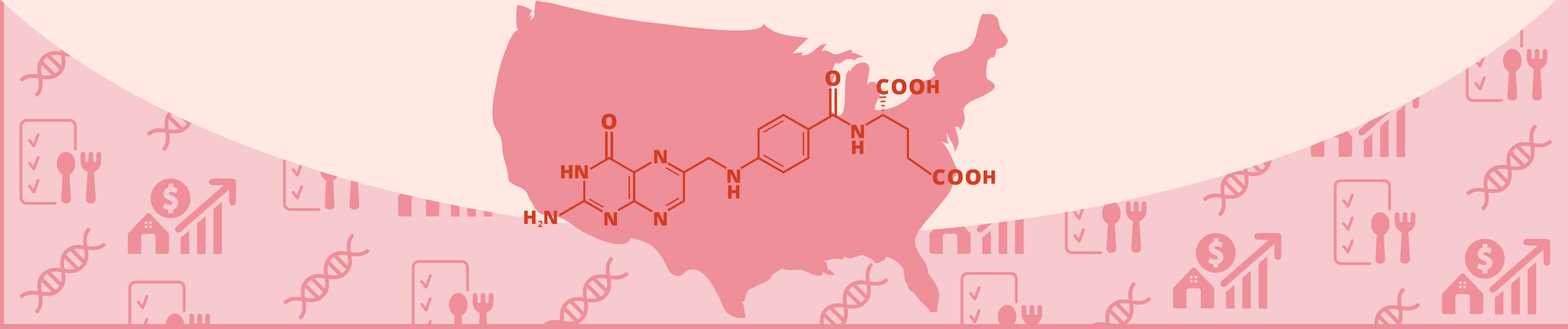

DELVING INTO THE DISPARITY: EXPLORING HIGH RATES OF SPINA BIFIDA IN HISPANIC COMMUNITIES IN THE UNITED STATES AFTER THE 1998 FOLIC ACID FORTIFICATION PROGRAM

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ABSTRACT

Background: Spina bifida (SB) is a congenital defect surrounding the development of the spine and spinal cord leading to neurological impairments, influenced by insufficient folic acid (FA) during gestation. Despite the U.S.'s implementation of a FA fortification program in 1998 to reduce the incidence of SB, there continues to be a discrepancy in SB rates between. Hispanic communities and non-Hispanic communities in the nation, highlighting a need for further exploration.

Objective: This paper explores the factors that contribute to the disproportionately high prevalence of SB in Hispanic communities as compared to the general population in the U.S. after the 1998 fortification program.

Methods: A literature review was conducted to assess the disparity in SB prevalence among ethnic groups in the U.S. post-1998, focusing on genetic, socioeconomic, and dietary factors.

Results: Analysis of the retrieved literature revealed that the combined effects of genetic, socioeconomic, and dietary factors correlated with a higher prevalence of spina bifida in Hispanic communities as opposed to non-Hispanic communities. Genetic studies highlighted elevated frequencies of the Methylenetetrahydrofolate reductase (MTHFR) gene variants in Hispanic populations. Additionally, socioeconomic disparities and dietary habits underscored challenges in the intake of folic acid and accessibility to fortified foods.

Conclusion: The findings in this paper support the following recommendations to help reduce the disproportionately high prevalence of spina bifida in Hispanic communities by; a) expanding the mandatory FA fortification program to include more products that are consumed by the Hispanic community and b) increasing community education about FA fortification.

INTRODUCTION

Spina bifida (SB) is a congenital disorder, and a type of neural tube defect (NTD), characterized by failed closure of the embryonic neural tube. The disorder encompasses myelomeningocele, meningocele, and spina bifida occulta. These vary greatly in regards to embryological development and prognosis. Myelomeningocele, which is the most common and most severe form of SB, is defined by an open spinal cord with a meningeal cyst. Meningocele refers to the protrusion of the meninges (filled with cerebrospinal fluid) through a defect in the skull or spine. SB occulta is a closed asymptomatic NTD in which some vertebrae are not completely fused. The symptomatology of SB varies greatly between cases, but it often causes individuals to exhibit motor and sensory neurological deficiencies below the level of the lesion. This can cause lower limb weakness, paralysis, lack of sensation, and urinary and fecal incontinence. SB is known to have a multifactorial inheritance, including numerous genetic and environmental risk factors, such as maternal substance use, maternal diet, geographic location, and certain demographic traits [1].

In 1996, the United States became the first country to introduce a FA fortification mandate to decrease the incidence of NTDs among its population. As of 2020, 72 countries have followed suit to implement FA fortification due to its success in United States [3]. The United States Food and Drug Administration declared that by January 1998, all grain cereal products on shelves across the nation be fortified with FA [3]. Findings from numerous papers that studied this intervention provide evidence that prenatal folic acid deficiency is a risk factor in the development of SB [4]. For instance, one population-based study investigated over 5000 NTD cases between 1995 and 1999 in 24 populations across the United States, calculating the prevalence of SB for births before and after the introduction of the 1998 FA fortification mandate. The study found a 31% decrease

[0.61 prevalence ratio] in cases, with a 95% confidence interval, reporting a temporal association between maternal prenatal FA intake and the prevalence of fetal SB development [5].

Hispanic refers to a person with ancestry from a country whose primary language is Spanish. Whereas, Latino, and its variations Latina and Latinx, refer to a person with origins from anywhere in Latin America [6]. There is no standard to how these terms are used within a research setting, for example, the Census Bureau now incorporates both as one category: "Hispanic or Latino" [7]. This paper will interpret the results of various studies using Hispanic and Latino as interchangeable terms, employing them as derived from their original publications.

Trends post-fortification in the period of 1999 to 2004 revealed that Hispanic infants had the smallest decrease in prevalence of neural tube defects (8%), in contrast to non-Hispanic White infants (11%) and non-Hispanic Black infants (14%) [8]. Furthermore, the prevalence of SB for Hispanic infants is currently reported to be the highest in the nation [9].

This article aims to explore contributing factors to the disproportionately high prevalence of SB in Hispanic communities as compared to the general population in the United States after the FA fortification program came into effect in 1998.

METHODS

A non-systematic literature search was conducted on relevant databases including PubMed, MEDLINE, Web of Science, and Cochrane. As well, grey literature from search engines (mainly Google and Google Scholar) were reviewed. In our search we employed Boolean operators and truncation features, with key search terms including, but not limited to: Hispanic, Latin, neural tube defects, spina bifida, and FA fortification. The populations of included papers had to have subgroups of Hispanics and/or Latino individuals. Additionally, the time frame of included articles was limited to those published in or after the year 1998 to eliminate studies that investigated SB prevalence before the implementation of the American folic acid fortification program. The outcome of interest was SB incidence. From the literature search, three prevalent themes were identified; genetic factors, maternal diet, and socioeconomic status. The connection between these three factors and SB will be further explored in this paper.

RESULTS

Genetic Factors

The disproportionately high prevalence of SB in Hispanic communities as compared to the general population in the United States may be partly attributed to genetic factors.

SB is known to have a multifactorial polygenic etiology, meaning that multiple genetic pathways are implicated in the development of the disorder [10]. The Methylenetetrahydrofolate reductase (MTHFR) gene is one such example and is involved in the creation of the MTHFR protein, which is important in folate metabolism [11]. Certain variants of the MTHFR gene are associated with an increased risk of NTD development, as they decrease the activity of the MTHFR protein [12]. For instance, the MTHFR C677T polymorphism results in a substitution mutation of cytosine for thymine at nucleotide 677, which alters the consequent amino acid from alanine to valine [13]. This missense mutation has functional consequences, reducing the activity of the MTHFR protein in folate metabolism [14].

Additionally, a review by Sharp and Little (2004) highlights the role of folate metabolism and MTHFR gene variants in various populations. Notably, U.S. Hispanics exhibit a higher prevalence of the TT genotype, a genotype that leads to decreased folate metabolism, as compared to African American and Caucasian populations [13].

The MTHFR C677T mutation varies in prevalence across different ethnic populations. A 2015 study with more than 1000 participants from across the United States reported that Hispanic Americans had the highest observed 677T allele frequency (42%), as compared to African Americans (16%) and Caucasian Americans (32%) [15]. These genetic differences correlate with the increased risk of NTD development amongst the Hispanic-American population [16].

Maternal Diet

Maternal diet, particularly concerning folic acid consumption, plays a crucial role in preventing NTDs, such as SB, including in Hispanic communities in the United States [17].

FA is known to significantly reduce the risk of SB when consumed before conception and during early pregnancy. However, due to dietary habits and disparities in prenatal education, insufficient FA consumption is particularly prevalent among Hispanic communities in the United States. Insufficient FA consumption may contribute to higher rates of SB in these populations may contribute to higher rates of SB in these populations [18].

Dietary preferences and habits of Hispanic populations reinforce the decreased impact of the 1998 FA fortification program on these communities.8 Though the United States government has mandated the fortification of cereal and some grain products with FA and successfully reduced the incidence of NTDs [16], these foods are not tailored to the diets of Hispanic communities. For instance, Hispanic diets are often dominated by corn-based products, as opposed to flour-based products, meaning they do not benefit from the FA fortification mandate to the same extent as the general population [19].

While introducing FA fortification in cereal is economically viable, due to its low cost per calorie and edible gram [20], it may have disproportionately benefited one culture's dietary intake as opposed to another. As reported by qualitative interviews with several Hispanic women from nine countries of origin living in the United States, Hispanic women's criterion for purchasing food is primarily based on familial preference [21]. As aforementioned, Hispanic communities tend to prefer corn-based products over flour-based foods [20]. This could justify the decreased impact observed in Hispanic communities after the advent of FA fortified cereals in 1998 [8].

Furthermore, access to health information likely impacts the understanding of the importance of FA consumption and prenatal care among Hispanic women [18]. Surveys of women aged 18-45 found that, as compared to Non-Hispanic White women, non-White Hispanic women had lower awareness of the importance of FA intake on preventing spina bifida [17,18,22-25]. A national survey of 1685 non-pregnant women in the United States analyzed the ethnicities of the women consuming the suggested 400 μg daily dose of FA through fortified foods and found that only 6.8% of Hispanic women met this recommendation, as compared to 8.9% of non-White Hispanic women [23]. Furthermore, when including nutritional supplements in daily intake measurements, still only 21% of Hispanic women consumed the recommended 400 μg FA, as compared to 40.5% of non-Hispanic white women [23]. These intake disparities may be attributed to the combined impacts of dietary preferences and educational inequities within the Hispanic community.

Socioeconomic Factors

Socioeconomic status (SES) is a way of describing one's educational attainment, income, and occupational prestige and is consistently correlated to an array of psycho-social and biological outcomes [26]; SB and other NTDs are not an exception to this commonality [27]. Hispanic communities on average have lower socioeconomic status (SES) than other ethnic communities within the United States, which could explain certain aspects of the disparity in SB prevalence between these groups. For instance, Hispanic households traditionally have a smaller median income than White households [28]. Furthermore, 33% of Latino youth are currently living in poverty, as compared to a 14% poverty rate for non-Latino youth [29].

Within the past 20 years, there has been a decline in studies that look at the effect of SES on SB prevalence in Hispanic populations. In particular, one study conducted in San Joaquin Valley determined that acculturated neighbourhoods were a protective factor in preventing NTD prevalence. Acculturated neighbourhoods are defined as those with a higher proportion of US citizens, greater English proficiency and a lower Hispanic population. There was an association between non-acculturated neighbourhoods and inhalable polluted particulate matter, whereas no association was observed in acculturated neighbourhoods. Furthermore, there was a correlation between NTD prevalence and non-acculturated neighbourhoods [30]. This points to a lack of data surrounding this topic and a call to action from the authors to generate more research to further establish the correlation between SES and SB prevalence.

DISCUSSION

An analysis of the association of genetic factors, maternal diet, and socioeconomic factors with SB prevalence within Hispanic and non-Hispanic communities in the United States provides a comprehensive overview as to why the former population has disproportionately high rates of SB when compared to the general population. The MTHFR C677T polymorphism – a common mutation of the MTHFR gene – is shown to be associated with the development of NTDs such as SB and is known to be more common in Hispanic American populations. Insufficient FA consumption in Hispanic communities in the United States due to cultural dietary preferences and educational disparities contributes to higher rates of SB despite the government's fortification efforts. SES's correlation with SB, an understudied subject in recent years, shows a greater prevalence with a lower SES due to factors such as neighbourhood characteristics and culture.

Taking into account the current factors that affect SB prevalence in the Hispanic population, this paper encourages the American government to explore interventions that address these disparities. One approach is to mandate FA fortification in foods that consider the diet of the Hispanic population. Currently, the United States FA fortification program mandates that cereal grain products be fortified; including bread, pasta, rice, and cereal [31]. As stated previously, Hispanic women's criterion for purchasing food is according to familial preference, and thus, typical Western foods that are fortified with FA may not be purchased [21]. Interestingly, to address the disparity found in Hispanic populations, the United States added voluntary FA fortification in corn masa flour in 2016 [31]. However, a study by Wang et al. shows that there was not a significant difference in the risk of NTDs amongst the Hispanic population after the introduction of this policy [32]. Investigating this further, Kancherla et al. found that implementing voluntary FA fortification did not achieve widespread popularity amongst corn masa flour brands [33]. The researchers received photos of 43 corn masa flour products from participants across 28 states and found that only 3 included FA. Additionally, zero corn tortilla products were fortified [33]. This presents a clear gap in the policy implemented by the United States government. Voluntary fortification has yet to have the intended effect on the Hispanic population, which may be due to the lack of culturally appropriate products that have FA fortification. In fact, this global systematic review identified the greatest reduction in NTD prevalence in Chile and Costa Rica after mandating FA fortification in all corn masa flour products. This significant reduction in Chile and Costa Rica was a 50% and 68% decrease, respectively, in NTD prevalence amongst Latinx communities [34]. However, it is also crucial to ensure that FA fortification of foods is regulated to adequately impact the intended target. The unregulated FA fortification program in Mexico resulted in 9-33% of women of child-bearing age consuming insufficient FA while 12% of children had an intake of FA above the tolerable limit [35], and the insufficient standards for fortification in Peru did not have an impact on NTD incidence [36]. Thus, the United States can explore a FA fortification mandate in corn masa products, similar to that of cereal grains, which is evidenced to have significantly decreased NTD prevalence amongst the general population. Thus, the United States can explore a FA fortification mandate in corn masa products, similar to that of cereal grains, which is evidenced to have significantly decreased NTD prevalence amongst the general population.

Fortifying commonly consumed foods can be applied to products that are utilized across all cultures, one of the most common ones being salt. This has been previously successful in preventing goitres in American populations by simply adding iodine to all salts [37]. Food nanotechnology and chemical engineering studies conducted in Canada have successfully created salt that can impart the recommended amount of folate (Vitamin B9) in iodized salt without affecting the flavour, colour, or sensory properties of the salt and remaining cost-effective [38,39].

An intervention that could be explored is to increase the awareness of FA supplementation among Hispanic populations. For example, there is a disproportionately larger proportion of Hispanic individuals from a lower socioeconomic status [28]. Further, Hispanic women have been found to have less prenatal education [18]. Furthermore, health education materials and outreach efforts may not always be culturally or linguistically tailored to effectively reach these communities [40]. A study conducted by deRosset et al. used the promotora de salud model to encourage the use of FA supplementation amongst Hispanic women in North Carolina [17,41]. This model leverages community health workers to support the health of Latinx community members. These workers can help provide information about FA, its importance, and address any barriers associated with having FA in the diet or via supplementation. This approach can ensure that culturally competent information is provided to members and leverages the existing trust between mothers and community health workers. The study showed a significant increase in FA supplement consumption between the baseline characteristics and at the 4-month follow-up. This provides support for the use of this model in encouraging the use of FA consumption in a culturally competent manner among the Hispanic population.

Although the Hispanic population has a disproportionately higher prevalence of SB, there are several interventions - such as making FA fortification mandatory for certain cultural foods and leveraging the promotora de salud model (a model that utilizes community identified leaders with shared social, cultural, and economic characteristics as community health workers)[17] - that the United States of America government can explore. This can lead to improved maternal and infant health outcomes and help narrow the disparities that exist in perinatal health among Hispanic communities. It is also important to engage with the Hispanic community to ensure that interventions are informed by their opinions. Thus, we also suggest future participatory action research studies to ensure bidirectional crosstalk between policymakers and the target communities [42].

LIMITATIONS

This paper has several limitations. Firstly, due to the paucity of research specifically pertaining to SB, many articles used in this review investigated the prevalence of NTDs overall. This is a limitation, as there are other NTDs beyond SB, such as encephalocele; therefore, the prevalence may not have been fully specific to SB. Future research may attempt to find SB specific prevalence data, or to potentially explore SB specific data. Additionally, as mentioned in the paper's introduction, the definition of the target population may not be comprehensive or may be overinclusive due to discrepancies between journals on the delineation of the Hispanic population. Furthermore, many articles included in this review used 'self-identification of ethnicity' as a method for assigning participants to research groups, which may result in inaccurate or inconsistent characterization of the target population. Further research may want to further investigate the extent of how "self-identification" may influence results, or to find research that does not rely on "self-identification." This could also contribute to the lack of insights from the Hispanic and Latinx communities. Their insights would be essential to learn from their own lived experiences. Future research endeavors should investigate the perspectives of Hispanic and Latinx communities.

CONCLUSION

The findings of this paper highlights genetic factors, maternal diet, and socioeconomic influences that contribute to the disproportionately high rates of SB in Hispanic communities compared to non-Hispanic communities in the United States. To address this health disparity, our research advocates for the expansion of mandatory FA fortification programs to encompass more products that are consumed by the Hispanic community. In addition, targeted community education about FA is recommended. Future research should prioritize the implementations of Hispanic communities perspectives, such as through participatory action research studies. Such an approach ensures that communities are involved with identifying the issues and are actively involved in the creation of culturally relevant programs that can address the observed discrepancy in SB prevalence. It is essential to gain their perspectives, such as through participatory action research studies. Such an approach ensures that communities are involved with identifying the issues and are actively involved in the creation of culturally relevant programs that can address the observed discrepancy in SB prevalence.

KEY MESSAGES

- Hispanic communities in the United States experience disproportionately high prevalence of spina bifida.
- Genetic factors, maternal diet, and socioeconomic factors are all associated with higher prevalence of spina bifida in the Hispanic communities.
- Expansion of folic acid fortification program to more products and community education about the importance of FA are ways to address the disparity in spina bifida prevalence.
- Future studies should adopt a participatory action approach to help identify and address the challenges faced by Hispanic communities in relation to spina bifida.

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ACCESSIBILITY IMPLICATIONS OF T1D INTERVENTIONS (MDI, CSII, AID) FOR CHILDREN AND ADOLESCENTS AGED 5-15 IN THE US AND CANADA: IDENTIFYING GAPS AND FUTURE ADVANCEMENT OPPORTUNITIES

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ABSTRACT

This narrative review delves into the growing challenge of Type 1 Diabetes (T1D) among children and youth, characterized by the autoimmune destruction of insulin-producing beta cells, necessitating lifelong management and impacting health and well-being. It explores the accessibility and challenges of commonly employed T1D interventions such as Multiple Daily Injections (MDI), Continuous Subcutaneous Insulin Infusion (CSII), and Automated Insulin Delivery (AID) in the pediatric population. The article focuses on financial and geographical barriers within the United States of America (US) and Canada and providing policy and integration suggestions for better accessibility of these interventions. The financial burden of T1D management is significant, with rising costs of insulin and diabetes management technologies posing barriers to access. Geographical disparities further exacerbate these challenges, particularly in rural areas where specialized care and resources are scarce. The need for policy reform strategies to mitigate financial barriers and enhance care accessibility is highlighted, emphasizing the role of telehealth and digital interventions to bridge the geographical divide. Future research should focus on the long-term outcomes of these interventions and the impact of policy changes on accessibility and health outcomes for children and youth. Throughout these future discoveries, age-appropriate adaptations to insulin delivery methods must be constructively considered. This review emphasizes the critical need for innovative, cost-effective, and accessible T1D care for children. Addressing financial and geographical barriers is essential to ensure equitable access to treatment and support for the pediatric population, emphasizing the need for a comprehensive approach that supports both physical health and overall well-being.

Type 1 Diabetes (T1D), children, youth, United States of America (US), Canada, accessibility, continuous subcutaneous insulin infusion (CSII), automated insulin delivery (AID), multiple daily injections (MDI), continuous glucose monitoring (CGM), geographical disparities in healthcare, rural healthcare access, telemedicine in diabetes care, financial barriers, T1D treatment

INTRODUCTION

Type 1 Diabetes (T1D) represents a significant and growing challenge in pediatric healthcare, characterized by the autoimmune destruction of insulin-producing beta cells in the pancreas [1]. This chronic condition, predominantly diagnosed in childhood and adolescence, necessitates lifelong management and has far-reaching implications for both physical health and psychosocial well-being [1,2]. The etiology of T1D is multifactorial, involving a complex interplay of genetic predisposition and environmental triggers, leading to a dependency on exogenous insulin for glucose regulation [2]. The incidence of T1D in children has been witnessing a steady increase globally, highlighting an urgent need for comprehensive research into its pathophysiology, early detection, and innovative management strategies [3].

This review aims to explore the accessibility in T1D interventions, namely Multiple Daily Injections (MDI), Continuous Subcutaneous Insulin Infusion (CSII), and Automated Insulin Delivery (AID) for the pediatric population, the challenges in management, and the potential pathways for improved care and quality of life for children and youth living with the condition. MDI is a regimen for managing T1D, involving several insulin injections daily to better mimic natural insulin release and offer dietary and scheduling flexibility, enhancing glucose control [4]. CSII, commonly known as insulin pump therapy, is a method of insulin delivery for managing T1D and involves using a small, portable device that delivers insulin continuously through a small catheter placed under the skin [4]. AID systems represent an advanced method for insulin administration, aiming to closely mimic the natural insulin release of a healthy pancreas by using continuous glucose monitoring (CGM) and insulin pump technologies in conjunction with a control algorithm to automate insulin delivery based on real-time glucose readings [5]. By delving into these critical aspects, this narrative review seeks to contribute to the growing body of knowledge which informs clinical practices and supports children and their families in navigating the complexities of T1D.

METHODS

The research question aims to take an extensive look into the pediatric population between the ages of 5-15 within the US and Canada. The initial literature search yielded results mainly from US and Canada, and thus the article will encompass a thorough investigation regarding the various interventions available in the treatment and management of T1D in these countries and will investigate the financial and geographical accessibility of these interventions for various pediatric patients. A literature search was conducted using databases such as PubMed, NCBI, and Web of Science, with key terms including Type 1 Diabetes (T1D), children, youth, United States of America (US), Canada, continuous subcutaneous insulin infusion (CSII), automated insulin delivery (AID), multiple daily injections (MDI), continuous glucose monitoring (CGM), geographical disparities, policy changes, among others. Upon discussing the current interventions available, the article will also highlight areas that require further advancement in accessibility in the scientific community, highlighting the shortcomings in current interventions.

RESULTS

Financial Accessibility

The financial challenges associated with managing T1D for children and youth significantly impact accessibility to essential treatments and technologies, highlighting the need for strategies to mitigate these barriers and enhance care. One study explored in this section investigated high costs of insulin with suggestions to improve access in the form of a policy review, and the second study analyzed data of 9,445 children with T1D showcasing the rising insulin prices and increased use of diabetes technology. The latter study addresses the rising costs of managing T1D for children but implicitly points to the critical accessibility and resource issue at hand for many families [6]. The most concerning financial burdens include the increase of average annual cost per patient from $11,178 to $17,060, the doubling of expenses for diabetes management technologies, and the significant rise in insulin prices [6]. These rising costs can create burdensome barriers for families, potentially leading to disparities in treatment and outcomes and hindering potential pathways for improved care of children with T1D. To ensure equitable access to the latest diabetes management technologies and insulin, strategies to control or reduce costs must be considered in policy formation. Addressing the financial barriers is essential to improve accessibility to high quality diabetes care, which is a pivotal step towards enhancing the overall quality of life for affected children and youth.

