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# Complex Comorbid Presentations are Associated with Harmful Behaviour Problems among Children and Adolescents with Cerebral Palsy

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# Complex Comorbid Presentations are Associated with Harmful Behaviour Problems among Children and Adolescents with Cerebral Palsy

**Aim**: Frequency and relationship between gastrointestinal symptoms, sleep problems, internalizing and externalizing symptoms, behaviour problems and autism spectrum disorder (ASD) symptoms, and predictors of behaviour problems were examined in children and adolescents with Cerebral Palsy (CP).

**Method**: Parents of 104 children and adolescents with CP completed the Gastrointestinal Symptom Inventory, Children's Sleep Habits Questionnaire, Child Behaviour Checklist, Social Communication Questionnaire and the Behaviour Problem Inventory-Short Form.

**Results**: High frequency of behaviour problems (88.5%), gastrointestinal symptoms (81.7%), sleep problems (81%) ASD symptoms (48%) and internalizing and externalizing symptoms (31.7%) were found. Relationships were found between gastrointestinal symptoms and sleep problems, and gastrointestinal symptoms and internalizing and externalizing symptoms. Relationships were found between sleep problems and behaviour problems. Intellectual disability, sleep problems, internalizing and externalizing symptoms, and ASD symptoms predicted behaviour problems.

**Conclusion**: Findings highlights the frequency of comorbidities that exist in CP and how these comorbidities affect one another.

Keywords: Cerebral palsy, gastrointestinal symptoms, behaviour problems, sleep problems, internalizing symptoms, externalizing symptoms, autism spectrum disorder

## 1. Introduction

Cerebral palsy (CP) is a neurodevelopmental condition occurring in infancy and persisting throughout the life cycle of that individual.[1] CP is a non-progressive disorder of the brain and is commonly defined as "a group of permanent disorders of the development of movement and posture that occurred in the developing fetal or infant brain". [1(p9)] Motor disorders of CP often inflict disturbances of sensation, perception, cognition, communication, and behaviour by epilepsy, and by secondary musculoskeletal problems.[1] The neurological disorder results from an injury that prevents the full development of the central nervous system or is caused by abnormal brain development or damage which occurs in the womb, during childbirth or within the first 3 years of life. [2,3]

CP is classified as one of the most severe disabilities in childhood.[4] The estimated prevalence rates of CP ranges between 2 to 5 per 1000 live births.[5] A CP diagnosis will depend on the area and extent of the brain damaged, which are classified into four main types: spastic, athetoid, ataxic, and mixed CP.[3] This diagnosis is typically made during the toddler years, however professionals encounter difficulties in providing an accurate prognosis as CP is a collective term for many different brain lesions.[6] More so, treatment of CP for children and adolescents will depend on their specific symptoms which can range from physical therapy to medication and surgery.[7] In addition to this, the presence of additional symptoms are likely to develop as the child matures, thus complicating matters as they may be overlooked.[8]

### *1.1 Cerebral Palsy and Comorbidity*

Comorbidity refers to the presence of two or more co-occurring disorders in the same person.[9] Children and adolescents with CP will experience structural abnormalities in the

central and peripheral nervous system.[2] Therefore, individuals will be more likely to develop problems in areas regulated by the nervous system such as, gastrointestinal symptoms, sleep problems and a range of other disorders that will have an impact on their CP diagnosis.[2] In addition to this, research has found that intellectual disabilities (ID), epilepsy, sensory issues, communication problems, psychiatric disorders, and autism spectrum disorder (ASD) are found to commonly co-occur with a diagnosis of CP.[10-13]

The high prevalence of ASD symptoms found in children with CP has gained increased attention in recent literature.[11,12] Comorbidities that co-occur with ASD include sleep problems, behaviour problems, gastrointestinal symptoms, attention-deficit/hyperactivity disorder, anxiety, toileting problems, feeding problems, and epilepsy.[14-23] Due to the overlap between ASD and CP, it can be hypothesized that comorbid conditions common in ASD may also be common in CP. However, limited research exists on collectively examining comorbidities in CP. Although the brain injury in CP is non-progressive, the co-occurring disorders can change over time, thus reducing function and quality of life. Further research is required as these comorbid disorders have a considerable impact on those with CP and their overall functioning.

### *1.2 Cerebral Palsy and Autism Spectrum Disorder*

The co-occurrence of CP and ASD is an area of research that has gained momentum in recent studies. ASD is a common neurodevelopmental disorder affecting 1 in 54 children.[24] The co-occurrence of CP and ASD in children shows a higher prevalence rate than in the general population.[10,11] The frequency of co-occurring CP and ASD was 6.9% and was reported higher among children with non-spastic CP at 18.4%.[11] It has been found that

8.7% (107 out of 1,255) of children with CP had a diagnosis of ASD. [25] It was also found that 8.2% of children with CP had co-occurring ASD and a total of 35% had epilepsy. [26]

Researchers have found that ASD can be diagnosed as early as 18-24 months and interventions should commence once signs develop.[27] However, both CP and ASD share common overlaps in symptoms such as communication problems, repetitive behaviours and sensory issues.[1,28,29] It is of major importance to identify the impacts comorbidities have on CP patients to promote their well-being.[30] Quality of life may significantly be improved among children with CP by identifying and treating comorbid conditions in a systematic way.

### *1.3 Cerebral Palsy and Gastrointestinal symptoms*

Gastrointestinal (GI) symptoms such as chewing and swallowing difficulties, vomiting, gastroesophageal reflux, and constipation have adverse effects on the quality of life and life expectancy of children with CP.[31] One study found that 92% of children with CP have clinically significant GI symptoms, with chronic constipation being the most prevalent at 74%.[32] It was found that 80-90% of a population of children with CP and neurodevelopmental disorders experienced chronic GI symptoms and were at risk of developing malnutrition due to uncoordinated swallowing, aesophageal reflux, and constipation. [33] The consequences of these GI symptoms are varied and can greatly impair the general wellbeing of these children while persisting into adulthood. Such factors include nutritional deficits, respiratory infections, and recurrent hospitalizations, therefore, an early detection of these GI symptoms may facilitate more efficient treatment or rehabilitation methods. [34]

### *1.4 GI symptoms and ASD*

Previous research has found that GI symptoms are highly prevalent in ASD.[35] Previous research found that 79.3% of children and adolescents with ASD presented with GI symptoms. [36] This research found that sleep problems in this sample predicted the presence of GI symptoms, and GI symptoms predicted comorbid psychopathology. Similarly, research has suggested that GI symptoms may be a potential cause of sleep problems in children with ASD. [37] Previous research has identified GI symptoms as potential predictors of behaviour problems in ASD. [38] Research has identified that children with ASD and GI and/or sleep problems exhibited more serious forms of behaviour problems.[39] The co-occurrence of CP and GI symptoms is a relatively new area of research that warrants further investigation. As GI and ASD commonly co-occur, this emphasises the importance of researching GI symptoms in children with CP.[40, 41, 42]

### *1.5 Cerebral Palsy and Sleep Problems*

Sleep problems are prevalent in children with CP, particularly those with visual impairment.[43] Sleep disturbances may result from motor impairment, pain from gastro-oesophageal reflux, muscle spasms, and epilepsy.[43,44] One study reported that children with CP revealed a significantly higher frequency of sleep disturbances and breathing difficulties in comparison to children without disabilities.[45] Furthermore, previous research investigated sleep disorders in children with CP and found that 23% had an abnormal total sleep score, with the main disturbance being epilepsy.[44] Additionally, they found that 44% of children had at least one clinically significant sleep problem[44].

The relationship between sleep problems and behaviour problems has been reported as a significant issue in the CP population. Research has found that approximately 50% of children with CP with an abnormal total score on the CBCL, measuring behaviour problems, had an abnormal total score on the Sleep Disturbance Scale for Children.[46] The relationship between these comorbidities are particularly relevant as there is increasing evidence of the high prevalence of behaviour problems in CP[6,47,48]. It is evident that there are a number of potential factors attributed to sleep problems in children with CP, however, there is a lack of research conducted on this topic. Future research on CP and comorbid sleep problems is required to increase understanding and facilitate treatment.