Moreover, the high cost of insulin is significantly influenced by market dynamics dominated by a few pharmaceutical giants, patent strategies, and complex healthcare systems particularly impacting accessibility to insulin in low- and middle-income households [7]. This is due to a few key stakeholders controlling the insulin market, namely the three big pharmaceutical companies: Eli Lilly, Novo Nordisk, and Sanofi [7]. These companies can charge higher prices for their insulin as there is little competition in the field, allowing for a few companies to dominate a large portion of the industry, eliminating the need to compete to maintain low prices [7]. Prices have risen as well due to the preference for expensive insulin analogs over their less expensive counterparts. This shift is attributed to the superior efficacy of insulin analogs, which react more quickly and maintain their presence in the body for extended periods, facilitating more effective glucose regulation than what is achievable with less expensive human insulin [7]. The study also addresses how pharmaceutical companies delay generic drugs by securing multiple patents and making minor modifications to existing drugs, impeding the availability of cheaper alternatives. Additionally, in the US, the high cost of insulin and the limited accessibility is due to the healthcare system. Moreover, insurance plans with high deductibles and copayments make it significantly more inaccessible for people to pay for their care. These problems are made even worse by government rules and regulations, such as with Medicare, a federal health insurance program in the US not being able to negotiate drug prices [7]. Thus, all these elements combined make it difficult for US citizens and other citizens from Canada that come from lower income households to regularly receive and pay for the insulin treatments they and their children require [8].

Geographical Accessibility

The current interventions that are widely used for the treatment of T1D for children highlight specific accessibility challenges, especially in terms of financial costs and for geographical disparities in the US and Canada, which necessitate targeted advancements in future interventions. The economic strain for T1D intervention is exacerbated in rural areas where access to affordable healthcare options is often limited [9]. Children in rural areas face challenges due to the long travel distances to healthcare facilities and specialized diabetes care centers. This affects the regularity of treatment and limits access to advanced care options and educational resources necessary for effective diabetes management [9]. Moreover, the remote locations of clinics in the US and Canada pose great difficulties for families who are forced to travel longer distances, often compromising the number of visits recommended by healthcare physicians. It is critical to improve T1D monitoring access ed by children who often depend on their caregivers to organize transportation for healthcare visits. This reliance can become particularly challenging in areas with significant geographical barriers, as coordinating travel around the caregivers' work commitments adds an extra layer of difficulty. Although there are interventions that can improve the frequency of visits for patients such as mobile healthcare units (MHUs), they must still be paired with accessible locations. A recent study evaluating the impact of telemedicine care coordination on access to care in Oregon's pediatric T1D population found that telemedicine significantly increased the number of T1D patients meeting goal visit frequency when paired with care coordination services [10]. This suggests that while telemedicine has the potential to mitigate access issues, its effectiveness is contingent upon adequate support systems being in place. Moreover, comparative analysis of urban versus rural access to T1D care reveals significant disparities such as limited availability of healthcare providers, longer travel distances to specialized clinics, and reduced access to advanced diabetes management technologies like CGM and insulin pumps in rural areas [10]. The study noted that prior to telemedicine expansion, patients in rural areas were less likely to receive adequately timed monitoring care compared to their urban counterparts [10]. This disparity highlights the need for targeted interventions to improve access in rural areas. In addition, the personal experiences of families navigating T1D care in rural areas provide valuable insights. Similarly, it was found that rural families may experience dramatic lifestyle changes post-diagnosis, with technology playing a crucial role in improving health management and flexibility [11]. These testimonials emphasize the challenges and adaptations faced by rural families in managing T1D.

DISCUSSION & FUTURE IMPLICATIONS

Future research should explore the long-term outcomes of digital interventions in rural settings and the impact of healthcare service inaccessibility on health outcomes. Investigating the barriers to the adoption of telemedicine and digital health technologies in rural areas can also provide insights on how to better tailor interventions to meet the needs of these communities. Amidst this exploration, policy changes targeting increased awareness of telehealth visit tools could be key to improving patient health outcomes by increasing engagement with the healthcare system, allowing for early screening of comorbid disease and mental distress [12].

When exploring the existing literature for age-appropriate accessibility implications of insulin delivery in children and youth diagnosed with T1D, it is evident that this area of research is lacking. Overcoming geographic and financial accessibility barriers are just as integral as ensuring safe delivery of insulin to these pediatric patients once they can regularly access them. Without sufficient research outlining the precise capacity for improvement in age-adapted insulin delivery methods, the most valuable and relevant available resources are recommendations from reputable academic and medical institutions regarding pediatric diabetic care. It is necessary to view these recommendations with the perspectives highlighted in the geographic and financial accessibility implications to better understand the foundational recommended guidelines in pediatric T1D care. According to recent reports, integrating a multidisciplinary team has proven to be an efficient, coordinated way to improve a child’s long-term health outcomes through simultaneous assessment of multiple patient health aspects, monitoring, and check-ins by nutritionists, social workers, doctors, and mental healthcare providers [13]. As children spend 30 to 35 hours every week at school, it is vital that there be school administrators and aides trained in supporting children with T1D ensuring that a proper diabetic care plan is carried out for any diabetic student [14]. One in every 300 Canadian children has diabetes, making it likely for there to be a minimum of one student with diabetes at any given school [14], reinforcing the importance of staff training so they may be aware of potentially undiagnosed diabetes or hypoglycemic complications occurring in real-time [14]. An equally concerning statistic supporting this change is that children aged less than five years old and early school-aged children make up the “fastest growing segment of the population” with T1D, further emphasizing the need for trained staff at all schools [14].

A holistic diabetic care plan would include an appropriate diet, constant glycemic monitoring, and insulin delivery alongside care from the multidisciplinary team, parents or guardians, and school staff [13, 15] . Children with T1D are recommended to perform blood sugar level checks at a minimum of four times daily depending on their individualized care plan, with others using CGMs reporting their levels every 5 minutes [14] . Younger-aged children who have not yet reached independence in fulfilling decision-making skills and everyday tasks may not be capable of checking their own blood sugar levels or ensuring their proper use of insulin delivery methods [14]. Policies and resources tailored towards these barriers must address future directions and recommendations to strongly improve the patients’ health outcomes, requiring consistent and safe diabetic care at even the youngest age group once diagnosed. Extremely young children diagnosed with T1D could greatly benefit from a CGM or AID device, allowing their families to adjust insulin delivery as per the child’s needs [15], if they are able to financially access such diabetic management tools. Though hybrid closed loop (HCL) insulin pumps used in conjunction with AID devices are typically provided to older children with T1D, they have been limited in use to clinical trials for children between the infant and preschool ages. As per the SickKids foundation, the blood sugar target range is 6-10 mmol/L for children under 6 years old, 4-10 mmol/L for children between 6 to 12 years old, and 4-8 mmol/L for children ages 12 years old and above [16]. This data emphasizes the necessity to expand HCL/AID system trials to cater to the precise and varying needs of these younger demographics. By doing so, one can ensure broader access to life-enhancing technologies and alleviate the financial strain on families, making a pivotal step toward equitable healthcare solutions for all children with T1D.

Caregivers arguably play the most integral role in the diabetic child's self-management skills and care journey over the years as they grow into complete independence [14]. Access to healthcare professionals or diabetes educators who can teach them strategies on easing the child's anxiety or pain with insulin delivery systems or frequent blood checks are crucial, so that the daily insulin delivery is not frightening nor unnerving, using distinctly age-appropriate calming strategies [15]. One example of a behavioural strategy may be 'think aloud', where the child can be verbally asked about which one of two injection sites they would like the insulin to be delivered, providing them an opportunity to feel ownership over their own health management [15]. A "supportive and emotionally warm parenting style" can promote these children's overall quality of life, thereby directly improving their long-term health status as well [15]. If the child uses MDI or CSII, there could be physical ways to manage pain in addition to these verbal and psychological standpoints, such as topical anesthetic cream applied before the insertion of a subcutaneous catheter insulin port for both injections and infusion [15]. These strategies must be considered when the preferred insulin pump technology is unaffordable or unavailable, causes negative skin reactions, or if the local healthcare provider in charge of delivering insulin to the pediatric patient is not experienced with their given age group [15].

With regards to financial accessibility and insulin prices, a 2021 study examining the high costs of insulin emphasizes the need for a strategic plan to lower them, which will require individuals from different fields to collaborate, which include policy makers in government and healthcare organizations such as Medicare [7]. The authors recommend bringing generic drugs and biosimilars to the market and advertising them to increase competition, which could lead to lower insulin prices for families. To prevent brand-name drug monopolies from perpetuating high insulin prices, the current patent systems and market exclusivity rules need to be re-evaluated and changed [7], making it easier for cheaper insulin options to enter the market. This study also emphasizes the importance of eliminating practices which harm competition, such as "pay-for-delay" deals that lead to difficulty in acquiring cheaper insulin variants [7]. Another vital recommendation involves enhancing the transparency of insulin pricing by clarifying the complex pricing structures so that families can better navigate the system and identify opportunities for financial savings [7]. The study advocates for policy reforms that would authorize government initiatives, such as Medicare, to directly negotiate drug prices with pharmaceutical firms, potentially reducing prices significantly [7]. It also calls for regulatory reforms to prevent pharmaceutical companies from exploiting consumers through practices such as filing numerous patents for the same medication and making minor modifications to extend patent protection [7]. Additionally, it highlights the advantages of adopting value-based pricing strategies and insurance models, supporting a pricing system that connects the cost of insulin with its clinical effectiveness and patient benefits [7]. In this manner, patients' out-of-pocket costs are more in line with the medication's therapeutic value.

Furthermore, when exploring other promising areas which could improve the quality of life for affected patients, telehealth emerges as a transformative approach in treating T1D by offering better care than during traditional in-person visits, enhancing convenience and accessibility to healthcare providers. By integrating advanced technologies like machine learning, telehealth not only improves the management of diabetes-related complications, but also supports mental health needs, making it an invaluable tool in the holistic treatment of T1D [17]. Telehealth has also been recognized as a valuable tool in managing the psychosocial impact of T1D in young people. Young individuals with T1D often face higher rates of psychological distress, including periods of burnout and challenges in coping with the daily management of their condition [17]. Family, peer, and psychological support, coupled with education on living with diabetes, are crucial for reducing distress and enhancing overall well-being and diabetes management [17]. Psychological screening tools and appropriate local referral pathways are recommended to ensure that young patients receive the mental health support they need. Various psychological and behavioral interventions – such as solution-focused therapy, coping skills training, motivational interviewing, cognitive behavioral therapy, and family-centered interventions – have been shown to significantly improve the quality of life and diabetes management in young individuals with T1D [18]. Furthermore, telehealth has shown to be particularly effective for younger patients with T1D, enhancing their engagement and adherence to treatment plans. A study on the CoYoT1 Clinic, a telemedicine appointment care model, demonstrated that young adults with T1D who participated in this program had significantly higher clinic attendance rates, met American Diabetes Association guidelines for appointment frequency more consistently, and reported greater satisfaction with their care compared to those receiving standard care [19]. This indicates that telehealth can effectively keep young adults engaged in their diabetes management during critical transition periods, closing the accessibility gaps caused by geographic and potentially financial challenges. However, some challenges have been noted in the literature, including disparities in access to telehealth services due to socioeconomic factors, digital literacy barriers, and potential limitations in the quality of remote care compared to in-person visits. Understanding these drawbacks is essential to optimizing telehealth's effectiveness and ensuring equitable healthcare access for all pediatric patients with T1D.

CONCLUSION

In conclusion, this review has discussed the challenges and critical needs in managing T1D in the pediatric population from ages 5-15 across the US and Canada. It is evident that while there are several effective treatments available, such as MDI, CSII, and AID, their accessibility is significantly hindered by a variety of factors [6,20]. Financial constraints remain a primary barrier, as the escalating costs of insulin and diabetes management devices place a substantial burden on families, particularly those with limited economic resources [6]. Geographical disparities further exacerbate these challenges, with children and youth in rural areas facing considerable difficulties in accessing specialized diabetes care due to long travel distances and a lack of local healthcare resources [20]. Moreover, the need for age-appropriate and emotionally supportive care emphasizes the necessity for a comprehensive approach to T1D management in children and youth. The review highlighted a pressing need for innovations in T1D care that are not only more cost-effective but also widely accessible, regardless of a patient's location or financial status. These interventions must be designed with an understanding of the diverse needs of the pediatric population, ensuring that every child with T1D can receive comprehensive, effective, and empathetic care that supports not only their physical health but also their overall well-being.

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VIRTUAL REALITY AS AN EFFECTIVE SOCIAL SKILLS TRAINING PLATFORM FOR IMPROVING SOCIAL AND BEHAVIOURAL INTERACTIONS OF AUTISTIC CHILDREN: A NARRATIVE REVIEW

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ABSTRACT

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by challenges in socio-pragmatic interactions and repetitive behaviors. Current research supports the use of tailored behavioral interventions to effectively enhance the well-being of Autistic individuals and their families. Traditional social skills training (SST) is a commonly used intervention in addressing social and behavioural deficits often associated with ASD. Given the barriers faced by autistic individuals when accessing traditional forms of SST, researchers have begun to investigate alternative, more accessible delivery mediums, including virtual reality (VR). The aims of this review are twofold: (1) to investigate whether VR is an effective tool through which SST can be delivered to autistic individuals; (2) to determine the impact of VR environments on the real-world social interactions of autistic children when used as a platform for SST. The results of this study demonstrate that VR is a promising, dynamic, accessible, and effective practice for the development and support of a range of social skills in ASD individuals.

Keywords: autism spectrum disorder (ASD), virtual reality (VR), social skills training (SST), social skills, emotion regulation, cognitive skills

INTRODUCTION

Defining ASD, Symptomatology, Etiology, and Prevalence

Autism spectrum disorder (ASD) is characterized by deficits in social communication and the presence of repetitive behaviors, interests or activities, as outlined in the DSM-5 [1]. The risk of ASD can be partially predicted through genetic factors, which also results in greater incidence in males [2]. As of 2023, ASD affects approximately 0.72% of the global population, though underdiagnosis remains a concern, particularly in developing regions [3]. In this article, we use “autism,” “autistic individuals,” and “ASD” interchangeably, aligning with a growing body of literature that discourages person-first language due to its potential to increase stigma [4].

Educational Interventions and ASD Treatment

At present, there is no singular accepted treatment for ASD. However, current literature supports the application of individualized behavioral and educational programs to positively impact the lives of autistic individuals and their families. As autistics' relational modalities appear one-sided, assistance in forming relationships is needed [5]. Studies have demonstrated that autistic individuals can learn to act in social situations if they have the opportunity to regularly participate in scenarios that mimic common daily interactions [6-8].It has also been shown that traditional educational interventions (e.g. social skills groups, video modeling, peer-mediated instruction) [9] for individuals with ASD can be expensive, inaccessible, and inefficient as a result of limited resources and low motivation on part of the individual [10,11].

Social Skills Training and Symptom Management

Traditional social skills training (SST) is a common intervention to help address social deficits associated with ASD [12]. This behaviour therapy involves modelling social interactions through face-to-face interventions to improve communication skills. These programs primarily rely on interactions between the child and a facilitator to reinforce positive social behaviours. In therapies involving operant conditioning, direct and explicit directions are provided to reduce repetitive or restrictive behaviours. This approach can increase skills (e.g. eye contact) which may be utilized in realistic social settings [13]. Due to the wide variety in symptomatology with ASD, the types of appropriate social skills training can vary with the age and functioning level of the individual. Current SST involves pivotal response training or prompts from facilitators supplemented with environmental modifications. SST can also involve interactions with other children through peer mentorship and with peers in large groups [14].

Application of Virtual Reality in Social Skills Training

Technology-driven therapies for ASD, including robotics [15-17], mobile apps [18-20], interactive video modeling [21-24], and virtual reality (VR) [25,26], are rapidly increasing in popularity. Autistic individuals in particular have shown interest and adherence towards learning through computerized programs, such as VR, due to its capacity to change environments and modulate to specific sensory levels. VR technology aims to create an interactive computer-generated virtual environment by combining hardware and software to provide individuals with the feeling of presence and an immersive environment [27]. Favourable properties of VR include cost-effectiveness, as evidenced by numerous quantitative evaluations of VR-based therapies as compared to traditional therapies, across various phycological disorders [28, 29, 30, 31]. Another favourable property of VR is the high self-determination index (SDI) for autistic individuals to engage in this form of SST. SDI is determined by subtracting mean external motivation from mean internal motivation. Therefore, since autistic individuals tend to have a higher mean internal motivation towards VR technology, they will be more likely to engage in VR-based SST [32]. Finally, the relative accessibility of VR has produced a large body of research targeting autism-related disabilities with this technology [33-36].

As such, this review focuses on the impact of VR-based SST on autistic children in North America. Traditional SST methods remain expensive and resource-limited [37], while VR offers a promising, scalable alternative. Given the cultural variability in ASD presentation, we examine VR's role in facilitating social development in North American children and its potential as a future ASD treatment [38].

METHODS

Literature was collected by conducting targeted searches on databases including Ovid Medline, PubMed, Web of Science, and CINAHL using a range of key terms.

EFFECTIVENESS & EFFICACY OF VR USE IN SOCIAL SKILLS TRAINING FOR AUTISTIC YOUTH

VR is a promising, dynamic, accessible, and effective practice for the development and support of social skills in ASD individuals [14,39]. A systematic review and meta-analysis by Karami et al. on the effectiveness of VR for the rehabilitation and training of ASD individuals [39] found that VR held varying degrees of effectiveness when targeting different social and life skills. From reviewing 26 uncontrolled and nine controlled trials, the authors concluded that "VR technology can be a viable tool for designing interventions aimed at enhancing and improving different skills" in autistic people at any age. The results of their analysis found the strongest effect of VR on improving daily living skills (i.e. shopping, driving, street crossing, and job interview skills), while moderate effects of VR were found in improving cognitive skills (i.e. attention and concentration, reasoning and problem solving, executive function, language, and metacognition), emotion regulation and recognition skills (i.e. emotion expression, affect recognition, stress, and anxiety management), and social and communication skills (i.e. social adaptation and interaction, communication, social reciprocity, social responsiveness, negotiation skills, and theory of mind). The same paper also found that performance on tests evaluating the aforementioned skills improved as age increased, but observed a substantial decline in the effectiveness of VR training on individuals diagnosed with some form of comorbidity alongside ASD. Ultimately, the results of the study showed promising efficacy of using VR in the various skills training of autistic individuals when compared to more conventional training programs, particularly when used to improve the effectiveness of daily living skills.

Another systematic review by Satu et al. on the use of immersive VR in neurodevelopmental disorders [40] found that immersive VR was mostly used in connection with ASD for the assessment of and intervention in social skills. From a review of 34 studies, the authors found that ASD intervention studies focused primarily on addressing social communication (i.e. improving nonverbal communication and social interaction, verbal and social communication, emotional and social adaptation skills, recognition of basic emotions and social skills, and joint attention skills); their results suggest that immersive VR "makes it possible to practice complex social skills in a controlled situation close to daily life." Erb, in their scoping review on the use of VR and augmented reality (AR) to teach various skills to children and youth with comorbid ASD and intellectual disabilities (ID), [41] further asserts that VR facilitates this practice because autistic individuals can practice their socio-emotional skills in a highly individually-relevant environment, without the "fear, anxiety, and potential consequences of real-life." [41] The review of 20 studies found that (given that autistic children and youth diagnosed with both ASD and ID typically demonstrate difficulties in initiating and maintaining social interactions) the use of VR and AR technology has been found to be a useful medium through which autistic individuals can practice various skills by simulating "real-world experiences in a safe and controlled environment.” [41] It is important to note, however, that the addition of AR adds complexity to the accessibility of this technology, particularly when compared to VR technology only. This can have further implications for the accessibility of VR / AR technology and social skills training for Autistic youth. [42] Overall, Erb concluded that the use of VR / AR demonstrated positive results with respect to teaching autistic individuals social skills, in addition to the generalization of such skills to real-world scenarios [41,43].

In conclusion, the exploration of VR as a tool for the rehabilitation and training of ASD individuals reveals promising avenues for enhancing various skills crucial to daily life. The remainder of this paper will investigate the effect of VR usage in addressing different basic and complex skills relevant to ASD.

APPLICABILITY OF VR IN SYMPTOM MANAGEMENT FOR ASD

The purpose of this section is to define some of the most common phenotypes and symptoms associated with ASD and how VR can be utilized as an alternative measure of treatment. We investigated whether VR could improve the measure of various symptoms over time, and whether there was a significant difference when compared to traditional, in-person forms of SST.

Stereotyped or Repetitive Motor Movements (stimming)

Stimming is a core characteristic or symptom that dictates the diagnosis of ASD [44]. The effects of stimming can affect real-world social interaction through repetitive motor movements and repetitive verbal language [44]. Stimming is defined as repetitive and unregulated motor movement [45]. Unmanaged, it can hinder engagement in activities, often requiring intervention [45]. Enriching the child’s environment has been found to be an effective method for practitioners and parents to reduce stereotypic behaviour in some children. This is most successful when there is a strong match between the sensory input provided by both the alternative and original stimuli [46]. In this case, VR technology provides stimuli meant to mimic real-world social interaction, which is the original stimuli. As VR technology advances and becomes more realistic, the gap between social interactions in VR vs. the real-world becomes smaller. Hence, virtual reality can be an effective tool for the management of stimming, since its immersive qualities can match the sensations felt from both stimuli.

A review from Chen et al. explores the efficacy of VR technology for social skill development in autistic. The study found that VR provides ASD individuals an inexpensive medium to learn, a motivating environment to practice skills, and a personalized setting where learners feel safe due to it being a controlled, familiar environment [47]. Another study by Frolli et al. found VR could decrease stimming but reported no significant difference between traditional SST and VR groups in reducing these behaviours [14].

Abnormalities in Eye Contact

One of the most common ASD symptoms is difficulty maintaining eye contact. Autistic children tend to have atypical viewing patterns because they show a greater fixation towards nonsocial objects than humans face during social interactions [48]. Evidence suggests that individualized interventions like VR immersion can address core vulnerabilities related to the lack of eye contact for ASD individuals. A study by Lahiri et al. found that VR provides a lower-anxiety platform for practicing social interactions, leading to a significant increase in the time spent fixating on faces among adolescent participants. Furthermore, this article also highlights that VR can be used to communicate with individuals from different parts of the world, which can improve eye contact when socializing with others from diverse backgrounds [49].

Deficits in Emotional Expression and Recognition

ASD is typically characterized, among other criteria, by deficits in social-emotional reciprocity [49,51]. These deficits often manifest as the reduced sharing and expression of emotions, as well as difficulty recognizing and interpreting the emotions of others [50-52]. Through social skills training and operant conditioning, autistic children are trained to associate facial expressions to specific emotions. According to a study by Didebhani et al., VR-based interventions may be able to improve the ability of children to recognize the emotion of others through facial expressions and tone of voice [53]. In this study, thirty children between the ages 7-16, completed two hours of VR training every week for five weeks, and had their emotion recognition skills observed. The results showed that VR resulted in an increase in emotion recognition, social attribution, and executive function of analogical reasoning, which suggests that VR interventions are effective for improving social impairments associated with ASD [53]. A study by Yuan et al. explored similar topics, investigating the efficacy of a VR-enabled training program on social and emotional skills for primary school autistic children. After six 1-hour training sessions, the authors found significant improvements in emotional expression and regulation, as well as a significant positive effect on social reciprocity [54]. Finally, a study by Frolli et al. compared a traditional emotional training procedure performed with a therapist to a training procedure that was VR-based [14]. The findings suggested that VR-based and traditional interventions have similar acquisition times for the recognition of primary emotions. Moreover, the VR-based intervention even showed shorter acquisition times for tasks requiring the use of both primary and secondary emotions. Overall, these collective findings support VR-based interventions as a promising asset to traditional emotional training and therapy methods.