### *1.6 Cerebral Palsy and Internalizing and Externalizing Symptoms*

Comorbid psychopathology refers to the occurrence of two or more forms of psychopathology in the same person.[9] Such disorders may range from mood disorders, anxiety disorders, Attention-Deficit/Hyperactivity Disorder (AD/HD) and other psychological disorders.[49] Higher levels of comorbid psychopathology are presented in children with CP than their typically developing peers.[50] Research has found that children with CP tend to present more emotional problems, hyperactivity, anxiety, and depressive symptoms than those without CP.[51] Previous research assessed mental health problems in children with CP and found that one in two children met the criteria for a psychiatric disorder whereby AD/HD was most prevalent.[52] Similarly, in a later study, it was identified that eight out of nine children with CP that presented with ASD symptoms met the criteria for AD/HD. [12] Findings of this research revealed that there was a significant

overlap between the presence of ASD symptoms and children meeting the criteria for AD/HD that have a diagnosis of CP.

Internalizing and externalizing symptoms such as anxiety have frequently been linked to the prevalence of behaviour problems in ASD.[50,53-57] Behaviour problems may in some circumstances, represent the atypical presentation of an underlying psychiatric disorder in individuals with developmental disabilities such as ASD. [58] The presence of ASD symptoms in CP is an important area for research as it may help address internalizing and externalizing symptoms common to ASD, that may also be common in CP. Overall, research concerning CP and internalizing and externalizing symptoms is rather limited and therefore, the current study will explore internalizing and externalizing symptoms and its relationship with CP to facilitate research in this area.

### *1.7 Cerebral Palsy and Behaviour Problems*

Behaviour problems have been defined as "culturally abnormal behavior(s) of such intensity, frequency or duration that the physical safety of the person or others is likely to be placed in serious jeopardy, or behaviour which is likely to seriously limit use of, or result in the person being denied access to, ordinary community facilities". [58 (p.44)] Based on this knowledge, individuals exhibiting behaviour problems not only put themselves at risk for injury and danger, but they also put those around them in danger. It has been reported that behaviour problems exhibited by children with developmental disabilities can affect family functioning and overall well-being of the child and their families. [47,59] It has also been reported that the presence of behaviour problems is associated with higher parental stress in various populations of children with developmental disabilities.[60-62] In addition,

previous research on ASD and ID populations has identified that if behaviour problems go untreated, they are more likely to persist into adulthood and continue to increase in severity as the child grows older.[63] Therefore, research investigating the predictors of behaviour problems in these populations is imperative.

In the context of CP, behaviour problems are a common co-occurring issue in this population.[47] A recent meta-analysis showed that 1 in 4 children with CP have a behavioural disorder compared with 1 in 10 typically developing children. [6,64] Attention difficulties, withdrawn, aggressive behaviour, and anxious/depressed symptoms are some of the most prevalent behaviours in CP.[49] It has been found that 48% of children with CP had high scores of behaviour problems, whereby aggression was reported as the most common behaviour problem. [49] In the ASD population, co-occurring disorders such as GI symptoms, sleep problems, and psychopathology have been reported to exacerbate the presence of behaviour problems. [38, 65-71]

Since ASD has been reported as a common comorbidity of CP, it could be hypothesised that the presence of such co-occurring conditions in CP, may influence the frequency and severity of behaviour problems in CP. Researching behaviour problems as a comorbidity in CP is important as it is currently under researched which requires intervention and attention. Research in this area of behaviour problems can help practitioners to not only treat the behaviours but to also prevent them. Identifying predictors of behaviour problems among children can prevent the worsening of these problems, as well as optimizing the wellbeing and safety of children with CP and their families.