Absence of Interest in Peers

Displaying little to no interest in other children has long been identified as a behaviour that can help differentiate autistic children from those without, especially in early years of life [55]. Moreover, ASD individuals have been found to be less likely to initiate social interactions with same-aged peers [56,57]. These behaviours can persist throughout an autistic individual's lifespan, and result in the inability to create stable friendships or relationships [58]. A recent exploratory study conducted by Ke et al. found that a VR-based social skills training program could lead to an increased level of social skills performance in autistic children, including significant improvements in the frequency and success of social interaction initiation [59]. This study provides preliminary evidence supporting the usage of VR-based interventions to increase social interest and engagement in autistic children.

DISCUSSION

Benefits of VR Interventions

VR interventions are crucial to SST for autistic children as they not only facilitate a safe, low-risk environment, but they increase engagement and help with the generalizability of said social skills to real-life scenarios. A traditional behavioural skills training intervention called video modeling is a type of observational learning where a person (an adult, a peer, or the student themselves) is recorded engaging in the targeted social skill (e.g., waving, putting up their hand in class, playing with their peers). Children who need to improve on those social skills will watch these recordings with a facilitator who will point out the important social skills presented as well as answer any questions [60]. While this has been an effective SST intervention [61] for teaching new behavioural skills in similar environments, it is not a sufficient long-term SST intervention because it does not effectively help children apply these behaviours when presented with a context different from the rehearsed task scenario [62]. This ineffectiveness of the intervention is emphasized in autistic children as they have a decreased ability to generalize knowledge from one context to another [62]. Thus, VR interventions are being explored to address this limitation.

Since VR tools inherently use electronic devices, it is important to highlight previous research on autistic children's relationship with technology in the context of learning and education. Computer-assisted instruction and electronic screen media (e.g. animated programs)[10] have been shown to be more accessible for autistic children as a majority of autistic children show comfortability with technology as well as show a reduction in 'problem behaviours' when using technology, allowing them to have an increased level of engagement with these types of interventions [63]. Additionally, ASD individuals might prefer the characteristics of computer-generated speech (e.g. monotone, less variable) because of their desire for "sameness." [63,64] Although advancing VR technology may introduce more varied voice outputs, the controlled environment of VR still provides a structured, low risk setting for SST.

In addition to the high motivational index that is created when using VR interventions for ASD individuals, virtual environments can encourage flexibility and generalizability by providing real-life role-play situations. In a pretest-post test study, Didehbani et al. investigated a group of autistic children immersed in VR-based social skills training involving role-play with avatars in daily life social scenarios. The results showed an improvement in the following measured outcomes: emotion recognition, social attribution, and executive function [53]. The study also mentioned how these VR interventions provide a platform for repeated practice of the same behavioural/social skills across different scenarios. The need for learning various applications of the same social skill is essential and can be accomplished with VR tools as they move away from rote learning and help facilitate the generalization of social skills from those learned in VR to those used in daily interactions [53]. While concerns exist regarding technology-based interventions promoting social isolation, research suggests that interactive behavioural intervention technologies like VR offer flexible, generalizable social skills practice in a safe setting for ASD individuals [25,66,67].

Limitations of VR Interventions

While the use of VR holds many benefits, it may also present its own limitations. Difficulties in accessing VR interventions, such as financial barriers / startup costs (e.g. purchasing softwares, headsets, controllers) and ongoing expenses related to maintenance, upgrades, and technical support, may pose barriers for some schools and families in accessing these interventions. Additionally, the individualized, "no-one-size-fits-all" nature of ASD means that interventions effective for one child may not necessarily be effective for another; this may make it challenging to create universally beneficial VR programs for social skills training for all ASD individuals. There is also a concern about the potential for overreliance on VR as a sole intervention method, which could limit exposure to diverse social situations and interpersonal dynamics that are essential for holistic social development. Further, the effectiveness of VR-based interventions may be influenced by factors such as the child's comfort level with technology, sensory sensitivities, and cognitive abilities, which need to be carefully considered and addressed in the design and implementation of VR programs for ASD individuals. Despite these limitations, ongoing research and advancements in VR technology offer opportunities to refine and tailor interventions to better meet the needs of ASD children and improve their social skills development.

Looking ahead, the integration of emerging technologies such as artificial intelligence (AI) and brain-computer interfaces (BCIs) could further enhance VR-based SST interventions. AI-powered VR could create adaptive learning experiences by responding dynamically to a child's progress, providing personalized feedback and adjusting scenarios in real time based on behavioral cues. AI-driven emotion recognition can also enable virtual avatars to display naturalistic facial expressions, tone, and social cues, making social interactions more authentic. Meanwhile, BCIs could revolutionize VR interventions by offering deeper insights into neurological responses during social training, allowing for highly personalized interventions tailored to individual cognitive and emotional needs.

While these advancements hold promise, they also remain susceptible to many of the current limitations facing VR technology today, including accessibility-based and ethical concerns. Nevertheless, ongoing research and technological innovations continue to refine the capabilities of VR in social skills training, offering exciting opportunities to improve interventions for autistic individuals.

CONCLUSION

The studies reviewed in this article underscore VR's efficacy in improving social, cognitive, emotional regulation, and daily living skills among autistic individuals across different age groups. VR's ability to simulate real-life scenarios in a controlled environment offers a safe and effective platform for practicing and generalizing socio-emotional skills. The accessibility and motivational appeal of VR hold great potential for addressing the challenges faced by ASD individuals, particularly when traditional interventions prove costly, inaccessible, or inefficient.[9] Some of the primary barriers to accessing traditional forms of social skills training is the need for facilitator, accessibility/requirements of a clinician, transportation, and time-intensive training, which can be addressed by the use of VR [12].

Initiatives aimed at integrating VR into educational curricula and therapeutic interventions may provide opportunities for schools and families to access funding specifically designated for acquiring VR technology. Future research should aim to explore the long-term effectiveness of VR interventions for autistic individuals, as well as investigate the optimal methods of incorporating VR into existing therapeutic practices and educational settings [68]. This research will not only contribute to the optimization of VR-based interventions, but also enhance our understanding of how technology can best support the diverse needs of ASD individuals across different developmental stages and functional levels.

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SELF-ADVOCACY DEVELOPMENT IN ONTARIO ADOLESCENTS WITH LEARNING DISABILITIES

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ABSTRACT

Learning disabilities (LDs) affect 3.2% of children in Canada and encompass a range of conditions impacting an individual's psychological learning processes. Self-advocacy, an individual's ability to speak up for themself, is a critical component of accessing supports and accommodations to manage LDs, especially as a child transitions throughout and beyond secondary education. This review will explore the development of self-advocacy in children with LDs through Ontario's existing support and skill development programming, systemic barriers to accessing self-advocacy supports, as well as stigmatization towards LDs. Ontario's current support infrastructure is a mix of resources that aim to increase students' ability to navigate life with their LD. These aims align with well-documented barriers hindering students' development of self-advocacy, such as attitudinal and societal factors, which stem from a lack of understanding surrounding LDs. Widespread stereotyping and stigma manifesting in depersonalization and social rejection further impact their relationship with their LD diagnosis and their overall perception of self. Overall findings highlight the need for research to gain a nuanced understanding of the gaps within Ontario's current system to more effectively address the challenges that adolescents with LDs face in developing self-advocacy skills.

INTRODUCTION

Learning disabilities (LDs) are a broad classification of conditions impacting an individual's ability to perceive, think, remember, or learn [1,2]. LDs are distinct from intellectual disabilities, as the former impacts an individual's psychological learning processes while maintaining typical thinking and reasoning skills, whereas the latter does not [3]. 3.2% of children in Canada are reported to have a LD, which represents 59.8% of all children with a disability [4]. Dyslexia is the most prevalent LD, impacting one's phonological processing and decoding, which often, in turn, hinders one's reading comprehension skills [1]. Other examples of LDs include dysgraphia and dyscalculia, referring to impairments in an individual's handwriting and mathematical processes, respectively.

LDs can have a significant impact on a child's development. Treatment and interventions intended to support specific needs can help children better manage their LDs. Broad focuses of intervention approaches include accommodations, such as additional time for work, and self-advocacy development, where students learn to communicate their needs [3].

Within the Ontario education system, the Education Act requires school boards to provide special education programs and services to students identified as "exceptional pupils" [5]. The act defines exceptional pupils as students who have been determined by an identification, placement and review committee (IPRC) as having behavioural, communicational, intellectual, physical, or multiple exceptionalities. Ontario special education programs for these students are continually modified based on their assessment and evaluation results and include an Individual Education Plan (IEP), with specific objectives and an outline of educational services that meet their unique needs. In early education, assessment and accommodations for LDs are primarily decided by educators and caregivers [6,7]. As a child progresses throughout their elementary, secondary, and post-secondary schooling, they can take on a greater role in obtaining accommodations and resources for their LD in Ontario.

Self-advocacy, referring to an individual's ability to speak up for themselves, is a critical component of self-determination, one's ability to make meaningful, personal decisions [8]. In the context of individuals with LDs, self-advocacy entails educating others about one's LD and how it impacts one's life [8-10]. Becoming an effective self-advocate requires communicating their needs with confidence and appropriately explaining requested accommodations to assist in the classroom. Stigma, a broad term referring to negative beliefs about an individual based on a certain characteristic, can have implications on adolescents' desire and ability to self-advocate as it confirms stereotype threat, defined as a specific situation where an individual fears confirming those aforementioned negative beliefs [9]. Both stigma and stereotype threat are important factors to consider in the context of self-advocacy and with anything in relation to social development, given that they act as key drivers of influence within a increasingly social setting such as the grade-school system.

The transition many adolescents in Ontario experience from elementary, to secondary, to post-secondary education (PSE) encompasses a critical opportunity for the application of self-advocacy skills. These skills become central in navigating education systems with autonomy and increased decision-making opportunities, despite pre-existing stigma and stereotyping.

An awareness of one's rights is vital to self-advocacy. It is important for adolescent students with LDs to be aware of their rights to maximize their learning experience, make decisions about pursuing PSE or other vocational training, seek career opportunities, manage daily living demands, and effectively advocate for their needs [10]. This article addresses the following research question: In adolescents (aged 10-19) with learning disabilities in Ontario, how might support programs, existing barriers, and stigmas influence the development of self-advocacy skills within their educational experiences? By exploring the development of self-advocacy skills in adolescents with LDs, the primary aim of this review is to act as an initial step towards guiding future research and system changes within students' self-advocacy skill development. Ultimately, this work contributes to a growing body of literature on inclusive education by identifying gaps in current support systems and offering insights for future research and systemic improvements.

METHODS

Searches for this review were individually conducted for the following elements: Ontario's support programming, barriers to self-advocacy, and stigma, with overarching terms including "learning disabilities" and "self-advocacy." The resources employed for this review were the Canadian Journal of Disability Studies, ERIC, Web of Science, Journal of Learning Disabilities, and OVID MEDLINE. Sources from grey literature searches, such as provincial governmental and Ontario school board reports, were also employed. Evidence was limited to English-language publications and focused on Ontario, but no limits were placed on the year of publication to gain a comprehensive view of LD treatment.

EXISTING PROGRAMS

The Learning Disabilities Association of Ontario (LDAO) provides resources catered to different learning styles and help develop self-advocacy in students with LDs [11]. For instance, the "Learning Styles for Youth" is an online course that enables adolescent students to identify methods that best support their learning inside and outside of school [11]. This workshop takes a broad approach to provide students with a foundational understanding of various learning styles, with a unit focusing on strengthening alternative ways of learning that might be unique to their LDs. Alternatively, the "Self-Advocacy for Youth" online workshop is a self-paced, self-directed resource to teach students how to effectively communicate the ways in which their LD affects them, and to self-advocate for the supports they need to optimize their learning [11]. To address misunderstandings of one's LD, unjust treatment, and discomfort with seeking help, this workshop provides students with information on their LDs, strategies to be an effective self-advocate, and their legal rights within the education system.

The transition between secondary and PSE is a pivotal stage for self-advocacy development, as system-level supports for LDs change significantly between these stages. Instead of mandated interventions present in earlier schooling, Ontario PSE requires students to identify and register themselves to receive accommodation [12]. Recognizing the need for self-advocacy in this process, the development of transition programming for students with LDs has been an ongoing process. The Summer Transition Program (STP), developed by the Learning Opportunities Task Force in 2006, is now carried out at all publicly funded Ontario colleges and universities [13]. The STP focuses on LD-specific campus orientation, awareness of one's condition, learning strategy skills, assistive technology training, and self-advocacy skill development [12]. Programming varies based on individual curriculums, with in-person and online options in addition to program lengths from one day to eight weeks. An evaluation of this program was conducted through a longitudinal cohort study with 151 postsecondary students at Durham College's and the University of Ontario Institute of Technology's Disability Centre [12]. Qualitative data from focus groups and interviews found positive perceptions of the STP, allowing students to become better acquainted with their institution's accessibility services, for example. Participation in the STP was overall correlated with an increased willingness for students to self-advocate and access student services. Thus, the intervention's focus on promoting self-awareness and awareness of their LD was particularly emphasized as significant in improving PSE students' self-advocacy skills [12].

SYSTEMS & BARRIERS

The Ontario Universities Accessible Campus defines

defines organizational and systemic barriers as practices and policies that impede individuals from participating fully. For students impacted by LDs, this can involve limited avenues by which students can showcase their understanding of course content [14]. Students with disabilities within the Ontario school system have expressed the need to address these persisting barriers [15]. The development of self-advocacy skills represents an area with several systemic gaps for students with LDs, including lack of involvement in students' IEP, lack of self-awareness of their disability, self-determination barriers, and teacher attitudes.

According to Regulation 181/98 of Ontario's Education Act, students at least 16 years of age have the right to participate in IPRC and be present in the committee's placement or identification [16]. Although the Ontario Minister of Education recognizes these meetings as a critical opportunity for students to develop self-advocacy, the development largely falls on students' willingness to participate [17]. Research suggests that adolescents with LDs face barriers during IEP meetings due to a lack of clarity surrounding educator's jargon-filled statements with minimal explanations and incomplete disability-related information [18]. Additional barriers for students to represent themselves at IEP or transition meetings stem from a lack of emphasis on preparing students to self-advocate and share their LD-related needs prior to said meetings [19,20].

The transition into PSE has further implications on self-advocacy. The Ontario Human Rights Commission's (OHRC) Disability Policy describes how students are responsible for registering with their institution's accessibility services office, participating in discussions surrounding accommodations, actively engaging in the accommodation process, and informing course instructors of their accommodations [21]. As students are typically 18 years of age during this transition, they are considered legal adults and can complete the process independently without requiring consent from a parent or guardian. However, they may choose to involve their parents or guardians, but this participation is allowed only with the student's consent. Students often face barriers in this process, primarily due to a lack of self-awareness about their LD and the need for self-advocacy. Without this awareness, they may struggle to recognize their specific challenges, articulate their needs, or seek appropriate support. These barriers can include difficulty recognizing their specific accommodations, fear of stigma, and uncertainty about the resources available to them [22]. This paper further expands on the implications of fostering a positive relationship with oneself, which can be enhanced by heightened self-awareness and understanding of one's own needs. An adolescent's understanding and comfort with their LD can shape their willingness to articulate their personal strengths and limitations within academic settings [22]. This can manifest as a heightened desire to complete academic work without assistance, where students may value independence to the extent that they are less willing to seek help or self-advocate. This reluctance to ask for support may stem from a belief that doing so could be perceived as a lack of capability or undermine their sense of self-reliance. Consequently, students might avoid using resources or accommodations that could aid their academic success [22]. Furthermore, the accessibility of programming designed to reduce these gaps is made complex through additional barriers. For example, thematic analysis from student evaluations of the STP identified time constraints and educator knowledge as significant factors influencing participation.11 Specifically, students who worked during summer or attended secondary schools with limited knowledge on transition programming faced more difficulty accessing these supports and subsequent opportunities for self-advocacy development [12].

The perspective of Canadian educators on the inclusion of children with developmental disabilities in educational settings was highlighted in a study by researchers from Ontario PSE institutions. Inclusion, in the context, refers to the practice of integrating students with developmental disabilities into general education classrooms and ensuring they receive appropriate support to participate fully in the learning environment [23]. Findings highlighted how teachers' attitudes surrounding disabilities have direct implications on their effectiveness at building inclusive classroom settings [23]. The research demonstrates that teachers with positive attitudes towards inclusion in the classroom have a greater likelihood of meeting the needs of students with disabilities. As primary and pertinent figures in students' lives, the evolving attitudes and awareness of educators have direct impact on students with LDs, thus translating to how students might view themselves and how they are motivated to obtain their needs as they develop. As teachers are equipped with different tools and perspectives, serving as a wealth of resources that students with LDs themselves might not be able to access, students might also come to know what works for them, and seek the same type of aid, with the greater hope that they would be able to voice these needs themselves. Therefore, it is important to foster knowledge and competence needed of classroom inclusion amongst teachers to appropriately support students with LDs and to minimize the barriers for students with respect to the improvement of their self-advocacy abilities [23].

STIGMA & STEREOTYPES

The vulnerability of adolescents with LDs to stigmatization and stereotyping has significant implications on the development of their self-advocacy skills. As individuals begin to navigate the education system, a LD diagnosis itself can be labelled as 'inherently weaker' compared to students without a LD, providing a basis for differential treatment by peers and educators [9]. This stigmatization can lead parents and educators to have lower educational expectations of an adolescent with a LD compared to their peers, thereby perpetuating stigma consciousness. Haft et al. define stigma consciousness as individuals expecting to be treated differently due to pre-existing stigma [9]. Stigma consciousness encompasses an individual's perception of societal views of their identity and directly impacts the extent to which individuals internalize negative beliefs and stigmatized views. Similarly, pressure to conform to the stereotypes associated with LDs is shown to impair achievement motivation, thus hindering individuals' self-advocacy abilities. This internalized stigma can be even more harmful than discrimination and negative interactions with peers, as it is associated with demoralization, feelings of hopelessness, and deterred professional help and accommodation [24].

Stereotyping attitudes towards individuals with LDs are defined as overgeneralized and simplified perceptions about the attributes for a minority group, and are further broken down by Kourdotis et al. into categorization attitudes (viewing a minority group as homogenous), depersonalization attitudes (stripping individuals of human qualities), and denigration attitudes (stereotyping a group according to the stereotyper's values) [25]. In a 2010 study, students from two research universities were provided questionnaires about stereotypes in individuals with LDs [26]. Low ability was identified as the most consistent metastereotype (stereotypes made within stereotypes), and many of the metastereotypes from the study directly parallelled the attitudes highlighted by Kourdotis et al [25,26]. Nearly half of participants reported believing that the general population thought individuals with LD were less intelligent than those without [26].

Attitudes toward individuals with LDs are further complicated by the fact that they may be stigmatized as having decreased social and interpersonal skills, leading to social rejection.26 Societal labels further perpetuate stigma consciousness, since it is the consequences of the LD, rather than the LD itself, that are visible. According to a 2021 study by Pfeifer et al. on the self-advocacy of individuals with LDs in the fields of science, technology, engineering, and math, participants who positively view their disability and demonstrate agency are actively engaged in self-advocacy [28]. Examples include students being willing to seek information or accommodation when problems are encountered. Conversely, individuals who view their LD in a negative or conflicted manner may struggle to practice advocacy and find it difficult to accept or take initiative in accessing accommodations [27]. In fact, the implication that one must adopt a "disabled identity" and embrace an ascribed label has resulted in some adolescents choosing to reject this identity to cope with stigma [27]. Due to these beliefs, individuals with LDs may be reluctant to identify with their disorder and have hesitations about self-advocating. Although different within the narrative for each individual, this suggests that the extent to which individuals view their LD as a part of their identity may have a degree of impact on their engagement in self-advocacy behaviors.

A primary way that self-advocacy groups target stigma is by changing negative attitudes, beginning with the self, and then of others. Kimball et al. find that emerging capacity for activism in high school students is associated with a developing sense of purpose [29]. These authors offer two definitions of self-advocacy: (1) the ability to communicate needs and wants, as well as locate services, and (2) being able to obtain necessary supports, have knowledge of self and one's rights, the ability to communicate, and ability to be a leader [29-32]. Kimball et al. propose that activism is rooted in larger concepts of identity, stigma, and purpose [29]. They found that students with disabilities who understood their needs and requested accommodation had stronger advocacy skills than those who did not. With encouragement to pursue this understanding and accommodation, students would be able to act as examples for others and create awareness so that the cycle of stigmatization is no longer perpetuated.

In a grander sense, enhancing self-advocacy skills not only creates a sense of solidarity in the community of individuals with LDs, but also allows individuals to be better integrated into the community and raise awareness. Anderson & Bigby found that self-advocacy alleviated stigma by allowing individuals with LDs to see themselves as capable and skilled, as well as affirmed and regarded as equals by peers [27]. Overall, while stigmas and stereotypes of LDs may initially cause individuals to refrain from advocacy, the process of navigating such misconceptions also has the potential to empower and create positive change for impacted individuals, especially with regards to self-advocacy skill development.

DISCUSSION

The intricate interactions between Ontario's existing support system, barriers, and societal stigmas are critical areas to investigate when examining skill development for adolescents with LDs. The benefits of existing programming aiming to foster self-advocacy through the LDAO and Ontario's education system are evident, encouraging an increased awareness for an adolescent's LD and ability to navigate the systemic processes. Despite these benefits, barriers continue to exist at the ground level, which could stem from the research gaps examined within this paper and previous research that was explored. By addressing these gaps and future areas for investigation, more insight into Ontario's climate on self-advocacy development in education can be strengthened, and students can be better supported as they navigate the provincial education system.

Current Research Gaps

When conducting a review examining the self-advocacy skill development of adolescents with LDs in Ontario, research was met with limitations as a result of the provincial scope. Currently, there is little evidence to support any conclusions relating to self-advocacy skill development among students with LDs, specifically across Ontario. On a broader and general level, the definition of self-advocacy may have posed a limitation in the review process. Self-advocacy is defined by a variety of terms throughout literature, including self-determination, goal setting, empowerment, self-assertion, and more.

We speculate some of these definitive inconsistencies to include different results being missed by authors due to various terminologies, slight deviations in definitions, or unverified/non-cited definitions, to name a few. Nonetheless, for the purposes of this review, the authors interpret "self-advocacy" based on a collective synthesis of the included literature. Additionally, the lack of longitudinal studies on this topic presents a methodological limitation, as it prevents researchers from capturing how adolescents' motivation evolves over time as they navigate Ontario's education system. Similarly, limited attention to comorbidities—despite the frequent co-occurrence of LDs with other conditions---hinders a comprehensive understanding of self-advocacy in these populations. 33 Although these limitations presented barriers within this review, they can be used to explore future developments within this area of research.

Potential Future Developments in the Field

A conceptual framework of self-advocacy provides avenues for development of these skills in adolescents, as well as improvements in student involvement throughout their educational experiences. Components of self-advocacy entail knowledge of oneself, such as strengths, weaknesses, interests, and attributes of one's specific LD, and knowledge of one's rights. While self-advocacy begins with understanding oneself and one's rights, the ability to apply this self-knowledge in group settings is essential for effective advocacy. Recognizing one's own needs and articulating them confidently enables individuals to navigate interactions with educators, support staff, and healthcare professionals, ensuring their voices are heard and their needs are met [34]. Moreover, a strong understanding of one's rights and personal needs fosters awareness of the roles and perspectives of team members. This awareness enhances collaboration, making advocacy efforts more informed and effective. Targeted programs or courses can support the development of these foundational skills, equipping individuals with the tools needed to engage in meaningful self-advocacy [37].

Parents and guardians play a crucial role in advocacy for adolescents with LDs, particularly in the education of younger students [34-36]. This includes attending IPRC meetings, providing relevant information, and representing their child's needs. A strong understanding of rights and responsibilities enables them to navigate the identification and accommodation process, including Ontario's formal appeal procedures. Factors such as socioeconomic status, educational attainment, language barriers, and knowledge of the education system, however, influence their ability to advocate effectively [37]. Various programs, including LDAO chapters across Ontario, offer parental advocacy support through communication training, system navigation assistance, and representation at school meetings. Expanding these services to include more personalized consultations and language-specific resources could enhance accessibility and impact [38,39].

CONCLUSION

Self-advocacy is a fundamental skill, providing the opportunity for students with LDs to feel confident, capable, and comfortable with the necessary tools to advocate for their needs throughout educational and employment pursuits. Gaps within Ontario's current programming still exist with respect to students' comfort, ability to speak up, and stigma, with significant implications for their experiences with their LD. Increasing adolescents' knowledge about their LD and understanding of the educational system are pivotal to self-advocacy skill development. Further research into Ontario-specific factors impacting self-advocacy is needed to build a system that adequately prepares students as they navigate the provincial education system and beyond.

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APPLYING THE BIOPSYCHOSOCIAL MODEL IN DGBI DIAGNOSIS

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ABSTRACT

Disorders of Gut-Brain Interaction (DGBI) are characterized by chronic gastrointestinal symptoms without structural abnormalities. There are several challenges in the diagnostic process due to difficulty identifying biomarkers for accurate diagnosis, the variability of symptom presentation, and the influence of culture on disorders. While the Rome IV criteria acknowledge cultural variations, existing cultural barriers complicate the diagnostic process. Discrepancies in diagnostic approaches across countries underscore the need to validate criteria like Rome IV in diverse cultural settings. Cultural differences also manifest in symptom expression and perceptions of pain, impacting diagnosis and treatment. Understanding these multifaceted influences is crucial for healthcare professionals to provide culturally sensitive care and improve diagnostic accuracy across diverse cultural contexts. The research suggests incorporating the biopsychosocial model can lead to more personalized and effective diagnoses. This paper advocates for incorporating the biopsychosocial model in diagnosis instead of solely using diagnostic criteria to offer a more comprehensive understanding of the illness.

INTRODUCTION

Formerly known as Functional Gastrointestinal Disorders (FGID), DGBI are a group of gastrointestinal (GI) disorders characterized by chronic GI symptoms with relation to intestinal motility disturbance, increased pain sensitivity in internal organs, altered mucosal and immune function, altered gut microbiota, and altered central nervous system processing [1]. The pathophysiology of DGBI is not yet fully understood, but it can be best identified as dysregulation of the gut-brain axis and presentation of any combination of motility disturbance, visceral hypersensitivity, altered mucosal and immune function, altered gut microbiota, or altered central nervous system processing [2,3].

DGBI heavily impacts the livelihood of children through the complex process of diagnosis and treatment. DGBI are associated with psychological disorders, emotional problems, school absenteeism, and poor school performance [4]. As such, timely and accurate diagnoses of DGBI can improve patient health.

Diagnosis of pediatric DGBI involves a medical history, physical examination, laboratory examination, and often a symptom-based assessment [5]. DGBI are diagnosed following the Rome Criteria, a diagnostic tool that classifies DGBI into 33 adult and 20 pediatric disorders based on symptom presentation [3]. These criteria assess the combination of physical symptoms present as well as their frequency and duration, and emphasize identifying symptom patterns without an identifiable organic cause [6]. Diagnostic criteria typically require that symptoms are present for at least three months, with onset occurring six months before diagnosis. Diagnostic tests such as endoscopy may be used to rule out structural causes. Additional tests such as balloon expulsion tests, anal surface EMG, or manometry may be employed according to the specific disorder presentation [5]. Some diagnostic consultations may consider psychological and social factors; however, these practices may be less formalized in the biomedical model, which assumes illness to be the consequence of exclusively measurable biological factors [7]. As the Rome Foundation acknowledges, minimizing this disorder to the symptoms presented in the Rome criteria can neglect the role that psychology and sociology can play. Applying the biopsychosocial model to children with DGBI provides a more comprehensive understanding of illness, enabling the examination of the combined roles of genetic, physiological, psychological and environmental factors that affect children with DGBI [8]. The biopsychosocial model has been widely used in healthcare since the 1970s to recognize the interplay of biology, psychology and sociology in medicine [9]. Since then, this model has provided a way to understand the discrepancies between biomedical thought and clinical observation [1]. Considering the biopsychosocial model in relation to DGBI allows for holistic assessment of the complexities in diagnosing DGBI as topics surrounding anxiety, social life, and culture can be considered in addition to factors such as genetics and cellular biology.

The absence of reliable biomarkers, variability of symptom presentation, and relevance of cultural context can lead to difficulties in diagnosing DGBI [5]. As such, looking at timely diagnosis based on individual circumstances guided by a patient, family and caregivers, and clinician beliefs may be beneficial.

BIOMARKERS AND DIAGNOSIS

A biomarker is a measurable characteristic that can be used to evaluate biological processes in an accurate and reproducible manner. Past efforts to identify biomarkers for DGBI diagnosis have been unsuccessful as they have not proven consistent enough to diagnose patients. Without a perfect reference standard to compare biomarker candidates to, many avenues for research end up with invalid results [10]. Early investigations have identified fecal volatile organic molecules and colonic mucosal immune cells as potential biomarkers. However, these findings are yet to be validated and tested in larger and more diverse sample sizes or in a diagnostic setting [11]. As such, there are currently no biomarkers that can be used to diagnose DGBI, posing a challenge to their timely and accurate diagnosis in people that are affected and seeking care [12,13].

A lack of reliable biomarkers increases the dependence on the symptom-based Rome IV criteria, the current gold standard for diagnosing DGBI. Diagnoses often rely on the process of elimination, which can be costly and tedious for patients and the healthcare system [7,14].
Additionally, the Rome IV criteria is limited in that it is based on its own previous versions, potentially introducing risk of bias. This has led researchers to look for biomarkers in many areas to try to deliver a more impartial and effective diagnosis to people afflicted by DGBI. However, a limited understanding of the pathophysiology of DGBI has slowed down investigations [15]. A potential option that takes this into account is the establishment of "surrogate endpoints," which are defined as biomarkers that may be substitutes for clinical endpoints to predict clinical benefit. Establishing surrogate endpoints may be able to guide researchers in biomarker investigations. An example of this would be establishing small intestinal microbiota as a surrogate endpoint and hydrogen breath tests as a biomarker candidate [11].

Another challenge in the search for DGBI biomarkers is the difference in presentation across several cultures and environments, making important prevalence and severity data difficult to gather. DGBI already presents differently from person to person, but the contrast may be more stark when compared across countries due to differences in symptom perception across cultures [10,16].

The Rome IV criteria is currently available in 33 different languages to allow for global consistency in diagnosis. However, there may be a need to implement further cultural and regional considerations for diagnosis. This is shown by instances such as inconsistent Irritable Bowel Syndrome (IBS), a common DGBI, epidemiology across different countries [17]. Establishing reliable biomarkers could help attain a clearer picture of global DGBI prevalence. However, it is largely unrealistic to try to target a single biomarker considering the variability of presentation [16]. Research into a broader scope of biomarkers is a current focus of DGBI research, as they are necessary for improved care and individualized patient treatment [11].

Considering these obstacles, DGBI research is moving towards innovative techniques that could provide a chance for a better diagnosis of DGBI. One of the potential avenues is the use of neuroimaging techniques. Current research has shown evidence of neural structural alterations in individuals with IBS. However, this finding alone is not robust to form new diagnostic criteria due to a 70% predictive accuracy. The primary hope for using neuroimaging in DGBI research and diagnosis is the reframing and recontextualization of the disorders falling into that category [18]. Another option, considering the high comorbidities experienced by people with DGBI, may be to identify psychomarkers to utilize in conjunction with future biomarkers and the symptom-based criteria already in use [12]. As such, searching for supplemental diagnostic criteria will likely have to consider the biopsychosocial model due to DGBI-specific diagnosis difficulties, such as a lack of biomarkers. The multifaceted nature of DGBI encourages innovative approaches in this sector.

VARIABILITY IN SYMPTOMS

DGBI, heterogeneous disorders, which are characterized by a wide range of symptoms, pose diagnostic challenges for clinicians showcasing the need for diagnosis through the biopsychosocial model [19].

Genetic predispositions demonstrate the biological aspect of symptom variability, as children with a family history of conditions like maternal anxiety or depression are at a heightened risk for developing DGBI [20,21]. Moreover, physiological factors, including the diversity of enteric neurons, have been associated with symptoms of DGBI, showcasing the role of biology in symptom expression [22]. Furthermore, research highlights the need to consider a child's unique genetic and physiological makeup, including the connections between genetic predispositions and environmental factors like diet, microbiota, and stress [14,21,23]. For example, the Children’s hospital of Philadelphia incorporated the biopsychosocial model through an integrated pediatric gastroenterology psychology program. Physicians are trained in providing DGBI diagnoses, cognitive behavioural therapy for abdominal pain disorders, treatment adherence, and feeding and eating difficulties. Additionally, there is training surrounding culturally competent care and considerations for people from marginalized groups. The program has experienced a larger increase in visit volume compared to the hospital's general GI division in the same decade. Outpatient visits have increased from 722 in 2011 to 6,348 in 2021 [24]. This approach showcases the need for an approach to diagnosis and treatment that accounts for the child's genetics and physiological state. Psychological aspects further complicate symptom variability. Mental health comorbidities like anxiety and depression, which are prevalent amongst children with DGBI, contribute to the variability of symptoms. These conditions worsen physical symptoms and complicate their management [25,26]. The high prevalence of anxiety and depression in children with DGBI highlights the importance of incorporating mental health support into the care plan for children with DGBI, addressing both the psychological factors directly influencing symptom severity and the co-morbid mental health conditions [25-27]. Other psychological factors, like catastrophizing, can exacerbate symptoms and reduce quality of life, demonstrating the need for psychological interventions as part of the diagnostic and treatment processes [26].

The social aspect of the biopsychosocial model also plays an essential role in the expression and severity of DGBI symptoms [25]. Family dynamics, peer interactions, cultural influences, and environmental stressors can significantly impact symptoms [25]. For instance, a family history of gastrointestinal symptoms can predispose children to DGBI, while negative peer interactions such as bullying can worsen symptoms through increased stress [25]. Furthermore, experiences of childhood abuse can lead to chronic stress, which in turn can aggravate symptoms, highlighting how social factors influence DGBI [21].

The complexity of DGBI in children highlights the need for a biopsychosocial approach to diagnosis and treatment [25]. Biologically, children's genetic and physiological makeup predisposes them to DGBI, psychological components can exacerbate symptoms, and social factors influence the severity of symptoms [23,25,26]. Given these factors, integrating the biopsychosocial model into healthcare settings can significantly advance understanding, management, and diagnosis of DGBI in children. By acknowledging and addressing the complex nature of these disorders, clinicians can diagnose more effectively [21]. This approach encourages a more nuanced view of symptom variability, moving beyond a solely biomedical perspective and including psychological and social considerations [22]. Clinicians can use assessments that account for the biological, psychological, and social aspects of a patient's health. This can involve collecting medical histories, and understanding the patient's mental health, lifestyle, and environmental factors. Clinicians can use this to create personalized treatment plans that can include therapies, social support, and medical interventions [28,29]. In summary, the biopsychosocial model creates a more comprehensive understanding of DGBI, increasing the potential for timely and accurate diagnosis and improving health outcomes [6-8,20].

CULTURE & ENVIRONMENT

Culture is characterized by values, beliefs, norms, and practices acquired by a group through learning and shared experiences. It serves as a guiding force influencing thought processes, decisions, and actions in predictable patterns [30]. Cultural influences can impact various aspects of the patient-physician dynamic, including the diagnostic process, treatment selection, and overall health outcomes [31]. It is notable how culture influences DGBIs through these aspects, a connection recognized in the collaborative development of Rome IV, which engages 117 experts from 23 countries and acknowledges the profound impact of cultural factors on disease manifestation and management [32]. However, despite this global effort, cultural barriers persist, complicating the diagnostic journey, particularly among pediatric populations.

From a biomedical perspective, there are some discrepancies in diagnostic approaches across different countries. As DGBI diagnoses depend on symptom description, it is necessary to establish the validity of the Rome IV criteria in different cultural settings. In particular, Hreinsson et al. conducted a study in 33 countries to assess whether or not the disorders in Rome IV aligned with their assigned symptoms in their particular populations. Findings indicate that North Americans are incompatible with the Rome IV symptoms for functional dyspepsia. This is potentially attributed to evolving clinical and diagnostic trends in the United States, where reflux disease is overemphasized, and dyspepsia's significance is underestimated [33]. Additionally, it was found that meal-related pain and abdominal pain limiting daily activities were additional symptoms correlated to IBS that are not currently included in the Rome IV criteria [32,34].Functional dyspepsia is another example of cultural differences that impact the biomedical perspective of DGBI. This condition is broken down into two subgroups: epigastric pain syndrome and postprandial distress syndrome [1,35]. Studies indicate that the prevalence of each sub-group varies between East Asian and Western populations and can potentially be attributed to factors such as Helicobacter pylori infection, which is more prevalent in Asia and may influence gastrointestinal motility and hormone production [36,37].

In childhood, cultural influences can shape one's psychological relationship with health. In particular, the stigma surrounding a functional disorder such as DGBI may manifest as shame or embarrassment to a child or their family, resulting in them minimizing their symptoms or downplaying its severity [38,39]. Moreover, cultural norms dictate the expression of pain, influencing a child's perception of symptoms [1]. For instance, in China, pain and bloating might be viewed as interconnected, with bloating seen as a less severe form of pain along a continuum [40]. Within a social context, familial experiences with chronic pain have been linked to more frequent episodes of pain in children. Children with mothers who have IBS exhibit increased occurrence of both GI and non-GI symptoms, along with more school absences and clinical visits [41]. While children with both parents experiencing chronic pain exhibited significantly worse health outcomes compared to those with only one or neither parent having chronic pain [42]. This may be explained through the social learning theory and the social communication model of pain, which suggests that children mirror their expression of pain based on how their parents demonstrate pain behaviour [43].

By simultaneously considering the biological, psychological, and social dimensions by applying the biopsychosocial model, healthcare professionals can attain a more comprehensive understanding of the factors influencing DGBI across diverse cultural contexts [43]. Particularly when utilizing internationally applicable criteria like the Rome IV, this approach ensures sensitivity to patients' cultural backgrounds in diagnosis. In a healthcare setting, assessing the emotional dynamics within a patient's family provides crucial insights into how relationships may affect the patient's health, including their contribution to health-promoting behaviors or disease progression through stress-related pathways or emotional dysregulation [44]. Enhancing clinicians' capacity to observe these relational dynamics and linking them to health outcomes via the biopsychosocial model facilitates broader systemic thinking and ultimately a clearer picture of DGBIs in children [44]. Consequently, future areas of study may benefit by looking into diagnostic patterns within immigrated/emigrated groups (e.g. a group in their native country compared to in a "melting pot" country like Canada or the United States) and associations between prevalence rates, perceptions of health, familial dynamics and culturally sensitive diagnostic processes.

CONCLUSION

It is essential to consider the biopsychosocial model when dealing with DGBI as the complex nature of this disease provides significant difficulty for children living with DGBI. Within an exclusively biomedical model, children living with DGBI could be classified or stereotyped as someone with a psychological disorder; this may invalidate the symptoms this child is feeling, and possibly even misdiagnose a child due to a misunderstanding of the effects of the DGBI.

The exploration of DGBI in children describes the complicated interplay of biological, psychological and social factors in their diagnosis and management. With an understanding of the limitations provided by a more traditional diagnosis, it is critical to advocate for the integration of the biopsychosocial model to develop a more holistic understanding of DGBI. By integrating genetic predispositions, physiological factors, psychological comorbidities and social influences into the diagnosis and treatment of DGBI, clinicians can develop better strategies that focus on the unique presentation of DGBI.

The limitations of diagnosis are apparent through the lack of diagnostic biomedical investigations, variability of symptoms, and cultural challenges which underscore the need for the biopsychosocial approach. Additionally, factors like genetic predispositions, physiological conditions, mental health comorbidities, family dynamics, and peer interactions, highlight the need for a nuanced approach to diagnosis [22].

With a variety of information to consider, clinicians may find it challenging to incorporate the biopsychosocial perspective, instead diagnosing children using the biomedical model. This may lead to increased risk of a misdiagnosis or stereotyping of physiological pain. The biomedical model is commonly used, but it is essential to consider all aspects of the biopsychosocial model when interacting with children to provide the patient with a diagnosis that accounts for their personal needs. By acknowledging the diverse nature of these disorders, clinicians can better address the complex needs of affected children, hopefully leading to improved health outcomes and overall better quality of life.

LIMITATIONS

It is important to note that the biopsychosocial model may present challenges in diagnosis or identifying symptom contributions in DGBI in children. For example, the subjective nature of social and psychological assessments may lead to variation in the interpretation of symptoms, which can complicate diagnosis [45,46]. Additionally, a multidisciplinary team comprising gastroenterologists, psychologists, social workers, and other professionals may be used to apply the biopsychosocial model [47]. This strategy may need resources that are not available and bring logistical difficulties. Furthermore, the evidence for the biopsychosocial model's use in children's DGBI diagnosis is still developing [22]. This gap in the literature suggests the need for more targeted research that can offer evidence-based recommendations for using the biopsychosocial model to diagnose and treat DGBI in children.

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EXPLORING THE NEUROLOGICAL IMPLICATIONS OF CLASSICAL MUSIC PEDAGOGY TRAINING FOR CHILDREN WITH AUTISM SPECTRUM DISORDER: THE ROLE OF MUSIC IN MEDICINE

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ABSTRACT

Music is the medicine of the mind [1]. Extensive research has explored the impacts of music training, through investigation across diverse musical domains and their neuro-focused aspects [2]. Knowledge of the direct implications of music education on cognitive development continues to evolve, with ongoing research demonstrating its positive effects on neuroplasticity. According to the Canadian Health Survey on Children and Youth, in 2019, 1 in 50 Canadian children aged 1-17 were diagnosed with Autism Spectrum Disorder (ASD). This paper delves into the neurological implications of Classical Music Pedagogy Training (CMPT) on children with ASD. Multiple studies reviewing the impacts of classical music, comparing the implications of musical training on typical development and ASD, highlight the benefits of CMPT for sensory processing, motor skills, and communication aptitudes in children with ASD. The paper further highlights the need for standardized terminology within this realm of research, and longitudinal studies examining the long-term impacts of CMPT on children with ASD.

INTRODUCTION

"Music Pedagogy" training is the study and practice of learning and teaching music [3]. More specifically, music pedagogy refers to the learnt ability, experiences, and knowledge that deepens understanding of an area of music [4]. In the context of this review, CMPT, a branch of music pedagogy, is explored to better understand the particular purpose and implications of CMPT as an intervention for child-related development. CMPT refers to the specific approach to music pedagogy and overall teaching. Particularly, classical music training is the approach to teachings, exemplifying learning music through the traditional skills and methods of that subject [5]. For the purpose of this paper, CMPT will be explored, through examining the neurological implications of such training as an intervention for children with Autism Spectrum Disorder (ASD).

ASD is defined as a neurodevelopmental disorder, frequently impacting a child's communication skills and social interactions [6]. ASD may present as differences in developmental domains, such as engaging in repetitive behaviours (e.g., stimming) or intense focus on specific interests, as well as challenges in social interactions [6]. The National professional development Center on Autism Spectrum Disorders (NPDC) conducted a comprehensive systematic review which was an extension of previous evidence-based practice review with the goal of outlining social communication interventions for children with ASD. Antecedent-based intervention, cognitive-behavioural intervention, naturalistic intervention, visual supports, video modelling, and more [7].

Nonetheless, the power of classical pedagogy training as an intervention, which applies several of NPDC's recommended social communication interventions, was not explored within that review [7]. This paper recognizes the importance of the work that exists pertaining to the role of music in medicine, and the potential neurological benefits of CMPT for children with ASD.

METHODOLOGY

A literature search was conducted utilizing terms broadly encompassing 'music education', 'autism spectrum disorder', 'communication', and 'neural processing.' A wide range of databases were consulted, including OVID Medline, Web of Science, and PubMed. Given the nature of the research landscape, several journals were used. In particular, the Journal of Cognitive Neuroscience, Journal of Neuroscience, and the Suzuki Associations of the Americas.

RESULTS

Social Communication and Autism Spectrum Disorder

When supporting an individual with ASD, effective communication strategies and interventions play a critical role, improving the child's overall quality of life. In terms of social communication related to ASD, research has emphasized the importance of early interventions.

A study revealed the importance of early intervention and detection of ASD. The study conducted a controlled trial examining 48 children diagnosed with ASD between 18-30 months old. The participants were further separated into two groups, one having the Early Start Denver Model (ESDM) intervention over two years, delivered by trained therapists and parents. The ESDM is an intervention that focuses particularly on early intervention for children with ASD, incorporating communication methods as a critical component of the intervention [8]. On the contrary, group two was referred to community providers for interventions that are commonly accessible within the community. Expanding on prior research, the study emphasized the efficacy of comprehensive developmental interventions for toddlers with ASD for its cognitive benefits [8].

Building upon existing knowledge, research continues to examine the longitudinal impacts of communication-based interventions targeted towards children with ASD. For instance, *Joint Attention, Symbolic Play, engagement, and Regulation (JASPER),* a naturalistic developmental behavioural intervention, which targets and analyses the impacts of joint engagement on a child's intimations of joint attention (IJA) skills, further examining whether here are associations between IJA skills and expressive language [9]. A few examples of IJA include pointing, showing, gaze following, coordinating attention, bringing objects to share, or commenting with gestures or vocalizations. Therefore, uncovering pathways that are important for social communication, and particularly expressive language display [9].

Rationale for CMPT as an Intervention in the Current Study

Studies show compelling evidence for the cognitive impacts of music pedagogy, through uncovering emotional and neurological benefits, however, there continues to be gaps between music teaching journals and neuroscience-based research. This paper aims to investigate the connections between literature within music education journals and the neuroscientific dimensions within the research landscape. Further seeking to bridge existing disparities within the context of ASD and music education. "Children with ASD are motivated to engage in music activities, and it can be a preferred medium to operate within. They thrive within structure and music fulfils this need for structure and routine [10]."

Of particular significance is a study published by the Journal of Cognitive Neuroscience, uncovering pitch sensitivity in individuals with autism. The paper expanded on past research which revealed the heightened strengths of participants with high-functioning autism, specifically, their ability to memorize picture-pitch associations, and recognize subtle pitch fluctuations. The paper revealed the ways in which individuals with ASD had increased pitch discrimination abilities. This investigation elucidated the findings of individuals with ASD outperforming typically developing populations in various low-level perceptual tasks [11].

The compelling evidence for the inherent cognitive and neurologic skills related to musical processing in individuals with ASD, coupled with early childhood classical music pedagogy, reveals the potential capabilities yet to be uncovered.

Neurological Power of CMPT for Typical Development

To truly cover the breadth of the neurological powers of classical music training for typical development, this section will discuss sensory processing and auditory-motor connectivity; communication, social skills, and emotional regulation; and fine motor skills, coordination, and executive functioning.

Sensory Processing and Auditory-Motor Connectivity

The Journal of Neuroscience (JNeurosci) examined the plasticity of the brain, as a result of 15 months of instrumental music pedagogy training in early childhood [12]. There were strong correlations surrounding structural brain changes in auditory and motor areas, with behavioural improvements on motor and auditory-musical tests. This paper was one of the first longitudinal studies to discover the brains structures at play in neuroplasticity, resulting in brain developments over time [12]. The study uncovered that a child who played and practiced a musical instrument, displayed stronger improvements related to motor skills. Which was measured through examining auditory-motor connectivity through finger dexterity and in auditory melodic and rhythmic discrimination skills [12].

The correlation between auditory-motor connections within the brain is critical to explore. The brain-behaviour correlations exist in motor and auditory brain regions for performance on motor and auditory tests [12].

When playing a musical instrument, through music pedagogy teachings, the primary motor area plays a critical role the execution and control of bimanual sequential finger movements and motor learning in general [12].