### *1.8 Current Study*

Research investigating behaviour problems in CP is needed as currently it is not appreciated clinically as an important comorbidity within this population. Other populations such as ASD have identified significant relationships between other comorbid conditions and the frequency and severity of behaviour problems. Based on this knowledge, the current study aimed to investigate the frequency of GI symptoms, sleep problems, internalizing and externalizing symptoms, and ASD symptoms as comorbid conditions in a sample of children and adolescents diagnosed with CP. A secondary aim of the current study was to examine the impact these comorbid conditions have on the frequency and severity of behaviour problems in CP.

## 2. Method

### *2.1 Participants*

The sample consisted of 104 children and adolescents with a diagnosis of CP. The sample comprised of 60.6% males (*n*=63) and 39.4% females (*n*=41). The mean age of the sample was 6.88 years (*SD* = 4.11), with a range from 2.5 to 18.5 years. The mean age of a CP diagnosis was 18.21 months (*SD* = 16.10).

Among the sample, spastic CP was reported the highest at 61.5% (*n*=64), followed by mixed CP at 26% (*n*=27), other forms of CP diagnoses at 7.7% (*n*=8), and 4.8% (*n*=5) reported ataxic CP. It was reported that most participants (55.8%) received their diagnosis from a neurologist (*n*=58), while 23.1% (*n*=24) from a paediatrician, 7.7% (*n*=8) from a neonatologist, and 13.5% (*n*=14) were from others, such as psychiatrists, special health care

needs consultants, geneticists, paediatric developmental behavior specialists, and orthopaedic surgeons. A total of 58.7% (*n*= 61) of participants had an intellectual disability (ID). It was found that 26.9% (*n*=28) reported having a mild level ID followed by 17.3% (*n* = 18) with a moderate ID, and 24.5% (*n*=15) reported having a severe level of ID.
A total of 60.6% (*n*=63) of participants were from the United States of America, while 17.3% (*n*=18) were from Ireland, 8.7% (*n*=9) were from the United Kingdom, 4.8% (*n*=5) from Canada, 1.9% (*n*=2) were from Australia, and 6.7% (*n*=7) were from other countries.

### *2.2. Measures*

#### *2.2.1 Demographic Information*

Parents or caregivers answered a self-constructed questionnaire providing information on their child's age, gender, and country of residence. Information was also provided on the age of CP diagnosis and classification, whether they have additional diagnoses such as ASD, ID or any other comorbid disorder, if they are currently receiving special educational interventions and the amount of hours a week, if the child is on any other sort of medical treatment or taking melatonin at present.

#### *2.2.2 Child's Sleep Habits Questionnaire (CHSQ).*

The CSHQ consists of a parent-report 52 item sleep-screening tool designed to screen for children's sleep habits.[72] The CSHQ is made up of eight sub-scales which include bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night waking, parasomnias, day-time sleepiness, and sleep disorder breathing. Forty-two of the items are rated on a three-point Likert scale, with the responses raging from 'Rarely', 'Sometimes', and 'Usually' in

relation to their child's sleep from the previous week. A total CSHQ score of 41 has been reported to be the clinical cut-off for identification of sleep problems. Although the questionnaire is designed for children aged 4 to 10 years of age, the CSHQ has been used in previous studies with younger and older children with a diagnosis of ASD.[66,73]

*2.2.3 Gastrointestinal Symptom Inventory (GSI).*

The GSI is a questionnaire developed by the Autism Treatment Network and is designed to identify GI symptoms in the past three months.[74] The GSI consists of 35 items, however there are additional items depending on a participant's symptomatology, overall including 77 items. It is scored dichotomously, meaning if the symptom is present or not. It also allows assessment of specific symptomatology including abdominal pain, abnormal bowel movements, reflux, constipation, nausea, and any other reported abdominal or stomach aches to allow a clearer identification for each problem. Although it has not yet been validated, it has been used in published research studies with children and adolescents with ASD.[75,76]

*2.2.4 Behaviour Problems Inventory-Short Form (BPI-S)*

The BPI-S is a shortened version of the BPI-01, an inventory used to measure the frequency and severity of behaviour problems.[77] The BPI-S consists of 30 items and is made up of three behaviour subscales: Self-Injurious Behavior (SIB; 8 items), Aggressive/Destructive Behavior (10 items) and Stereotyped Behavior (12 items). Each item is rated on a 5-point frequency scale (0 = never to 4 = hourly), and a 4-point severity scale (0 = no problem to 3 = severe problem).