In terms of sensory processing, CMPT has incredibly structured and organized auditory patterns. Studies that compare non-musicians and musicians identify four areas in which music-training neuroplasticity determines: number of years of continuous training, age of training onset, amount of practice, and aptitude. Neuroscience research published by *Perspectives* Journal expands on this evidence of neuroscience research showing the possibilities of music training leading to changes in the auditory system [13]. For example, the neural representation of timing and harmonic features of speech signals the presence of background noise is stronger in musicians than non-musicians. Therefore, the evidence suggests that musicians possess enhanced cognitive and sensory abilities that improve their ability to process speech in challenging listening environments, when comparing their listening abilities to non-musicians [13].

Communication, Social Skills, and Emotional Regulation

Forgeard et al. examines the implications of practicing a musical instrument in childhood found that the children outperformed the control group in verbal abilities, specifically vocabulary, and non-verbal reasoning [14]. The results were further strengthened as the study found that the duration of music training further predicted the outcome of the child's performance on the assessment measures; Raven's Advanced Progressive matrices and Vocabulary test [14].

Although not exclusive to classical music, Moreno et al. explored the impacts of music training on verbal intelligence, further strengthening communication abilities[15]. Moreover, the nature of music pedagogy training can have positive impacts on the child's overall emotion regulation. For example, music pedagogy training often incorporates structured activities such as rhythm exercises or expressive musical play, which can help children develop emotional self-regulation by providing a safe space to express emotions, learn impulse control, and practice various coping strategies [15]. Through examining the psychological inquiry of musical behaviour the study revealed music's powerful ability to foster emotional self-regulation through engaging children in structured yet expressive activities [16,17] .

Fine Motor Skills, Coordination, and Executive Functioning

CMPT pertaining to instrument learning, has the profound ability of strengthening fine motor skills. A study by Costa-Giomi, who conducted a longitudinal investigation, discovered that children who received two years of piano pedagogy training, improved greatly in terms of their motor proficiency test [14].

The results of the study from Journal PLOS, revealed that children who received instrumental music pedagogy training for three or more years outperformed non-musically trained participants (control) [14]. Specifically, they outperformed their counterparts in facets related to fine motor skills, in both hands, as well as decreased ability to discriminate between melodies. The assessment was conducted through the Gordon's IMMA and the Melodic Discrimination Task [14].

Neurological Power of CMPT for children with ASD

There has been growing interest surrounding the exploration of neurological implications of CMPT as an intervention for children with ASD. This section aims to synthesize existing literature that explores the neurological power of classical music training, specifically for children with ASD. As mentioned, classical music training due to its structure, pattern and composition, has profound neurological purposes for typical development. By examining the neuroscientific evidence behind classical music pedagogy, this section seeks to understand the potential neurological implications that may arise when children with ASD undergo CMPT.

Sensory Processing and Auditory-Motor Connectivity

Through examining the research in the JSTOR digital library, various studies were uncovered, which discussed the improved sensory processing and auditory-motor connectivity for children with ASD, who are musically trained. Specifically, a study by Thaut examined the structured auditory patterns in classical music. Further identifying that the specific arrangement of harmonies, and rhythms in classical compositions provide a consistent pattern. That pattern is also predictable, in many ways. The structure is incredibly beneficial for children with ASD, as they can experience challenges in processing complex auditory stimuli [18]. As well, the paper delves into the benefits of the structured auditory pattern in supporting an individual with ASD in identifying and interpreting sensory information with a higher degree of effectiveness [18].

The passive listening techniques such as Auditory Integration Therapy (AIT) have been highly used to address behavioural difficulties and auditory hypersensitivity in children with ASD [18]. Moreover, children with ASD may have difficulties modulating sensory inputs which can as a result manifest as enhanced perception of auditory and visual stimuli. Nonetheless, Sensory Integration therapy through music has abilities to strengthen auditory-motor abilities [19].

Communication, Social Skills, and Emotional Regulation

Building communication abilities is a crucial aspect of interventions for children with ASD. The study by Hayoung A Lim., separated children with ASD into three distinct groups. The various groups underwent music training, speech training and non-training. The results revealed that participants in music and speech training experienced a significant increase in their verbal production, when comparing the pre to posttest results [20]. Further identifying that children with ASD understand important linguistic information that is incorporated in music stimuli organized in patterns of perception, and further result in functional speech production [20].

In terms of communication, research has uncovered the abilities of music-based interventions to address core ASD related impairments in joint attention, social reciprocity and nonverbal and verbal communication. Furthermore, children with ASD can experience difficulties related to direct social engagement, therefore music related interventions provide a socially embedded group music activity, providing opportunities to make room for comfortable social interactions [19].

Fine Motor Skills, Coordination, and Executive Functioning

In terms of fine motor skills, it is suggested that children with ASD experience impairments in gross motor skills, including bilateral motor coordination, balance, and gait, as well as fine motor delays [19,21]. Musical activities embedded within pedagogy are typically used to address such difficulties, and have been found to build significant progress in the child.

Furthermore, 4-6 years old who experienced 2-month music and movement programs displayed improvements in their gross motor skills [22]. Specifically, displayed through jumping and dynamic balance, measured through Motor proficiency test comparing children who engaged in non-musical programs [22]. Another study revealed that children between the ages of 4-6 years old who received a 10-week Dalcroze-based musical integration program, coupled with a physical education program, outperformed children who solely received a general movement exploration program [23].

DISCUSSION

Decades of research on the neurological impacts of music have demonstrated its profound influence on cognitive, emotional, physical, and social development [24]. CMPT experiences have been shown to enhance neuroplasticity, facilitate communication, and support sensory process, which are key areas of development for children with ASD. However, despite these promising findings, significant gaps remain in the literature regarding the standardization of methodologies used to assess CMPT interventions for children with ASD. This has contributed to a fragmented research landscape, where inconsistencies in terminology, methodologies, and evaluation frameworks hinder progress.

One of the primary challenges in this field is the lack of a universally accepted framework for assessing the effects of CMPT on specific developmental outcomes within ASD. Neuroscientific and music education research often employs different vocabularies and methodologies, making cross-disciplinary collaboration difficult. Meta-analyses rely heavily on standardized terminology to ensure consistency in interpreting findings, yet there remains no consensus on how to define and measure the effectiveness of CMPT. Establishing a shared language is essential for advancing research, facilitating interdisciplinary collaboration, and ensuring that findings can be accurately compared and applied in real-world settings.

The heterogeneity of ASD further underscores the need for personalized approaches to CMPT. ASD manifests in highly diverse ways, meaning that a pedagogical technique effective for one child may not yield the same results for another. This variability introduces biases in participant pools and complicates the generalizations of findings. Additionally, barriers such as financial constraints, limited access to music education, and varying levels of parental or guardian support contribute to disparities in CMPT accessibility [25,26]. Future research should address these barriers to ensure that all children with ASD have the opportunity to benefit from CMPT, if they wish.

Barrett et al. (2022) conducted an exploratory study examining early childhood educators' beliefs about music education, emphasizing its perceived value as a creative and inclusive tool for fostering social and emotional development [27]. While music is widely acknowledged for its positive impact, the specific mechanisms by which CMPT influences neurological and behavioral outcomes in children with ASD remain insufficiently explored. Existing research lacks a comprehensive analysis of the key characteristics of classical music pedagogy—such as melody, tempo, dynamics, and intonation—that contribute most significantly to positive developmental outcomes. Identifying these critical factors is essential for refining instructional strategies and optimizing the effectiveness of CMPT interventions for children with ASD.

Furthermore, much of the existing literature focuses on short-term benefits, with limited research examining the long-term effects of CMPT. Longitudinal studies are necessary to determine whether early CMPT engagement has lasting neurological and behavioral impacts in children with ASD. By addressing these gaps, through standardized methodologies, consensus on terminology, and long-term research, a more comprehensive understanding of CMPT's role in ASD interventions can be established. Ultimately, this will allow for more effective implementation of music-based therapies, leveraging the power of classical music as a meaningful and accessible tool for cognitive and emotional development in children with ASD.

CONCLUSION

In the broader context, this research underscores the neurological benefits of music education, and the specific implications CMPT can have on child development domains. The role of music in medicine is complex and at times elusive, however, it is evident that there is a purpose for music within medicine. The intricate ways that music interacts with neural networks, further cognitive development and carry an overall positive impact on the well-being of children with ASD. While the study outlines various gaps, limitations and controversies within the literature, there is undeniable potential for future developments within this area of research. There must be a commitment towards advancing knowledge in the nuances of music’s power on the neurodevelopment of children with ASD, to build a future where music and medicine can work hand-in-hand.

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INVESTIGATING THE RELATIONSHIP BETWEEN ADVERSE CHILDHOOD EVENTS AND CHRONIC PAIN IN PEDIATRIC AND ADULT POPULATIONS

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ABSTRACT

Chronic pain is a highly prevalent condition in pediatric and adult populations, with epidemiological rates increasing over time. Literature has highlighted a correlation between adverse childhood experiences (ACEs) and chronic pain conditions, with a great focus on adult populations. This paper explored this existing research, while diving deeper into studies pertaining to children and adolescents with chronic pain. This review also examined proposed models that can explain this correlation to explore conceptual understandings, alongside the underlying biological mechanisms that facilitate this relationship to investigate physical processes. Adverse childhood events, and trauma is correlated with chronic pain and other pain conditions in both child, adolescent and adult populations, with differences in prevalence and intensity present depending on the adverse experience, and the number of experiences. A need for further research in pediatric populations is highlighted.

OBJECTIVE

Chronic pain is a severe public health concern with high prevalence rates in child, youth and adult populations [1-3]. This paper seeks to provide a comprehensive overview on: 1) How does the presence of childhood experiences and psychological trauma correlate with the development of childhood and youth chronic pain and other pain disorders; 2) How do these experiences reflect in adult chronic pain models; and 3) What are the underlying mechanisms and proposed models that facilitate this relationship? The purpose of this literature review is to investigate the existing literature on children and adolescents with regards to chronic pain and its connection to psychological distress and examine the ways in which adulthood chronic pain is connected to these adverse childhood experiences. Given the plasticity of the brain in youth and their higher activity levels than adults, the underlying processes in the emergence of pain may differ from the observations in adults, thus a comprehensive investigation into both populations would be beneficial. Further research in this field may elucidate treatment targets that could enhance existing psychological interventions earlier than adulthood for those who may experience or be predisposed to experiencing chronic pain.

BACKGROUND

Chronic pain is longstanding pain that persists typically defined as lasting longer than 3 months [1] involving an interaction of physiological and psychological variables, affecting daily functioning [4]. Chronic pain is a major public health concern with prevalence rates between 11-38% in children, with similar estimates found in adult populations [5] There is increasing evidence to support a significant association between early-life adversity and the incidence of chronic pain both in childhood, adolescence and later in life [5]. Despite high prevalence rates of chronic pain conditions in child and adolescent populations, the relationship between adverse childhood experiences (ACE) and chronic pain conditions is less researched as majority of research has investigated this matter in the context of adult populations [6].

A review of epidemiological rates found that chronic and recurrent pain is prevalent in child and adolescent populations, with girls generally experiencing more pain than boys, and that prevalence rates increase with age [1]. Further research suggests that the prevalence of childhood pain increased over the last few decades [5], and supports the idea that clinicians and researchers should be aware of this problem and the long-term consequences. Chronic pain is also highly prevalent in adult populations [6] with approximately 20% of adults worldwide experiencing chronic pain [7], making it a significant public health concern.

Considering the connection between ACE and pain, traumatic experiences are quite common in childhood [8]. The results of a large epidemiologic study found that approximately 30% of youth experienced one or more traumatic events by age 16, with 13% of those youth endorsing symptoms of post-traumatic stress [8]. In pain literature, the relationship between Adverse Childhood Events (ACEs) and increased risk for pain outcomes have been investigated, however, this research is largely conducted in adult populations and in the context of retrospective accounts. In addition, it is often limited to abuse and trauma alone [6]. The lack of empirical research in this subject matter is concerning, as chronic pain conditions are highly prevalent in youth and can be disabling [1, 2].

RESULTS

The Correlation Between Childhood Adverse Events and Pediatric Pain

The relationship between psychological trauma and pain in adolescence has not been investigated thoroughly, however, of the research that has been conducted, pediatric populations with chronic pain conditions such as juvenile onset fibromyalgia and migraine have a higher likelihood of reporting an ACE [9]. The 1998 Adverse Child Event study investigating the relationship between childhood abuse and household dysfunction to death, demonstrated that the association between ACEs and chronic conditions was frequency dependent [10]. This finding is further substantiated by evidence from a 2020 study that described a dose-dependent relationship between ACEs and chronic pain during childhood and adolescence [11]. In contrast, some research has found that average pain intensity does not significantly differ across the number of ACEs reported [12]. Nonetheless, after controlling for demographic and clinical factors in the United States, a study found that exposure to one or more ACE was associated with a 60-170% increased likelihood of experiencing chronic pain [11]. Furthermore, in a retrospective study, the highest proportion of participants who reported greater than or equal to 3 ACEs had a primary pain diagnosis of widespread musculoskeletal pain [12]. Interestingly, among youth with traumatic brain injuries, PTSD symptoms predict pain symptoms [13] further demonstrating that the effects of trauma can increase pain. Interestingly, the relationship is not the same for all kinds of pain. For instance, research has shown that adolescents with widespread pain are more likely to report frequent abuse or trauma than those with chronic migraines [11].

Empirical evidence also suggested a strong correlation between exposure to potentially traumatic interpersonal events (PTIE) and consequent psychological distress with recurrent headaches in adolescents [14]. Increased exposure to PTIEs was associated with a higher prevalence of recurrent headache disorders, possibly indicating a dose-response relationship [14]. Interestingly, in this study, generally, twice as many girls as boys reported recurrent headache, and prevalence increased with age in girls but not boys [14], indicating some sex differences in this relationship.

Investigating younger populations, a cross sectional study in Portugal found a dose response association between the number of ACE's and reported pain outcomes in children aged 10, suggesting that the pain effects of ACE's start earlier in childhood than previously reported and investigated [15]. There is some preliminary and limited research in pediatric complex regional pain syndrome (CRPS). For instance, an article by Wager et al highlights that there is limited understanding of the relationship between psychological factors and complex regional pain syndrome (CRPS) in children, particularly regarding which factors trigger CRPS and which arise as a consequence of the chronic pain [16] However, they have found that these children experienced a larger number of stressful life events than controls [16]. This research highlights that prospective long-term studies are needed to further explore this correlation. Interestingly, a recent prospective study found that boys with recurrent abdominal or pelvic pain at age 7 were more likely to report headaches, abdominal/pelvic and musculoskeletal pain at age 13 [17], demonstrating that early childhood pain associated with psychological distress can predict pain in adolescence as well. A systematic review supports this finding, discussing that some types of pain may become chronic in childhood and be predictive of long-term pain related disabilities [1].

A comprehensive meta-analysis by Davis and colleagues revealed that adults who have endured childhood abuse and neglect exhibit heightened pain symptoms compared to those who were not exposed to trauma [18]. This finding is aligned with the growing body of evidence that illuminates an important correlation between early life adversity and a greater incidence of chronic pain in adulthood [19]. Moreover, research highlights that individuals who have reported stressful events in childhood were more likely to develop back pain and chronic low back pain (CLBP) compared to individuals who did not undergo such adversities [20, 21]. Similarly, Creed et al. observed this association with adverse events like interpersonal difficulties and abdominal pain [22]. Notably, while severe childhood abuse did not directly impact chronic pain, physical abuse emerged as a significant factor [21]. Furthermore, women with Chronic Pelvic Pain (CPP) showed a significantly higher incidence of severe childhood sexual abuse than patients with headaches or no pain [21, 23]. These findings collectively suggest a distinct correlation between sexual abuse and CPP and a general association with physical abuse and chronic pain.

Pain is frequently reported among individuals with PTSD [24]. Researchers have proposed that traumatic events and their subsequent PTSD symptoms can increase the risk for development or worsening of chronic pain, and that these factors are mutually maintained [25]. Individuals who reported trauma exposure were found to be 2.7 times more likely to manifest a functional somatic syndrome [26]. A systematic review found that people with psychological trauma face a 3.3 times heightened risk of developing generalized pain disorder, and temporomandibular pain, 2.2 times more susceptible to OBS, and 2.5 times more likely to develop fibromyalgia [27]. Notably, the age of trauma has no influence on these results, rather, the most robust association was observed with high levels of emotional stress, surpassing that of physical abuse [26]. In a separate investigation with 62 females from a community-based sample, emotionally abused individuals had reduced heat pain tolerance [19]. Furthermore, Scott et al. identified a correlation between neglect, family violence, abuse, or family criminal behavior and symptomatic pain conditions [28]. Collectively these findings substantiate that there is a complex interplay between trauma and chronic pain.

The first MRI study on patients with Irritable Bowel Syndrome (IBS), a chronic condition in which individuals report visceral pain [29], found an increased perception of pain in patients with a history of abuse had a stronger activation of the left, middle, and posterior cingulate cortex compared to controls. These brain regions are related to attention regulation, which has been interpreted by authors as an indication of pain amplification [30]. Furthermore, the authors found less activity in the left supragenual anterior cingulate cortex which is involved in pain inhibition and emotional arousal. It was similarly found that sexually abused individuals showed changes in neuronal processing with higher activation in the lateral and medial superior frontal gyrus and lower hippocampus activations than controls [31]. Moreover, patients with a history of physical and sexual trauma exhibited a greater temporal summation which indicates increased pain to repetitive painful stimuli, a larger number of pain sites, and more pain related disabilities compared to patients without trauma [32]. This suggests altered neuronal processing of perceived pain in patients with a trauma background.

Proposed Mechanisms and Models Facilitating the Relationship Between Childhood Adversity and Pain

The Stress Allostatic Load Model of Chronic Pain:
Evidence supports the role of the Hypothalamic-pituitary-adrenal (HPA) axis, the immune system, the opioid and endocannabinoid system as well as epigenetic mechanisms on the connection between early life adversity and chronic pain.

The stress allostatic load model of chronic pain posits that persistent pain can develop from a sustained endocrine response, such as consistently high levels of cortisol in the brain, predisposing individuals to sensitization - remaining in a state of arousal, alongside a reduced hippocampal volume. With repeated trauma and exposure to ACEs, the sympathetic and parasympathetic responses become dysregulated and allostatic load in addition to the dysregulation of the HPA axis may occur [6]. Interestingly, while acute stress can elicit analgesia in humans, early life and chronic stress acts more pronociceptive, increasing cold allodynia and heat hyperalgesia and increasing sensitivity to noxious inflammatory stimuli [33, 34]. Thus, the modulations of the HPA axis associated with early life stress can dysregulate corticosterone signaling and contribute to heightened pain responses. According to a study on cortisol secretions in fibromyalgia (FM) and rheumatoid arthritis (RA) patients compared to healthy controls, saliva samples for cortisol analysis, over two days showed that HPA axis dysfunction both with increased and decreased cortisol levels has been reported in these chronic pain patients [35]. FM and RA patients had higher average cortisol levels than healthy controls, but there were no differences in cortisol diurnal patterns or reactivity to psychological stress. Despite reporting lower stress levels, the patient groups differed from controls on stress measures. Adjusting for psychosocial and lifestyle factors did not alter these cortisol findings. These findings provide additional evidence of hypothalamic–pituitary–adrenal axis disturbance [35].

The Cognitive Appraisal Model of Chronic Pain:
Furthermore, a prolonged or exaggerated stress response associated with psychological trauma can perpetuate cortisol dysfunction with exaggerated physiological responses. For example, catastrophizing can lead to altered cognitive appraisals of stimuli as being threatening, creating sensations of pain [36]. Along a similar model, Nelson et al. has proposed that youth with a history of chronic stress or ACEs engage in cognitive distortions that surround perceived stress like pain, which influence coping styles, length of pain, and impairment overall [36]. Thus, the neuronal remodeling that follows chronic stressors can exacerbate pain perception due to the loss of anti-nociceptive signaling in the central pain matrix.

Pro-inflammatory Signaling in Trauma and Pain Perception:
Pro-inflammatory signaling is directly associated with chronic pain [36]. As childhood is a sensitive period, when combined with acute and even toxic levels of stress, evidence suggests that it is predictive of increased inflammation in preadolescent and adolescent years [37], demonstrating a physiological response to psychological distress. Slopen et al. particularly found that adverse events in middle childhood (ages 6-8) and

cumulative adversity from birth to the age of 8 was associated with elevated levels of CRP and IL-6 by age 10 and 15, respectively [37]. Interestingly, adverse events experienced earlier in childhood (aged 1.5 to 6 years) were not associated with the same level of inflammation. The brain increases production of cytokines such as tumor necrosis factor-alpha (TNF-a), interleukin-6 (Il-6) and C-reactive protein (CRP) in response to acute and prolonged stress in child populations [38, 39]. In a study of 92 individuals, levels of CRP were directly associated with somatic pain complaints [40]. Family violence, assessed with regards to inter-parental conflict and aggression, has also been correlated with biomarkers of increased cortisol signaling in children and adolescents. In addition, caregiver depression is a predictor of inflammation in early adolescence.

Furthermore, epigenetic and inflammatory models describe how stress has been linked to alterations in DNA which feeds into pro-inflammatory signaling. Particularly, DNA methylation and histone acetylation patterns in the brain increases the expression of pro-nociceptive neurotransmitters [41]. This DNA methylation can diminish glucocorticoid receptor sensitivity leading to resistance [42]. As glucocorticoids are anti-inflammatory agents, desensitization to this system increases inflammatory signaling and consequently pain [43]. Interestingly, another epigenetic mechanism has been posited to be involved in the pain trauma pathway. Childhood trauma has been found to induce demethylation of the gene FKBP5 which alters the expression of the FKBP51 protein; a critical mediator of chronic pain [44]. From these mechanisms, it is apparent that inflammatory signaling acts as a mediator of the relationship between psychological trauma and chronic pain, modulated by epigenetic changes.

The Role of Endogenous Opioid and Cannabinoid Systems in Pain Signaling:

The endogenous opioid and endocannabinoid systems play a role in the underlying enhanced nociceptive behavior, possibly explaining pain hypersensitivity in individuals exposed to childhood adversity. Rat studies have shown that Maternal deprivation (MD) reduced overall brain opioid receptor binding with altered expression of endogenous opioids in key brain areas involved in the modulation of nociceptive processes [45]. Early life stress has been shown to alter the endocannabinoid system in both the short and long term. For instance, MD results in decreases in cannabinoid receptor type 1 (CB1) receptor expression with increases in CB2 expression [46]. Evidence shows that endocannabinoids regulate pain and are modulated by stress. Further, they are antinociceptive in inflammatory and neuropathic pain models [47], therefore, deficits have been proposed to underlie hyperalgesia. Sensitivity to pain is often present in chronic pain patients.

DISCUSSION

This review highlights the intricate relationship between adverse childhood experiences (ACEs) and chronic pain, with emphasis on pediatric and adult populations. While a correlation between ACEs and chronic pain is well-documented, nuances in this relationship warrant deeper exploration. This section will expand on key findings, examine mechanistic insights, and propose directions for future research.

Pediatric populations with chronic pain conditions have a higher likelihood of reporting an ACE in a dose dependent fashion [9-12]. More robust associations have been observed between high levels of emotional stress, surpassing that of physical abuse [19]. Interestingly, the relationship is not the same for all kinds of pain and trauma. For instance, sexual abuse in childhood can be considered a risk factor for the development of CPP [21,23] while physical abuse is more associated with CLBP [20]. Emotional stress, surpassing physical abuse, emerges as a robust predictor of heightened pain responses and pain-related disabilities. Additionally, findings suggest that the type of pain experienced, such as musculoskeletal pain versus migraines, may be influenced by the nature of the trauma. Furthermore, a recent cohort study found that the pain effects of ACE's start earlier in childhood than previously reported and investigated [15]. This finding indicates a need for further research and specificity.

Interestingly, studies have noted that traumatic events and their subsequent PTSD symptoms can increase the risk for development or worsening of chronic pain and that these factors are mutually maintained [24,25]. These findings highlight the reveals the bidirectional relationship between psychological distress and pain. Adults who have endured childhood abuse and neglect exhibit heightened pain symptoms than those who were not exposed to trauma [18,19]. Specific studies have noted this association with chronic lower back, pelvic, and abdominal pain as well as pain disorders such as fibromyalgia, IBS, and other generalized pain disorders in addition to altered pain perception with temporal summation [20-23,30]. Neuroimaging studies have shown alterations in brain regions such as the cingulate cortex and frontal gyrus as well as hippocampus, all associated with pain perception, and emotional arousal [29,31,32], highlighting structural changes in the brain as a mediator of chronic pain in patients with a trauma history.

Emerging evidence indicates that the timing of ACEs plays a critical role in determining their impact on chronic pain. For example, pain effects of ACEs appear to manifest earlier in childhood than previously recognized. Studies have demonstrated that pain symptoms, such as recurrent abdominal or pelvic pain at age seven, can predict the presence of chronic pain in adolescence. This emphasizes the necessity of early intervention to disrupt the trajectory from childhood trauma to long-term pain disorders. The findings demonstrate that early childhood pain associated with psychological distress can predict pain in adolescence as well.

Mechanistic studies provide valuable insights into how ACEs contribute to chronic pain. Dysregulation of the hypothalamic-pituitary-adrenal (HPA) axis, pro-inflammatory signaling, and epigenetic modifications are key pathways implicated in this relationship. Researchers have posited that adverse childhood experiences have a physiological impact by altering cortisol signaling with marked changes in the HPA axis [6], and DNA, altering inflammatory pathways posited to cause peripheral sensitization [33,34] which can lead to hyperalgesia and chronic pain alongside other pain disorders [36]. Other models have proposed changes in cognitive appraisals of stimuli, creating sensations of pain; a phenomenon known as catastrophizing as a response to exaggerated stress responses associated with psychological trauma [6,36]. Endogenous signaling molecules are also implicated in this process, with evidence indicating that endocannabinoids and opioids are sensitive and responsive to early childhood stressful experiences.

In summary, this review underscores the multifaceted nature of the relationship between ACEs and chronic pain, emphasizing the need for a nuanced approach that considers trauma types, developmental timing, sex differences, and mechanistic pathways. These insights provide a foundation for advancing research and clinical care in this critical area.

FUTURE DIRECTIONS

Several limitations in existing research must be addressed to advance the field. Current studies focus mainly on physical and sexual abuse, neglecting other ACEs like household dysfunction or parental mental illness. Most research relies on cross-sectional designs, limiting causal inference. Prospective longitudinal studies are needed to explore the temporal relationship between ACEs and chronic pain, particularly in pediatric populations, where brain-based mechanisms of stress and pain remain poorly understood [6].

Future research should examine cumulative and dose-dependent effects of multiple ACEs on diverse pain conditions, while considering environmental and behavioral factors such as socioeconomic disparities and healthcare access. Addressing these gaps will improve prevention and intervention strategies for trauma-exposed populations.

Finally, existing models linking stress and pain in adults and youth often overlook the complex interplay of physiological, psychological, behavioral, and environmental factors.

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UNVEILING THE IMPACT: THE ROLE OF CHILD-LIFE SPECIALISTS IN THE CANADIAN HEALTHCARE SYSTEM

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ABSTRACT

Certified Child-Life Specialists (CCLS) play a vital role in supporting infants, children, youth, and families navigating the complexities of illness, trauma, disability, loss, and bereavement. This review delineates the six core domains of care within which CCLS operate, and underscores the novel findings regarding the efficacy of CCLS interventions. Through a synthesis of existing literature, the tangible benefits of integrating CCLS within healthcare teams are demonstrated, transcending traditional hospital settings to positively impact paediatric care outcomes across diverse healthcare contexts. Moreover, the current study contributes to the flourishing discourse surrounding paediatric care in Canada by addressing pertinent issues such as workforce diversity and burnout among healthcare professionals. By advocating for the enhanced integration of CCLS within the Canadian healthcare system, this focused review underscores the urgency of prioritizing psychosocial care to meet the evolving needs of paediatric patients and their families. By supporting the well-being of CCLS practitioners and promoting their seamless integration within healthcare systems, institutions can enhance patient care quality and advance the goals of patient-centered care.

BACKGROUND

Certified Child-Life Specialists (CCLS) are healthcare professionals who guide infants, children, youth and families in uncertain times surrounding illness, trauma, disability, loss and bereavement [1]. Traditionally, CCLS have primarily provided support in hospitals, as hospitalization is a major stressor in a child's life [2]. CCLS are not only employed in children's hospitals, but general hospitals as well to provide psychosocial support for children whose parents or loved ones are hospitalized [3,4]. In Canada, CCLS dispense child life services in a variety of settings beyond hospitals. This can include, but is not limited to, community programming, advocacy centers, non-profit organizations, and academia [5].

CCLS provide their support through a myriad of ways to engage children and families, making their scope of practice broad and unique. Within this unique scope, they function within 6 domains of care;

1. Resilience focused,
2. Play-based,
3. Individualized approach,
4. Developmentally grounded,
5. Relationship-orientated,
6. Trauma-informed.

CCLS will have completed a minimum of a bachelor's degree in any field of study, ten college-level courses in child life or a related department/subject including a minimum of one child life course taught by a CCLS, and a minimum of 600 hours of child life clinical experience under the direct supervision of a CCLS [6]. Once certified, CCLS look to identify and help children navigate medical environments (i.e., length of stay, medical procedures), family factors (i.e., parental anxiety, parental involvement in care, overall family support), and even individual factors (i.e., age/developmental level, temperament, trait and state anxiety, and coping style). CCLS routinely use play which can range from a child's definition of a "fun" activity or healthcare play where CCLS will use materials related to health care to clarify misunderstandings or work through painful experiences. They can also use an array of other interventions like family facilitation, therapeutic dialogue, developmental support, and procedural support [7]. This focused review endeavors to elucidate the indispensable role of CCLS and their interventions, advocating for their enhanced integration within the healthcare system.

Therefore, the research question of this review is: what are the short-term and long-term impacts and implications of FASD on a child's cognitive development? This will allow us to identify what is needed to support the challenges that stem from this domain.

Our review followed a non-systematic approach and both a web search and electronic database search were performed to find published literature that shed light on this topic. The results from the literature are presented below.

RATIONALE

To understand the need for expanded CCLS integration, it is important to consider how pediatric hospital care has evolved over time. The hospital setting for children has evolved over the years. A typical children's hospital in the early parental contact, cognitive stimulation and/or learning experiences, and a sense of comfort for children [8]. These are all aspects that hospitals in the 2000s aim to value. This shift could reflect changes not only in hospital infrastructure [9], but also in the mindset of healthcare providers. With the advent of the patient-centred care model, the introduction of CCLS and the maximization of their efforts is necessary, as CCLS are a manifestation of the switch from a biomedical care model to a patient-centred one.

Currently, Canada has one of the lowest physician-to-population ratios globally [10]. Given the prevalent phenomenon of burnout among paediatric nurses, the allocation of sufficient time for interactive play with paediatric patients, a crucial component of paediatric care, presents a considerable challenge [11,12]. As such, the integration of CCLS is integral to the paediatric care model.

Child-Life care in Canada was introduced more than 40 years later than in America as the "Child in Hospital" resolution advocated for by the Canadian Paediatric Society. In 1978, 93% of 180 general hospitals had play spaces and 37% of those spaces had salaried staff. 77% of Canadian child-life programs have one to three CCLS on staff. Figure 1 represents the number of CCLS across Canada as of 2020. With Ontario in the lead, 30 CCLS belong to McMaster's Children's Hospital [5]. This shows an uneven split among of CCLS among community hospitals and in rural communities. This article aims to show the urgency for the Canadian healthcare system to promote a greater reliance on CCLS in a balanced way that prevents burnout and compassion fatigue for all healthcare providers involved.

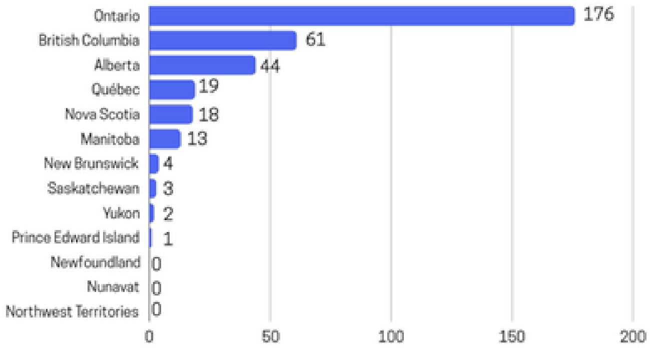

__Figure 1__. A row bar graph representing the number of child-life specialists in each Canadian Province/Territory as adapted from the Association of Child Life Professionals (2020) [5].

KEY FINDINGS

This literature review provided insights into the benefits of having an empowered CCLS on a patient's care team. As their role has been seen to spread to a variety of settings, so can their ameliorative qualities be seen in different settings. In a traditional hospital setting, American studies have shown decreased anxiety in paediatric populations with a CCLS present in the hospital [2,13-15] , allow for children to complete intimidating procedures [14,16-18], and allow for children to bypass abrasive treatments (ie. sedation) [15,20,21]. Studies in Canada have adopted similar methodologies and reached similar outcomes [1,22,23], which points to the efficacy of CCLS interventions surpassing global differences and being a robust intervention. However, much of the existing research remains small-scale or descriptive, highlighting the need for more rigorous empirical studies to confirm and extend these findings. While these benefits highlight the value of CCLS interventions, the literature also reveals several ongoing challenges that may limit their full potential.

CHALLENGES & FUTURE DIRECTIONS

Despite the documented advantages of CCLS interventions in numerous studies, both CCLS and its broader community encounter significant challenges. Certain themes echoed in the literature include; 1) insufficient empirical examination of their services, 2) a lack of a diverse CCLS workforce, and 3) compassion fatigue & burnout. There is particularly a lack of robust, outcome-based research assessing the measurable impact of specific CCLS interventions. Much of the existing literature relies on qualitative approaches or anecdotal insights, with limited quantitative, intervention-focused studies that can demonstrate causality or support large-scale implementation.

A scoping review by Boles et al ., 2021 concluded the lack of empirical evidence about CCLS in four databases. From 1998 to 2017, an increase was seen in the number of articles that made mention of Child Life. Yet, only 14% of these articles had content surrounding Child Life and 2% of these articles focused on Child Life [24]. Mentions of CCLS typically ask child-life professionals about their opinions regarding relevant topics within the hospital or important subjects in paediatric care [25,26]. In the absence of robust empirical evidence substantiating the efficacy of CCLS and their interventions, policymakers and healthcare professionals may find themselves lacking the necessary foundation to effectively advocate for the comprehensive integration of CCLS within the broader healthcare landscape. Without empirical validation of the tangible benefits and positive outcomes associated with CCLS services, the potential for optimizing paediatric patient care and enhancing overall healthcare outcomes may remain unrealized, impeding progress towards a more holistic and patient-centered approach to healthcare delivery.

An important consideration to make in healthcare delivery is the diversity of a profession's workforce. It is known that a workforce must be as diverse as the population it aims to serve [27]. Currently, the CCLS workforce is characteristically Caucasian, female and 34 years of age. This can pose a challenge in serving the diverse populations of Canada [8]. By mitigating other challenges associated with the role, it is anticipated that the job description will resonate with a more diverse array of populations, thereby narrowing this gap. In fostering a more inclusive and welcoming environment within the profession, coupled with targeted outreach and recruitment efforts, the potential exists to attract individuals from diverse backgrounds who may not have previously considered a career as a CCLS. Furthermore, by embracing diversity within the CCLS workforce, healthcare institutions can better meet the unique cultural, linguistic, and social needs of the diverse populations they serve. Many developmental theories in CCLS education also lack cultural context [28]. This deficiency underscores the critical necessity for an updated curriculum that places a heightened emphasis on equity, diversity, and inclusion. By integrating cultural considerations into the educational framework, CCLS programs can better prepare future professionals to effectively navigate and address the diverse needs of the paediatric populations they serve. Moreover, an updated curriculum that prioritizes equity, diversity, and inclusion not only enhances the cultural competence of CCLS practitioners but also ensures that their interventions are culturally responsive and relevant. A proactive approach is essential for promoting positive outcomes and fostering trust and rapport with patients and their families from varying cultural backgrounds.

Given the inherently empathetic nature of their role, many CCLS experience compassion fatigue and burnout for their day-to-day roles. Consequently, the prevalence of compassion fatigue and burnout among CCLS underscores the imperative for comprehensive support mechanisms and strategies to safeguard their emotional well-being and professional resilience. In a cross-sectional survey, it was reported that CCLS that frequently consulted with colleagues and a multidisciplinary team was a protective factor from compassion fatigue and burnout symptoms [29]. This points to the necessity of seamlessly integrating CCLS within a healthcare team. By fostering a cohesive and supportive interdisciplinary environment, healthcare institutions can not only mitigate the risk of burnout among allied health professionals like CCLS but also enhance the overall quality of patient care. Ultimately, this integration not only protects the well-being of healthcare providers but also enhances the quality and continuity of care for the vulnerable paediatric patients and families they serve.

In addition to these workforce-related challenges, structural barriers such as inconsistent funding, limited job positions, and a lack of standardized policies across provinces also hinder the full integration of CCLS services. Hospitals in smaller or rural communities may not have the financial resources to employ dedicated CCLSs, despite the demonstrated benefits [5]. National policy support and dedicated healthcare funding streams for psychosocial care are essential to ensure equitable access to CCLS interventions across Canada.

CONCLUSION

In summary, the role of CCLS in guiding infants, children, youth, and families through the complexities of illness, trauma, disability, loss, and bereavement is pivotal within the healthcare landscape. While historically confined to hospital settings, the expanding scope of CCLS practice now encompasses diverse healthcare environments across Canada, including community programs and academic institutions. Employing a multifaceted approach within six distinct domains of care, CCLS utilize a diverse array of evidence-based interventions tailored to address the psychosocial needs of paediatric patients and their families.

Despite the demonstrable benefits of CCLS interventions, notable challenges persist. These include the paucity of robust empirical evidence validating their effectiveness, the imperative for a more demographically diverse workforce reflective of the populations served, and the pervasive issue of compassion fatigue and burnout among CCLS. Addressing these challenges necessitates the adoption of a proactive and interdisciplinary approach that prioritizes equity, diversity, and inclusion. Efforts to integrate cultural competencies into CCLS' education and practice are paramount for ensuring culturally responsive care delivery. Furthermore, fostering a supportive interdisciplinary environment within healthcare teams holds promise for mitigating the risk of burnout and enhancing the quality of patient care.

In addition to these workforce-related challenges, structural barriers such as inconsistent funding, limited job positions, and a lack of standardized policies across provinces also hinder the full integration of CCLS services. Hospitals in smaller or rural communities may not have the financial resources to employ dedicated CCLSs, despite the demonstrated benefits. National policy support and dedicated healthcare funding streams for psychosocial care are essential to ensure equitable access to CCLS interventions across Canada. Collaboration between hospital leadership, professional associations, and provincial health ministries will be critical in ensuring that CCLSs are recognized, funded, and strategically placed to support all pediatric populations.

By prioritizing the well-being of CCLS practitioners and advocating for their seamless integration within healthcare systems, institutions can better meet the psychosocial needs of paediatric patients and their families, thereby improving health outcomes and advancing the overarching goals of patient-centered care. Ultimately, strategic investment in CCLS education, research, and institutional integration is essential to achieving equitable and holistic paediatric healthcare in Canada.

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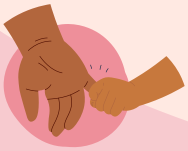



INVESTIGATING THE IMPACT OF MATERNAL-INFANT BONDING IMPAIRMENT DUE TO POSTPARTUM DEPRESSION ON CHILD IQ AND ADHD OUTCOMES

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ABSTRACT

Postpartum depression (PPD) is a psychiatric disorder following childbirth that is associated with health complications, such as chronic depression, changes in sleep patterns and appetite, and feelings of worthlessness, when left untreated. While PPD directly influences the mother, there are underlying consequences on child cognitive development, such as a link with lower child Intelligence Quotient (IQ) scores. This review aims to better understand the impact of PPD on maternal-infant bonding, and how this may influence child Intelligence Quotient (IQ) scores and attention-deficit hyperactivity disorder (ADHD) outcomes. A literature search on PubMed, OVID Medline, Cochrane Library, and Google Scholar was conducted using keywords relating to PPD, ADHD, and IQ. Current literature has suggested that PPD-induced impaired maternal-infant bonding is a risk factor for child ADHD outcomes and is associated with lower child IQ scores. However, there are limitations to consider, such as the validity of IQ scores, as the association between PPD and impaired maternal-infant bonding on child IQ scores is complex and multi-faceted. There are many potentiating risk factors and confounders that may influence the mother's experience of PPD and the developmental outcomes in the offspring.

INTRODUCTION

Maternal postpartum depression (PPD) is a psychiatric disorder consisting of depressive symptoms that persist for at least two weeks and interferes with the mother's ability to return to normal function following postpartum [1,3]. Symptoms of PPD consist of depressed moods, emotional withdrawal, changes in sleep patterns and appetite, feelings of worthlessness, psychomotor agitation, and other symptoms that may have a severe impact on the mother's mental state, quality of life (QoL), and relationships with others [1,4].

Maternal-infant bonding dynamic refers to the quality of emotional connection and interaction between the mother and her offspring [3]. When PPD is left untreated, it may lead to impaired bonding dynamics between the mother and her offspring, as PPD may hinder the mother's ability to engage and interact with her child by responding to infant cues [3]. Maternal PPD has been correlated with a decrease in responsiveness to infant cues and needs, and disengaged parenting outcomes that are thought to contribute to lower child Intelligence Quotient (IQ) scores and attention-deficit hyperactivity disorder (ADHD) outcomes [3]. It is also important to note that not all mothers with PPD show poor responsiveness or disengaged parenting behaviours that may lead to these outcomes [3].

Although PPD is a severe health concern for both the mother and her child, this paper will primarily focus on the complications of PPD on the offspring [1,2]. Currently, to this paper's knowledge, there are no reviews written in English with a specific focus on investigating the effects of impaired maternal-infant bonding due to maternal PPD on either child IQ scores or ADHD outcomes. Thus, this paper seeks to provide a review of recent literature to address the three following research inquiries: 1) What is the impact of PPD on maternal-infant bonding dynamics? 2) How does PPD-induced impaired maternal-infant bonding affect child IQ scores? and 3) Is there an association between PPD-induced impaired maternal-infant bonding and ADHD?

RESULTS

The Influence of PPD on Impaired Maternal-Infant Bonding

The relationship between postpartum depression (PPD) and impaired maternal-infant bonding is well-established in current literature [5]. Research suggests that PPD may negatively affect the bonding dynamic between the mother and her offspring due to a decrease in maternal responsiveness and engagement [5]. Gilden et al. involved sixty-four women with PPD, 61.1% of whom reported self-perceived impaired bonding with their offspring that could result in "undesirable" bonding experiences with their child [5]. In this study, "bonding" referred to the mother's feelings towards her child, with impaired bonding deemed to be undesirable, as it could impact the child's cognitive long-term development [5]. However, it is important to note that bonding was measured using the self-administered Postpartum Bonding Questionnaire (PBQ), thus the results may not entirely translate to the actual quality of maternal-infant bonding. Other studies have found similar findings, including an association between impaired maternal-infant bonding and child behavioural challenges during early childhood, as well as a risk for maltreatment later in life [6,7].

Mother and Baby Units to Improve Maternal-Infant Bonding:

Although impaired maternal-infant bonding can result in undesirable child developmental outcomes, an adequate treatment regime will usually improve bonding in mothers with PPD [5]. In the study by Gilden et al., a significant association between a decrease in depressive symptoms and improvement in maternal-infant bonding was identified among participants with PPD over an eight-week period following admission to a Mother and Baby Unit (MBU) [5]. MBUs are non-pharmacological interventions that involve guidance from nurses regarding ways mothers can engage with their offspring, connect with support groups, and learn from video-based instructions [5]. The aim of MBUs is to facilitate healthy mother-infant relationships and provide inpatient psychiatric support for mothers and their infants [8]. Moreover, a systematic review involving twenty-three studies investigated the outcomes for women admitted to MBUs [8]. The results indicated that higher levels of insecure attachment in infants were observed in mothers with PPD and other affective disorders which are characterized by severe disruptions in emotions; however, the quality of mother-infant interactions generally improved following MBU admission. Quality was based on the mother's engagement with her offspring and her response to infant cues [8]. In addition to MBUs and other non-pharmacological interventions, pharmacological treatment options, such as tricyclic antidepressants or selective serotonin reuptake inhibitors, are commonly prescribed medications to treat depressive symptoms of PPD and improve maternal-infant bonding [3]. Thus, adequate treatment plans are important to improve the well-being of the mother and her offspring and mitigate the effects of PPD on impaired maternal-infant bonding.

Relationship Between PPD and Child IQ Scores

Several studies have indicated a negative association between maternal PPD symptoms and child cognitive development [2,9-11]. PPD has been linked to impaired maternal-infant attachment and bonding that is thought to contribute to changes in cognitive developmental outcomes, particularly lower IQ scores in children [2,9]. Mothers diagnosed with PPD were found to exhibit a decrease in sensitivity to infant cues and response to fulfilling infant needs [10]. A study by Sharp et al. [11] found a relationship between a lack of maternal engagement and responsiveness due to PPD and lower IQ scores in children. Children of mothers with PPD demonstrate a greater likelihood of developing attentional deficiencies, difficulties with Piaget's object permanence tasks, and other cognitive-related tasks [11]. During the first year of maternal PPD, children demonstrated a decrease in performance on standardized tests of intellectual attainment at four years old; however, these lower IQ scores were not displayed in children of mothers who became depressed after the first year of PPD [11]. Although further research is required to understand the relationship between the onset of PPD symptoms on child IQ scores, these results suggest that maternal PPD has a greater effect on child IQ scores during infancy than later stages of child development.

Sex Differences in IQ Scores:

Another study found that three-year-old children of mothers with maternal PPD at six months postpartum had a notable difference in full IQ scores, but no major difference in performance IQ (PIQ) or verbal IQ (VIQ) [12]. Full IQ is composed of both PIQ and VIQ, which can be further broken down into subcategories, and measure an individual's visuospatial intellectual abilities and spoken language abilities, respectively [12]. Interestingly, the three-year-old male participants were disproportionately affected by maternal PPD than female children, as they exhibited the lowest full IQ scores in the study [12]. A meta-analysis by Sui et al. [13] found similar results; significant differences in cognitive test scores were identified in male children, whereas no significant differences were observed in female children's test scores. It is hypothesized that male children are more sensitive to the effects of PPD partly due to maturational disadvantages and differences [13]. However, some studies found conflicting research that indicated an insignificant difference in IQ scores among male and female children [11]. Based on the inconclusive findings, there is insufficient evidence to prove whether there is a difference between maternal PPD and the child's IQ in males compared to females. However, there is consistent research evidence that demonstrates that PPD is a predictor for lower child IQ scores.

Brain Activity:
It is hypothesized that the negative relationship between PPD and lower IQ scores could be explained by an infant's electrical brain activity and a lack of synchrony and reciprocity between an infant's actions and maternal response [13]. Research suggests that infants of mothers without PPD demonstrated a greater likelihood of exhibiting electrical activity within the left frontal cortex, whereas infants of mothers with PPD are more likely to exhibit electrical activity within the right frontal cortex [13]. These findings are significant as electrical activity in the left frontal cortex has been shown to establish positive emotions and "normal" behaviour, whereas electrical activity in the right frontal cortex is more responsible for negative emotions and feelings of distress [13,14]. As a result, greater activity expressed in the left frontal cortex in the infants of mothers with PPD is linked to higher rates of distress, negative emotions, and reactions that may have a negative impact on the infant's ability to react to novel stimuli and hinder learning abilities [14,15]. The change in electrical brain activity in the offspring may result in frequent distress that prompts cognitive or intellectual developmental delays [14,15]. However, more research on the brain activity in infants or children of mothers with PPD is needed to confirm this hypothesis.