*2.2.5 Child Behavior Checklist (CBCL)*

The CBCL is a 113-item, parent-report form questionnaire used to assess psychopathologies and behavioural problems in children and adolescents aged 1½ to 18 years, based on the past 6 months.[78] The CBCL is divided into two separate age groups, from 1½ to 5 years of age (categorized into seven behaviour subscales) and from 6 to 18 years of age (categorized into eight behaviour subscales). The subscales are grouped into internalising problems, externalising problems and other problems based on the following: anxiety and depression, social withdrawn, somatic complaints, social problems, thought problems, attention problems, rule-breaking behaviour, and aggressive behaviour. Parents are instructed to answer on a 3-point scale: '0' (Not true), '1' (Somewhat or Sometimes True), and '2' (Very or Often True).

*2.2.6 Social Communication Questionnaire (SCQ)*

The SCQ is a 40-item parent-report questionnaire, which screens for symptoms of ASD.[79] The questionnaire evaluates children's communication skills, social interaction, and stereotyped, repetitive behaviour. Each item in this scale requires a 'Yes' or 'No' response and each scored item receives 1 point for abnormal behaviour and 0 points for absence of abnormal behaviour. Total scores can range from 0-39 and a total score of 15 or above is suggestive of the presence of ASD symptoms. It has been widely used and validated to use as a first screening tool for ASD in clinical samples.

***2.3 Informants***

Informants were parents or caregivers of children and adolescents with Cerebral Palsy. Parents were given information and instructions on each of the questionnaires and these were completed independently.

### *2.4 Procedure*

Ethical approval was granted by the School Research Ethics Committee in the University. Parents and caregivers were recruited through social media, CP worldwide organisations, online forums and parent support groups for CP. If parents wished to participate in the study, they were provided with a participant information form and a consent form to complete. Parents provided consent for their children as they were completing questionnaires on their behalf. Assent for minors was not obtained. Once consent was obtained, the informants were provided with the questionnaires and they could complete them in their own time. Rating scales were completed by parents independently according to the instructions which were printed on the top of each questionnaire.

## 3. Results

### *3.1. Analyses*

A series of Pearson's correlations were carried out to examine the relationship between GI symptoms (total score of GSI), sleep problems (total score of CSHQ), internalizing and externalizing symptoms (total score of CBCL) and ASD symptoms (total score of SCQ) on behaviour problems based on the BPI-S subscales. In addition, five hierarchical multiple regressions were conducted to investigate whether GI symptoms, sleep problems,

internalizing and externalizing symptoms, and ASD symptoms predict behaviour problems in children and adolescents CP based on the BPI-S subscales.

### *3.2. Descriptive Statistics*

Parent report of comorbid diagnoses were obtained from the sample. Intellectual disability was the highest co-occurring diagnosis reported in 58.7% (*n* = 61), epilepsy was reported in 38.5% (*n* = 40), AD/HD was found in 15.4% (*n* = 16), ASD was reported in 12.5% of the participants (*n* = 13), and Pervasive Developmental Disorder in 10.6% (*n* = 11). A combination of Occupational Therapy and Speech and Language Therapy was reported in 64.4% (*n* = 67) of the sample, followed by Physiotherapy in 39.4% (*n* = 41) and Applied Behavior Analysis in 13.5% (*n* = 14). Hours of intervention received per week was then measured. Two hours of therapeutic intervention was received in 17.3% (*n* = 18), 15.4% (*n* = 16) received 5 hours per week, followed by 11.5% (*n* = 12) receiving at least 3 hours, and 7.7% (*n* = 8) receiving 20 hours of interventions. The mean time of intervention in the whole group was 7.93 (*SD* = 7.86). Furthermore, 58.7% (*n* = 61) of the sample reported that they were currently on medication, and 19.2% (*n* = 20) reported they were taking melatonin at the time of the study.