Impaired Maternal-Infant Bonding and Child ADHD Outcomes

Attention-deficit hyperactivity disorder (ADHD) is a psychiatric condition that tends to involve inappropriate levels of inattentiveness and hyperactivity, difficulty concentrating, disorganization, and other symptoms that affect a child's ability to function [16]. Symptoms of ADHD can be broadly categorized into inattentive- and hyperactive symptoms [16]. Inattentive symptoms include difficulties sustaining attention, missing small details, avoiding tasks that require more cognitive effort, and forgetfulness [16]. Hyperactive symptoms include fidgeting, inability to engage in activities quietly, and difficulty waiting for their turn in group situations [16]. ADHD may also affect an individual's ability to function through procrastination, low-self-esteem, mood instability, and difficulty controlling emotions [16]. It is also important to acknowledge that ADHD may manifest differently among individuals, with variations in symptom prevalence, severity, and impact, reflecting a spectrum of experiences that can change over time [16,17].

According to study findings, PPD is a risk factor for child ADHD as PPD may result in symptoms of impaired maternal-infant bonding, including maternal inattentiveness, inability to cope with the care of the child, and a lack of nurturing behaviour [3,17]. However, the specific relationship between a lack of nurturing behaviour as a result of PPD on child ADHD outcomes is not entirely known [3,17]. A meta-analysis that investigated the general relationship between PPD and child ADHD found a statistically significant correlation between PPD and ADHD with a 95% confidence interval of 1.27 to 2.26 among nine cohort studies, which indicates that there is a 95% likelihood that children were 1.27 to 2.26 times more likely to have ADHD or exhibit inattentive or hyperactive ADHD symptoms if their mother was diagnosed with PPD compared to mothers without a PPD diagnosis [3].

Bowlby's Theory of Attachment:
Bowlby's theory of attachment highlights the significance of the dyadic relationship between the mother and her infant in shaping early attachment, which refers to the emotional bond that forms between the mother and her child during early years of life [18,19]. This early attachment influences cognitive and emotional development, as well as internal working models that shape future relationships [18]. Infants rely on their mothers for engagement, interaction, and emotional regulation, making the quality of early attachment an important factor in psychological well-being and social development [18,19]. Research suggests that variations in attachment styles may be associated with differences in impulse control, patience, self-regulation, and self-soothing abilities, which are traits linked to ADHD [18,19]. PPD has been connected to disruptions in maternal-infant engagement, which may influence these developmental processes [18,19].

Early Breastfeeding Cessation and Child ADHD Outcomes:
Breastfeeding is an important physiological and emotional bonding experience between mothers and their infants [18]. Mothers with PPD tend to engage in less healthy nurturing habits such as early breastfeeding cessation, which is a risk factor for child ADHD [18]. Current literature suggests that breastfeeding has protective properties against child ADHD, which could account for the increased likelihood of child ADHD outcomes when mothers are unable to consistently breastfeed for an adequate period [18]. Furthermore, a prospective study by Hamdan et al. found a reciprocal relationship between breastfeeding and a reduced risk of PPD [20]. The results of the study found that: 1) mothers who were breastfeeding after two months following postpartum had a lower risk for PPD in later months of postpartum, and 2) mothers with PPD were linked with a decreased period of breastfeeding [20]. Another study found a similar association between having higher depressive symptoms at two weeks following postpartum and early breastfeeding cessation around three-months postpartum [21]. It is hypothesized that a reciprocal relationship exists between PPD and reduced breastfeeding rates, as early breastfeeding cessation may exacerbate the risk and duration of PPD [19,21]. According to these studies, children of mothers with PPD are more likely to exhibit ADHD symptoms due to early breastfeeding cessation; however, the specific mechanisms and strength of this correlation are not well understood [19,21].

disabilities reported feeling very or extremely concerned about managing their child's behaviour, stress, anxiety, and emotions.[17]

Lastly, experts in the field are also worried that the rates of FASD may rise during the COVID-19 pandemic due to increased alcohol consumption. The Canadian Centre on Substance Use and Addiction revealed that 20% of Canadians have increased their alcohol consumption during the pandemic due to the lack of routine, boredom, stress, and loneliness. Furthermore, women have been found to consume more drinks than recommended per day according to Canada's Low Risk Alcohol Drinking Guidelines.[15] Additionally, intimate partner violence, a risk factor for FASD, as it may increase maternal alcohol consumption, has increased during the pandemic.19 Lockdown and stay-at-home measures have increased safety concerns among women who live with controlling or coercive partners, and it has been found that the number of calls to women's support services has significantly risen during this time.[18] With both the rise in alcohol consumption and intimate partner violence, there may be a potential increase in alcohol-exposed pregnancies.[18] Strategies that can help prevent FASD during the pandemic are inclusion of FASD education and prevention in public health messaging, provision of free contraceptives, and collaboration among healthcare providers for substance use and intimate partner violence to provide holistic care to women at risk of substance use during pregnancy.[18-19]

Hence, it is clear that the COVID-19 pandemic has impacted FASD in a number of facets, including the disruption of support services for children with FASD and their caregivers, the impact of social isolation on the mental health of children with FASD, and the potential increase in FASD prevalence in the near future.[12,15]

CURRENT RESEARCH GAPS & FUTURE DEVELOPMENTS

Given the short-and long-term implications on a child's cognitive development, alongside the complexities of FASD, over 90% of individuals diagnosed with this disorder are estimated to experience mental health challenges.[20] Despite the prevalence of such poor implications, little research has investigated the impact of interventions that address these persistent challenges for youth with FASD. In fact, a recent systematic review conducted in 2020 aimed at exploring the effectiveness of mental health and substance use interventions, revealed that only three out of 10,000 screened studies assessed their efficacy in FASD individuals.[20] The need for these future developments are imperative during this time, given the impacts of COVID-19 in exacerbating mental health challenges for these individuals. Thus, future research is warranted to investigate the effectiveness of such interventions in FASD youth specifically. Apart from the poor mental health outcomes that many children with FASD experience, many other repercussions associated with the primary and secondary disabilities of FASD also exist. While their implications have been well-established, again, there is limited research on specific behavioural and cognitive strategies that may improve cognitive functioning. Developing a deeper understanding of the effectiveness of these strategies is critical to the implementation of intervention programs that can improve the physiological, psychosocial and physical health outcomes amongst youth with FASD.[21] Moreover, pharmacological interventions are often prescribed to manage the behavioural issues associated in children with FASD. However, if specific brain receptors that these medications act on are altered by prenatal alcohol exposure, then they may be ineffective.[6] Thus, future research should be aimed at developing a deeper understanding of the various changes that FASD can cause at a cellular level in order to facilitate more effective pharmacological interventions for youth with FASD.[22]

Given the rising concerns of caregivers, especially during the COVID-19 pandemic, it is pertinent that stronger support services are put in place. The 2015 Ontario provincial roundtable report on FASD found very little to no immediate crisis support or services outside of business hours.[23] Furthermore, participants of this roundtable report also called for more funding for services so that families and caregivers have increased access to supports that would assist them in coping with their mental health challenges as well. Finally, support currently exists in silos. As such, streamlining resources for children with FASD and their caregivers will be essential to reduce barriers in accessing information about these programs. For example, participants recommended establishing a one-stop 24/7 source where they could obtain the most updated information about FASD services, such as a national website or toll-free number.

By improving the current research gaps and enhancing support services, caregivers and children with FASD will be more cared for, thus leading to better outcomes with the child's cognitive and overall health development. Stakeholders, such as governmental jurisdictions, are beginning to see these gaps and take action for the future. For example, the Ontario government is investing $10.1 million in annual funding towards FASD programs and services and diagnostic services for people impacted by FASD.[24] This funding will go towards expanding FASD diagnostic services through new clinics and programs.

DISCUSSION

There are existing meta-analyses that examine the relationship between postpartum depression (PPD) and child attention-deficit hyperactivity disorder (ADHD) outcomes, as well as the relationship between PPD and lower child Intelligence Quotient (IQ) scores. However, research specifically exploring the impact of PPD-induced impaired maternal-infant bonding on these developmental outcomes remains limited. While PPD can influence child IQ and ADHD risk through multiple pathways, this paper focuses solely on one specific mechanism: the role of impaired maternal-infant bonding.

Studies have demonstrated an association between maternal PPD and decrease in maternal-infant attachment and bonding, which may affect reasoning and problem-solving abilities [2,9]. A lack of maternal engagement during early stages of child development has been linked to a decrease in performance on standardized tests of intellectual attainment, particularly in children of mothers who are diagnosed with PPD within the first year of postpartum [11]. There are conflicting findings on the influence of sex differences in IQ scores among children [11]. While some studies have found that male children appear to be disproportionately affected by impaired maternal-infant bonding, exhibiting lower full IQ scores than female children, others suggest that the difference is not statistically significant [11-13]. Thus, further research is needed to determine if PPD may influence the IQ scores of male and female children differently. Although the studies evaluated in this paper demonstrated that PPD has a negative association with lower IQ scores and poorer performance on standardized tests, understanding the limitations of these tests and potential confounders are necessary for interpreting these findings accurately [2,9-11]. Such confounders may include the child's sex, birth order, socioeconomic status, and family adversity [11].

Although IQ tests are an important indicator of intelligence as they are designed to measure cognitive functioning abilities, such as comprehension, reasoning, and judgment, there are limitations associated with IQ tests [22,23]. Researchers find that IQ tests may fail to accurately measure more cognitively complex or nuanced aspects of mental functioning, which may influence the predictive validity of IQ scores [22]. Additionally, there are external factors, like motivational influences and affective states, that may drive variations in performance on IQ tests [22]. Studies demonstrate that individuals who are more motivated, open to new learning experiences, and willing to search and process information, may perform better on IQ tests than if they were unmotivated to do so [22,23]. Affective states refer to the emotional state and expression of a person and can also influence IQ test performance [22]. Negative affective states, such as test anxiety and stress, are associated with lower IQ test performances [22]. Thus, when evaluating the IQ of a child, it is important to evaluate the validity of these tests, and how this may influence our understanding of the relationship between maternal PPD and offspring cognitive development.

Furthermore, although it is unclear if a causal relationship exists between PPD-related impaired maternal-infant bonding and ADHD outcomes, a correlation has been identified by several studies [16,18,19]. PPD increases the likelihood for a child to exhibit insecure attachment styles and receive a lack of maternal attentiveness, which are associated with symptoms of ADHD, such as having impulses, non-self-regulation, and impatience [18,19]. PPD has also been associated with early breastfeeding cessation that may negatively affect the bonding experience between the mother and infant [18]. Literature suggests that breastfeeding has protective properties against ADHD and has found an association between early breastfeeding cessation and an increased risk of ADHD in the offspring [18]. While associations have been identified between PPD and early breastfeeding cessation, as well as between early breastfeeding cessation and increased child ADHD, more research is needed on how the interconnected relationship between all three considerations (PPD, early breastfeeding cessation, and child ADHD outcomes) might affect the lives of mothers and their children [18,19].

CONCLUSION

The findings of this review suggest that impaired maternal-infant bonding due to maternal postpartum depression (PPD) is associated with lower Intelligence Quotient (IQ) scores and attention-deficit hyperactivity disorder (ADHD) in children. Adequate treatment plans are necessary to improve maternal-infant bonding, as well as the general health and well-being of mothers with PPD and their offspring. While a correlational relationship exists between PPD-induced impaired maternal-infant bonding and lower child IQ scores, as well as increased risk of child ADHD, evidence supporting a causal relationship is limited. Thus, to better understand the relationship of impaired maternal-infant bonding and maternal PPD on child IQ and ADHD outcomes, further research is required to explore the underlying mechanisms, assess the long-term impacts, and develop targeted interventions that support maternal mental health and promote healthy developmental trajectories for children.

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THE IMPACT OF MUSIC EDUCATION ON STUDENTS' COGNITIVE DEVELOPMENT OF CREATIVITY IN ELEMENTARY SCHOOLS

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ABSTRACT

Music education in the elementary ages of a child's life has proven to be connected to cognitive outcomes. However, how it influences a child's creativity is a field that has not been thoroughly researched. Understanding the impact of musical education on a child's neurological development and creativity is paramount to gaining a holistic understanding of cognition in children. Literature exploring the development of divergent thinking, executive functions (EF), and neurological functioning from music training provides insight into the multiple ways that music impacts cognition. Databases including OVID Medline, ScienceDirect, and PubMed were searched , and the CRAAP test was used to assess validity. Findings suggest a strong positive correlation between high intrinsic motivation levels among students in music education and divergent thinking skills. Neuroimaging results pointed out an underlying co-activation in the Inferior Frontal Gyrus, a brain region associated with divergent thinking and creative idea generation, particularly evident during musical improvisation [1]. Results also indicate varying strengths in associations between music training and the development of three EFs (working memory, inhibition, and cognitive flexibility). Greater cognitive flexibility and lesser inhibition are also correlated with creativity, thus demonstrating the need to further research the relationship between EFs, music education, and creativity. Musical education is also associated with increased cognitive functioning, including auditory processing, reasoning, and language abilities. Additionally, certain styles of musical education are hypothesized to increase perseverance and attention. Such findings imply that further research can be done to understand the mechanisms through which musical education impacts creative thinking in children and would be useful for further applications of musical education in educational and recreational settings.

INTRODUCTION

Within multiple theoretical frameworks that pertain to the evolution of children, the concept of creativity is found intricately weaved into the field of cognition. For example, in Piaget's theory of cognitive development, the emergence of symbolic and imaginative play is seen in the preoperational stage that is often associated with ages 2-7 – two concepts which are associated with creativity [1]. In Vygotsky's sociocultural theory, a child's interaction with their cultural surroundings is hypothesized to shape their cognitive development [2], and a child's creative abilities evolve with increased life experiences [3]. Defined by Li as "the ability to generate novel and potentially useful ideas or solutions to problems" [4], creativity in education, or the lack thereof, has become a prominent concern due to heavy reliance on test scores (i.e., IQ scores) within the elementary school curriculum [5]. The elementary school grade range is defined from kindergarten to grade 8 (approximately ages 4 to 14) [6]. To further opportunities and increase experience for students in creative expression, artforms such as music, drama, dance, and visual arts have been pushed to be integrated into early childhood education [7]. Particularly, the instrumental side of music education has been recognized for its notable influence on an individual's cognitive development. For example, according to Chan et al., adults who have had at least 6 years of musical training before the age of 12 recalled approximately 16% more words than those without, proving greater verbal memory [8]. However, literature on the relation between music and cognition in traditional education is still considered novel, and consequently under-addressed. Thus, this review aims to explore the impacts of elementary music education on children's divergent thinking, executive functions, and neurological functioning to elucidate how musical training can influence a child's cognitive development of creativity.

METHODS

In pursuit of our objective, we conducted a comprehensive database search associated with music education and its relations to traditional elementary education and cognitive-creativity development. Databases including OVID Medline, ScienceDirect, and PubMed were searched , and boolean operators were employed to ensure data relevancy and accuracy. MeSH terms were also integrated to increase specificity and filter large quantities of database results, with search terms pertaining to elementary education, children's cognition, and musical training. To ensure reliability, citations were reviewed and assessed using the CRAAP test. Recovered literature was separated into article types, such as trials or opinion articles, to ensure separation of factual evidence from personal perspectives. For the review's inclusion/exclusion criteria , regarding exclusion, articles outside of North America (i.e., Canada, United States) were not included, with exception of literature from the United Kingdom and Hong Kong that were only included to add details to the current evidence discussed in this review. With regards to the inclusion criteria , only studies from 1991 to 2023 were incorporated into the review to cover a broad spectrum of findings and trends relevant to our objective.

DIVERGENT THINKING

Introduction to Divergent Thinking

The ability of Divergent thinking (DT) is considered an educational skill that is pronounced in younger children and decreases with age. It is a critical component in developing creative thinking. DT involves the use of a thought process that enables students to scan a problem or concept from multiple directions, helping them to free-flowingly generate solutions [9]. It differs from convergent thinking, which seeks a single correct answer, by encouraging exploration of various perspectives and unconventional ideas.

Experiment on Divergent Thinking: Impact of Intrinsic Motivation

A study by Wolfe et al. investigated intrinsic motivation and divergent thinking in music education within forty Midwestern third graders who demonstrated high convergent ability [10]. They measured the children's Intrinsic Motivation Level (IML) by observing time spent engaged in music-making.

The results indicated a statistically significant relationship between intrinsic motivation and divergent thinking. In divergent thinking challenges, students with higher IML scores outperformed those with lower intrinsic motivation levels. Specifically, the High Intrinsic Motivation group (HIM) scored significantly higher on the Measures of Creative Thinking in Music Tests (MCTM) compared to the Low Intrinsic Motivation group (LIM) ($t = -2.79$, $p < .01$). These findings suggest that intrinsic motivation crucially enhances divergent thinking.

Improvisation in Music Education - Benefits and Integration

As technology progresses, North American elementary music education reflects other aspects of divergent thinking. This is seen through varying experiments showcasing musical improvisation as a core component of North American music education curriculum . Pedagogical approaches such as the Kodály Method further establish its importance [9,11]. Notably, a study by Lewis examined divergent thinking scores of instrumental music students in the United Kingdom with improvisation instruction and observed an increase in divergent thinking for fluency, originality, and flexibility [9]. Contrastingly, the control group had no increase in DT, suggesting that improvisation can encourage individuals to break from schemas. This finding potentially extrapolates to North American practices due to the shared use of the Kodály Method.

Moreover, neuroimaging studies captured in reviews by Sowden et al. have consistently shown activation in the Inferior Frontal Gyrus (IFG) during musical improvisation, an aspect of the brain that is connected to forming creative ideas via divergent thinking [12]. In addition, Beaty et al.'s research demonstrated statistical significance in divergent thinking, predicting 57% of the variance in expert judgment regarding the creative quality of improvisation [13]. The co-activation of IFG suggests a common mechanism that underlies divergent thinking and musical improvisation.

EXECUTIVE FUNCTIONS

Executive functions (EF) are a group of cognitive operations that lead to purposeful and goal-oriented behaviors. They recruit several brain structures, particularly among the prefrontal cortex [14,15,16]. EFs play a critical role in an emerging child's cognitive development as they contribute to learning and behavior [15]. Furthermore, there are 3 core brain functions that make up executive functions in children: inhibition, working memory and cognitive flexibility [14]. Inhibition encompasses a child's ability to resist impulsive actions [14,15]. Working memory (WM) is defined as the capacity to retain information over short time durations while also achieving other cognitive tasks [14,15,17]. Lastly, cognitive flexibility is the ability to shift attention between different behaviors and mindsets, especially in adapting to change [14,15].

The systematic review by Rodriguez-Gomez et al. evaluated various EFs in three age groups: preschool, school-age, and adolescent [14]. Among these categories, 23 studies yielded results on inhibition, 19 studies yielded results on WM and 17 studies yielded results on cognitive flexibility. The authors identified a greater benefit of music training in inhibitory control while there were more mixed results for the remaining 2 core EFs. Studies evaluating the relationship between music training and inhibition ranged from a variety of methodologies including Go/No-Go, Stroop, Simon, and the NEPSY tests. These tests are cognitive and neuropsychological assessments designed to evaluate key aspects involved in executive function such as inhibition, selective attention and cognitive control. For instance, in a 2021 Canadian study examining 50 children, children in the non-music intervention group demonstrated greater progress in NEPSY inhibition than in the corresponding control and motor groups. Authors hypothesize that the role of attention, control, and integration of multiple sensory stimuli in music training contribute to this positive trend. Additionally, three of eight studies examining working memory yielded positive results in preschool children while the remainder of studies involving school-age children and adolescents did not identify significant differences between music and control groups.

A similar trend was observed in the case of cognitive flexibility. Some studies in preschoolers were able to identify statistically significant improvement in several cognitive tasks (DCCS, WCST, or the NEPSY-II subtest: "Animal Sorting.") while no associations were found in larger sample sizes of adolescents and school-aged children. Moreover, the studies which focused on preschool children exhibited a relationship between music training and EF development. However, improvements were found to decline with increasing age, especially among adolescents. This can be partly attributed to the extensive maturation and overall neurodevelopment occurring during the ages 3-5. More research can help better explain the effect of age on EFs and music training. In addition, it is important to consider that there exist several challenges in this field of clinical research as the definitions of music education can be variable from study to study and certain variables are not standardized such as measures of creativity. Overall, the complexity of these results can be clarified with further research and new hypotheses examining more focused types of music education.

A 2023 systematic review by Pasarín-Lavín et al. elaborates on the relationship between EFs and creativity in children and adolescents [18]. A number of papers demonstrate that creativity is positively correlated with flexibility and negatively correlated with inhibition. Authors suggest that this is because a person with lesser inhibition and greater flexibility demonstrates a higher creative capacity. In comparison, there was insufficient evidence to support a relationship between working memory and creativity. Authors also highlighted the need for standardized tests as the variability between studies can limit the conclusions made. Limitations included potentially objective measures of creativity, a limited article sample and a risk of publication bias demonstrated by the overwhelming positive results in research literature.

All in all, the results from the systematic reviews conducted by Pasarín-Lavín et al. and Rodriguez-Gomez et al. emphasize the implications of this field of research on a child's development as there is a complex interplay between a child's education and cognitive skills.

NEUROLOGICAL FUNCTIONING

Music engages the brain in a variety of ways, and the cognitive abilities that it develops can be transferred to other domains of creative thinking and neurological function via transfer effects. Evidence collected by Hille et al. Has shown an association between musical education and higher cognitive functioning. Specifically, their data suggests an association between musical training, general abilities, and spelling abilities [19]. In a retrospective study observing children between the ages of six and eleven, Schellenberg et al. observed a correlation between full-scale IQ scores and academic performance in children who received keyboard or signing lessons (20). In the study conducted by Forgeard et al. observing eight-to eleven-year-old children, it was .found that children who had received three years of music training performed better than the control group in the assessment of fine motor skills and melodic discrimination. Furthermore, results from this study suggest a correlation between receiving musical training during childhood, and increased IQ levels, academic performance, vocabulary size, and reasoning scores [21]. Schellenberg et al. examined the impact of music lessons on enhancing an individual's sensitivity to emotions. The ability of six-year-old children to identify anger and fear was studied, and it was observed that children who received one year keyboard lessons performed better than children who received no lessons [22]. Musical training impacts the development of auditory processing in the auditory cortex, which may explain the findings that Fujioka et al. observed between musical training, general intelligence, and auditory processing in four to six-year-old children. Children with musical training exhibited differences in the processing of violins sounds, which was demonstrated by a pronounced morphological change in neural responses during a time frame of 100ms to 400ms. This research suggests that children training under the Suzuki method may develop more advanced skills, or an increased abilities to sustain focused attention [23].

DISCUSSION

Divergent Thinking

Previous studies, such as Wolfe et al.'s, have questioned the reliability of divergent thinking's testing approaches [10]. Alternative methods of assessment (e.g., product rating) may be further explored to measure complex constructs like musical creativity to increase reliability. The variability of the grading system between elementary schools suggests a future researchable sector on the impact of music education's extrinsic rewards (e.g., grades and awards) on students' divergent thinking.

Although there have been previous studies establishing that IFG's pre-activation affects how the brain processes stimuli [11,13] , there are limitations;untested approaches within Sowden et al.'s study [12]. Acknowledging research gaps, these assumptions are: (1) that other types of improvisation besides musical improvisation similarly activate the IFG, and (2) pre-activation of the IFG during musical improvisation is long enough to influence subsequent divergent thinking tasks. This paper, Sowden et al., helps guide our approach by linking IFG activation to divergent thinking, which we examine through musical improvisation—a spontaneous process often associated with creative thinking. Longitudinal neuroimaging research, which assesses the duration of IFG activation, could provide validation but faces challenges such as a lack of a golden-standard model and interpreting complex growth patterns [24]. Tackling these challenges is key to advancing developmental cognitive neuroscience.

Executive Functioning

This review clearly indicates the complexities of a child's creative cognitive development in relation to music education [14, 18]. In current research literature, the relationship between EFs and creativity has been explored separately to EFs and music training. A beneficial impact of music training on inhibition was identified while a lesser impact was noted concerning cognitive flexibility and working memory [14]. Overall, there is an age-dependent trend of the benefits of music training as the strongest associations were demonstrated in preschoolers while weaker associations were shown in adolescents [14]. This calls for further research to be conducted to better understand this trend and the broader implications of a child's musical experiences on their cognitive development in their early life. It was also found that greater cognitive flexibility and lesser inhibition are correlated with creativity [18]. These mixed findings demonstrate the advantages and drawbacks of certain cognitive functions, especially inhibition.

Additional research in this emerging field can help make clearer connections between EFs, creativity and music training. This can have more meaningful implications for the design and execution of education programs in promoting optimal cognitive development. Nevertheless, the studies from both research questions had many of the same limitations, primarily the lack of standardization of variables [14, 18]. This can be found between cognitive testing strategies, follow-up times, music training times and key definitions. Thus, standardization can allow researchers to make more generalizable and reliable conclusions. In addition, it is beneficial to explore varying frameworks of creativity to improve the accuracy of data in testing hypotheses amidst several proposed models. This can help limit objective measures while gaining greater perspective on the research question at hand. It is also important to recognize how this field of research is of relatively new interest and thus presents gaps in knowledge. Thus, it would be beneficial to further explore these associations between music education, creativity, and EFs.

Neurological Functioning

There is extensive research comparing groups of children receiving music lessons to groups of children receiving different types of enrichment, such as sports, or to control groups who are not engaging in any type of enrichment [20]. Thus, the opportunity to compare and contrast the differences that exist between the impacts that different instruments have on cognition may be easily missed.

Though there is extensive research that compares children trained in music to those untrained, there is limiteded research to justify which instruments in music facilitate a larger impact on a child's cognition [21]. Additionally, many studies investigating the impacts of music on cognition often have correlative results, and do not present causal relationships [19,20,21]. Causal relationships are more reliable than correlational when making evidence-based conclusions. This may be due to difficulties limiting a study's confounding variables within this field. It can be hard to isolate intelligence as an outcome when factors such as family situation, income level, genetic predisposition, health condition, mental well-being, and the presence of learning disabilities may all impact a child's mental abilities, and therefore impact the way that their intelligence levels are perceived by researchers.

Another limitation observed in this topic is the challenge to standardize and interpret cognitive outcomes and measurements that quantify cognitive improvement. There are various outcomes observed, such as IQ scores and the completion of tasks involving detection and recognition of patterns, such as auditory discrimination [20,21]. Furthermore, many of the studies we consulted which examine the impact of music on cognition often have small sample sizes, which can reduce the generalizability of the findings [21,22]. Across this literature, there is a lack of standardization on what quantifies a music lesson, making this subject difficult to generalize. Factors such as the consistency, frequency, structure, objectives, and presence of an instructor are all things which should be considered when trying to determine the quantification of a music lesson.

Lastly, there is a wide range of research investigating the effect of music on outcomes like IQ and auditory discrimination, as they are easier to test for and quantify [21]. However, there is minimal research focusing on the impact of music on a child's creativity, making this a challenging area to measure amongst students.

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SCREEN SMARTS: THE FACTORS AND INTERVENTIONS OF MEDIA AND DIGITAL HEALTH LITERACY IN CHILDREN AND ADOLESCENTS

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ABSTRACT

As children and adolescents begin using social media at a younger age and social media becomes a prominent source of health information, media and digital health literacy are necessary to help children and adolescents critically evaluate the health information widely available on these platforms. Media and digital health literacy can provide a protective influence to minimize the risk of adverse health outcomes and disparities. This opinion review explores different factors influencing media and digital health literacy in children and adolescents, including education of adolescents and caregivers, self-efficacy, and socioeconomic status. Interventions such as incorporating media and digital health literacy education into school curriculums, health education programs targeted towards caregivers, and nontraditional education frameworks attempt to address these issues. This review explores successful interventions to identify effective strategies for improving media and digital health literacy and discusses the importance of involving children in research. By understanding key risk factors and effective strategies, this review hopes to inform the targeting and design of future media and digital health literacy programs.

INTRODUCTION

Social media has become an integral part of day-to-day life and an important medium for connecting people [1,2]. In 2023, most Canadians had a minimum of one social media account, with 33.1 million total social media users in Canada, out of the 40.5 million individuals living in Canada in 2023 [3,4]. Despite the potential benefits of social media, including the facilitation of community, connection, and self-expression, there are also negative consequences on users, with social media being found to impact adolescent mental health, sleep quality, self-esteem, diet, and the regulation of their behaviour [5,6,7]. A 2023 survey found that 40% of children in America aged between 8-12 years use social media, despite most social media platforms having a minimum age requirement of 13 years to create an account [5]. Uptake is even higher among adolescents that have met the minimum age requirement, with a survey conducted in 2018 in America finding that 97% of adolescents aged 13-17 use social media platforms [8]. Adolescents and children are eager to engage with social media and are regularly exposed to it. For the purpose of this opinion review, we define children to be prepubescent and adolescents as the transition between puberty and adulthood.

Social media platforms, most notably Facebook, YouTube and Instagram, have become popular tools for obtaining health-related information both actively and through passive exposure [9]. A Pennsylvania-based cross-sectional study found that while only 3.5% of adolescents reported using social media for health information, 66.7% to 91.7% of these respondents reported seeking health information on social media about topics such as fitness and sexual health when asked specifically about said topics [10]. However, the convenience of social media comes with a caveat, as it can be a significant source of misinformation and disinformation [11]. According to the Canadian Centre for Cyber Security misinformation is defined as, “false information that is not intended to cause harm” [12]. Disinformation on the other hand is false information that is created and spread deliberately with the intention to “manipulate, cause damage, or guide people, organizations, and countries in the wrong direction” [12]. Social media is filled with both mis- and disinformation which is more likely to be shared and reposted than accurate information.12 Disinformation is produced more rapidly and makes bolder, attention-grabbing claims compared to scientifically-backed content [12]. For instance, a post that claims a serious health condition has been completely cured will attract more attention than a carefully-worded post that explains the scientific process and limitations of the research. The spread of inaccurate health information can have detrimental effects, including delaying proper treatment to patients [13]. Given the increasing popularity of the internet and social media as sources of health information and the prevalence of mis- and disinformation on social media platforms it is important to develop media health literacy to provide adolescents with the skills required to critically and confidently evaluate online health information [11,24].

Media health literacy is defined as addressing or solving a health problem through the application of knowledge gained from health information found, understood, and appraised from society's means of mass communication.14 In contrast, digital health literacy is defined by the World Health Organization as, "the ability to seek, find, understand, and appraise health information from electronic sources and apply the knowledge gained to addressing or solving a health problem."15

Many risk factors overlap between poor personal health and low media and digital health literacy, including, but not limited to, age, race/ethnicity, socioeconomic status, and education level [15]. These lower levels of media and digital health literacy may lead to broadening existing gaps in health equity [15]. Adolescents with low health literacy have shown an increase in risky behaviours and obesity, and a decrease in healthy behaviours [16].

This opinion review will examine central factors related to media and digital health literacy and associated interventions with the goal of informing future strategies for developing these skill sets in children and adolescents. Central factors including self-efficacy, socioeconomic status, and the educational background of adolescents and their caregivers, were determined through the 2022 systematic review by Jindarattanaporn et al. and more recent research as this field is rapidly expanding [15,17]. In this review we will discuss the importance of identifying and analyzing the determinants contributing to low levels of media and digital health literacy and implementing targeted interventions. We will also discuss targeted interventions including incorporating media and digital health literacy education into school curriculums, health education programs targeted towards caregivers, and nontraditional education frameworks.

METHODS

The literature review process followed a structured approach to ensure comprehensive coverage of relevant studies. Initially, a broad scan of existing literature, including gray literature, was conducted using Google and Google Scholar to identify key themes and gaps in research. Following this preliminary stage, searches were performed using PubMed, Ovid MEDLINE, SAGE Journals, and Web of Science to retrieve peer-reviewed articles. Searches included terms related to media health literacy, digital health literacy, adolescents, children, factors influencing media health literacy, and interventions. The search terms were combined through truncations and Boolean operators, "AND" and "OR".

To assess research quality, studies were prioritized based on methodological rigor, relevance, and credibility. While a formal empirical assessment was not conducted, preference was given to peer-reviewed studies with clear methodologies, including systematic reviews and primary research. Review papers provided synthesized insights, while primary studies contributed specific data on media and digital health literacy in children.

Articles were eligible for inclusion if they: (1) were available in English; (2) contained relevant information on factors and associated interventions affecting media and digital health literacy in children; and (3) written in the last 24 years. Studies aligning with the framework from Jindarattanaporn et al. (2022) were emphasized, with additional research post-2020 incorporated for up-to-date perspectives [17].

Data interpretation was guided by thematic analysis, identifying common patterns across studies related to factors influencing media health literacy. Findings were synthesized to highlight key determinants, challenges, and intervention strategies, ensuring a well-rounded perspective on the topic.

FACTORS & INTERVENTIONS

Education of Children and Adolescents

The education of children and adolescents, and its impact on cognitive development depends on the quality, availability, and accessibility of teaching within the school system.18 Education plays a crucial role in media health literacy as the dissemination of health information, and thus achievement of health literacy, requires knowledge, comprehension, understanding, appraisal, and evaluation [18]. A study conducted with adolescent students in Lithuania suggests a positive correlation between individuals with more school achievements and high media health literacy.19 This identified relationship has been attributed to the development of cognitive abilities attained through higher education [18]. Conversely, lower education levels, low educational aspirations, and reading, spelling, or mathematics difficulties correlate to low media health literacy [19].

In order to improve the media health literacy of children and adolescents, interventions must be implemented within the school system. To this end, the Learning for Life (L4L) intervention aimed to promote media health literacy and healthy lifestyle behaviours by providing diverse health education resources to teachers [20]. Teachers can tailor the resources provided to suit different learners and implement resources into the standard classroom curriculum [20]. The intervention effectively enhanced students' media health literacy, particularly their ability to identify ways technology can support their health and identify good and poor health information [20]. The results suggest that interventions such as L4L can potentially improve children and adolescents' media health literacy and should be considered an essential component of school-based health education programs.20 Furthermore, another promising intervention for promoting media health literacy involves adopting the health promoting school education framework [21]. This framework is designed to achieve educational goals by addressing health issues [21]. This approach provides students with opportunities to develop skills in self-advocacy and empowerment in alignment with media health literacy [21]. Despite the promising evidence supporting the health-promoting school, more evidence is needed to establish it as the definitive standard for promoting media health literacy [21].

Education of Caregivers

The formal education level of caregivers has been found to impact their children's digital health literacy positively [14]. It has been suggested that a higher education level in caregivers provides a stronger parental foundation of digital health literacy than when caregivers had less education [14]. It has been posited that this academic foundation can then be relied upon while discussing media and related critical thinking skills with their children [14]. The demonstrated relationship between education and digital health literacy appears to hold across countries and cultures [14]. For example, Israeli children whose mothers were in school for 15 or more years had greater digital health literacy than children whose mothers had less than 12 years of education [14]. Similarly, a Swiss study found a positive association between a caregiver's understanding of the importance of a healthy lifestyle and their children's health literacy [22]. Lastly, a study from Taiwan showed that caregivers with a lower education level had lower levels of digital health literacy and tended to have children with lower levels of digital health literacy [23].

Given that caregivers' education level and digital health literacy impact their children's digital health literacy, interventions focused on teaching caregivers digital health literacy would seem to improve that of their children [23]. The implementation of digital health literacy courses into parenting programs has been proposed as one way to improve the digital health literacy of caregivers and children [24]. In a 2021 review of digital interventions on health literacy in caregivers with children with a health condition, 60% of the included studies reported satisfaction with their respective intervention [24]. All of the study interventions included an educational aspect, with 80% of the studies incorporating interactive communication components, a portal to connect caregivers with health care providers, videos, and online discussion channels [24]. Measured by a positive change in health behaviour or increase in knowledge, all of the studies in the review found improvement in caregivers' health literacy [24]. The review highlighted the lack of digital health interventions developed for caregivers [24].

Self-Efficacy

The Social Cognitive Theory (SCT) states that learning, functioning, and actions result from the ongoing and polydirectional interaction between personal, environmental, and behavioural factors [25]. A key component of SCT is self-efficacy, an individual's perception of and general trust in their abilities [25]. Self-efficacy is vital for successful health outcomes and explains how health behaviours are sustained when an individual believes in their ability to start, complete and maintain a desired behaviour [25]. Self-efficacy is critical for adolescents using the internet and social media as it impacts their ability to access, evaluate, and apply health information.25 According to one study conducted in Australia, 21 participants ages 12-17 completed an eHEALS self-report digital health literacy measure to assess their perceived digital health literacy [25]. Then, they participated in a practical search task using both a think-aloud protocol and an interview to assess their demonstrated digital health literacy.25 The results showed that overall, participants had high levels of perceived digital health literacy and reported using strategies to search and appraise online health information but did not understand or demonstrate these strategies in practice during the search task [25]. This study concluded that adolescents desire to improve their digital health literacy so they can confidently appraise online health information, but there is a discrepancy between their perceived and demonstrated digital health literacy skills [25].

In order to enhance adolescents' self-efficacy and digital health literacy, it is important to involve adolescents in the design and development of interventions [25]. Co-designing school-based and healthcare-based interventions can improve efficacy and uptake and demonstrate trust in and appreciation of adolescents' knowledge and creativity [25]. School-based interventions can promote the development and progression of adolescents' digital health literacy through the curriculum and by providing a supportive environment [25]. Healthcare professionals should assume that adolescents use online health information during appointments and discuss this with them [25]. Overall, interventions should focus on improving adolescents' self-efficacy as it is pivotal to developing and retaining healthy behaviours and a predictor of digital health literacy [25].

Socioeconomic Status

Household socioeconomic status (SES) strongly predicts health outcomes and behaviours in adolescents [25]. Media and digital health literacy is directly involved in the relationship between low SES and unhealthy lifestyle behaviours [26,27]. A systematic review identified five studies where high SES was significantly associated with higher scores on a digital health literacy scale among adults, carrying significant implications for adolescent development given the parent's role as a teacher [28,20]. In California, fifth graders from lower-SES households were more likely to consume sugar-sweetened beverages, believe incorrect health information, and underestimate the impact of advertisements, indicating poor media health literacy [29].

SES's strong association with media health literacy deficits makes it the perfect marker for targeting health literacy initiatives. High-quality education interventions are especially effective when administered to younger or more disadvantaged adolescents [30]. Investments in combatting SES inequality lead to long-term societal cost savings as adolescents grow to be more productive and healthier [30]. *The Bigger Picture* (TBP) is an arts-based public literacy program that educates high schoolers on health literacy in the context of systemic injustice and youth empowerment [28]. TBP successfully increased student media health literacy in a low SES high school despite being moved online due to the COVID-19 pandemic [31]. Interventions such as TBP help in the short-term by increasing youth awareness of health risk factors through personal connections and discussions [31]. The program also addresses SES in the long term by promoting civic engagement and social justice by prompting students to acknowledge their identities and their role and power in the systems surrounding them [31].

DISCUSSION

This opinion review explored four key factors associated with adolescent media health literacy [17]. The mechanisms behind these relationships are difficult to determine due to the observational nature of studies that explore environmental factors and the length of time between a potential environmental change and a measurable difference in media health literacy [18]. Nevertheless, there is value in studying these environmental risk factors to help identify the need or potential value of media health literacy programs. Lower levels of adolescent self-efficacy, SES, adolescent education level, and parental education level were all identified as risk factors for poor adolescent media health literacy. Future programs may benefit from prioritizing higher-risk communities, indicated by these factors, to prevent widening gaps in health equity.

The reviewed research included a variety of strategies for improving adolescent media or digital health literacy. Online information sessions effectively improved media health literacy when combined with arts-based mediums (poetry and poster-making), suggesting that virtual interventions may be effective for reducing potential cost barriers and that arts improve engagement [31]. Adolescent empowerment is another key feature of successful interventions since programs that involved adolescent empowerment through their incorporation in program design, an emphasis on self-advocacy, or civic health literacy [21,25,30]. Programs may also find success in targeting caregivers who impact their children's media health literacy through their environment and direct education. Beyond their at-home environment, successful interventions implemented health education resources into the standard classroom curriculum [23,24]. These strategies should be considered when designing future adolescent media health literacy programs.

With reliance on media for health information rapidly increasing and its suggested association with adolescent health outcomes, more research is required on the contributing factors to create targeted, evidence-based interventions to best promote the health of adolescents [2]. More specifically, future research is needed to ascertain which factors most indicate poor media health literacy and which interventions work best for adolescents.

As research moves forward in examining the media health literacy of adolescents, it is important to consider how to ethically involve this population in such research. It is critical to utilize primary data from adolescents as their health literacy is thought of as distinct from that of adults [18]. This stage, as well as any developmental stage, while interconnected, comes with unique milestones, with adolescents growing as independent decision-makers and forming health attitudes, beliefs, and behaviours related to health literacy [32-34]. Research on health interventions benefits from the involvement of the target population to inform the prioritization of intended health outcomes [35]. The Children's Advisory Group established by the London South Bank University Ethics Committee provides an example of how to prioritize children's perspectives in health literacy research [36]. Child-friendly research methods, revised consent forms, and more accessible methods of results dissemination are examples of changes that can be applied to health literacy research for more active integration of children's perspectives [36].

By understanding the potential risk factors associated with poor adolescent media and digital health literacy and identifying successful intervention strategies, future programs may best address the broadening gaps in health literacy and equity.

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2024 CHILD HEALTH CONFERENCE
WINNING ABSTRACT

Bot-ched Data: Dealing with Bots and Bad Actors in Online Autism Research

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BACKGROUND

There is a recent and dangerous threat to online research and data collection: bots and bad actors. Bots (artificial intelligence (AI)) and bad actors (human participants who do not truthfully complete surveys) are growing in their strength and numbers when it comes to infiltrating studies. In the field of autism research, these present additional barriers to an already underrepresented population. As these fraudulent participants continue to evolve past current methods which aim to combat them, it is imperative researchers consider all options of prevention, detection, and elimination, without compromising their survey's integrity or increasing participation barriers for valid participants.

OBJECTIVE

To synthesize current reviews of bot and bad actor prevention, detection, and elimination from online research surveys.

METHODS

A search of major databases was conducted for independent studies and reviews, resulting in 19 papers found to be most applicable. The literature was then summarized by what methods of bot and bad actor prevention, detection, and elimination were explored, how authors used each method, and the effectiveness of these.

RESULTS

The search resulted in 19 articles, 12 of which were independent studies which explained authors firsthand experiences dealing with bots and bad actors [1-12]. The remaining 7 were reviews which assessed common strategies for bot/bad actor prevention, detection, and elimination [13-19]. Of the independent studies, 2 focused on dealing with bad actors, 1 focused on bots, and 9 focused on both. For the reviews, none focused solely on bad actors, 1 focused on bots, and 6 discussed dealing with both.

Across these articles, 64 distinct methods were identified as strategies, however 11 of these were discussed most frequently.

(1) CAPTCHA

CAPTCHA stands for "Completely Automated Public Turing test to tell Computers and Humans Apart". This tool can detect bots in research surveys by employing various methods such as image recognition tasks. While effective in combination with other measures against bots, it may not be foolproof, as bots are evolving to bypass CAPTCHAs [1,3,6-10,13-16].

(2) Email Screening
Email screening involves the verification of email addresses to authenticate research participants. While successful in identifying some fraudulent activities, it is susceptible to bots and bad actors using valid email addresses. Its efficacy is contingent upon the overall implementation of complementary measures [1,3,5, 11,15,16].

(3) IP Address Tracking
An IP address is a unique number which identifies any device accessing the internet. Tracking IP addresses can be useful to identify participants who attempt to complete surveys multiple times or used to block specific addresses. This method demonstrates moderate effectiveness by identifying repeat entries and geographic inconsistencies but faces limitations with VPNs (virtual private network; masks a user's IP address) and dynamic IP addressing (a temporary IP address which continually changes over time) [3-6,8,10-16,18].

(4) Consistency Checks
Consistency checks involve analyzing response coherence, for example, asking "how old are you" at the start of the survey and later "what year were you born". An inconsistency in responses to these can point towards fraudulent participation. While showing potential for detecting AI and bad actor responses, effectiveness is not consistently defined across articles [2,3,8,10,11,13,15,16].

(5) Timestamp Analysis
Examining survey completion times and response durations proves useful in detecting suspicious activity such as extremely rapid response times. Although, this may not account for atypical response times in human participants across diverse and inclusive study populations [1-3,6,11,13-15,17].

(6) Attention Checks
Attention checks request specific actions, such as leaving a question unanswered or selecting a particular answer option. These are shown to be moderately effective, though they may be incorrectly answered by valid respondents, emphasizing the need for integration with other strategies [3,7,8,10,12-15].

(7) Two-Step Opt-In
Two-step opt-in processes require multiple steps from participants before they may enter the study. For example, pre-screening potential participants followed by unique survey links sent to those deemed valid. This aims to limit public access compared to surveys posted on more public platforms. Effectiveness does vary based on other strategies used and potential link exposure by scammers [1,4,6,7,9,11,14,15].

(8) Collection of Contact Information

The collection of personal contact information including phone numbers and physical addresses is varied in its effectiveness, relying heavily on proper integration with other validation techniques to ensure participant authenticity [4,9,14,16,17].

(9) Clarifying Compensation Rules

Compensation is often an incentive for bot and bad actor infiltration. Clarifying compensation rules can prevent fraud by explaining compensation policies and only offering compensation post-data verification. Its direct impact on reducing bot responses is not explicitly quantified, though public compensation advertisements are noted to increase fraudulent participation [5,7,9,14-16].

(10) Domain Knowledge Assessment

Domain knowledge assessment is the evaluation of participants' knowledge related to survey content. For example, asking participants in an autism study to explain their lived experience. This method yields varied results as part of an anti-fraud strategy and it may pose challenges for certain populations [3,10,14].

(11) Interviews Pre- and Post-Data Collection

Interviews conducted pre- and post-data collection (i.e. prospectively or retrospectively), exhibit high efficacy in preventing bot participants and eliminating bot respondents. This strategy is also effective in deterring and detecting bad actors. However, interviews may deter eligible individuals and can remove anonymity [2,5,12,16].

CONCLUSIONS

Current strategies employed to tackle bots and bad actors in online autism research is a complex and nuanced landscape. A synthesis of 19 relevant studies revealed several distinct approaches. However, none of these methods are completely effective in isolation. This emphasizes the necessity to combine multiple strategies to enhance their overall efficacy.

Another recurring concern surfaces throughout the discussion: the imminent obsolescence of current strategies in the face of rapidly evolving AI capabilities. While these methods can have effective outcomes, the relentless progress of AI technologies poses a formidable challenge to their sustainability. Thus, it becomes evident that a dynamic and adaptable approach is needed. Researchers across disciplines must collaborate to find novel method combinations and novel strategies.

As the use of online questionnaires in all research—especially studies on autism and other underrepresented populations—continues to grow, it is increasingly important to keep participation barriers low while collecting valid data.

QR Code to Full Research Poster:

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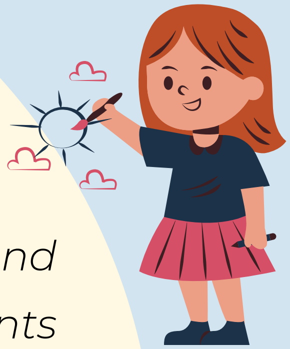

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