### *3.3. Comorbid Conditions in Cerebral Palsy*

Table 1 presents the frequency of GI symptoms, sleep problems, internalizing and externalizing symptoms, behaviour problems, and ASD symptoms in participants with CP. Behaviour problems was the most prevalent comorbidity (88.5%), with the most common topography being stereotyped behavior (*n* = 77, 74%), followed by SIB (*n* = 73, 70.2%) and

aggressive/destructive behavior (*n* = 60, 57.7%). GI symptoms was the second most common comorbidity in the sample. Regarding specific GI symptoms, constipation was the most prevalent (*n* = 66, 63.5%), followed by abdominal pain (*n* = 55, 52.9%), diarrhoea (*n* = 39, 37.5%), nausea (*n* = 35, 33.7%), other GI symptoms (*n* = 32, 30.8%) and bloating (*n* = 27, 26%). Regarding internalizing and externalizing symptoms, 32.7% (*n* = 34) had cut-off scores for borderline and clinical ranges for internalizing problems, and 23.1 % (*n* = 24) had borderline and clinical scores for externalising problems.

---Insert Table 1 about here---

A summary of the means and standard deviations of BPI-S subscales, CSHQ subscales and total scores, CBCL subscales and total scores, SCQ total score and the GSI total score are presented in Table 2.

---Insert Table 2 about here---

### *3.4. Correlational Analysis*

#### *3.4.1. Behaviour Problems.*

A series of correlations were carried out between GI symptoms, sleep problems, internalizing and externalizing symptoms, and ASD symptoms on behaviour problems based on the five subscales of the BPI-S. Significant correlations were found between behaviour problems based on the five BPI-S subscales and internalizing and externalizing symptoms, sleep problems, and ASD symptoms. Table 3 presents a summary of these findings.

---Insert Table 3 about here---

*3.4.2. Internalizing and Externalizing Symptoms.*

Internalizing and externalizing symptoms based on the total scores of the CBCL had significant relationships across all variables within the study. A significant relationship was found between internalizing and externalizing symptoms and GI symptoms based on GSI total scores ($r$=.24, $p$=.015). Additionally, significant correlations were found between internalizing and externalizing symptoms and ASD symptoms based on the SCQ ($r$=.22, $p$=.024) and between sleep problems based on the CSHQ ($r$=.37, $p$=.001).

*3.4.3. GI symptoms and sleep problems.* Of the sample, 67.3% ($n$ = 70) had both comorbid GI symptoms and a comorbid sleep problem. A significant positive correlation was found between total GI symptoms and total sleep problems ($r$=.34, $p$=.001).

### *3.5. Hierarchical Multiple Regressions*

A series of five hierarchical multiple regressions were conducted to examine if age, gender, presence of ID, GI symptoms, sleep problems, ASD symptoms and internalizing and externalizing symptoms predicted behaviour problems in children and adolescents with CP. In each model, age, gender, and presence of ID were entered in the first step. The CSHQ total score was added in the second step followed by the addition of the total score on the GSI in the third step of the model. The presence of ASD symptoms was entered in the fourth step of the model and lastly, the total score on the CBCL was entered in the fifth step. The same steps were carried out for each of the five behaviour problems subscales:

SIB frequency, SIB severity, aggressive/destructive behaviour frequency, aggressive/destructive behaviour severity, and frequency of stereotyped behaviour.

### *3.5.1 Predictors of SIB frequency.*

The first block with age, gender and presence of an ID significantly predicted SIB frequency $F_{(3,100)} = 2.93$, $p < .05$, with an adjusted $R^2$ of .053. The presence of an ID was the only significant contributor to the variance explained. Subsequently, the addition of GI symptoms in the second block did not contribute significantly. The third block with the addition of the total sleep problems scores significantly contributed to the model $F_{(5, 98)} = 3.42$, $p < .05$, with an adjusted $R^2$ of .105. Block 4, with the presence of ASD symptoms did not contribute to model significantly. Lastly, the fifth block, with the addition of the CBCL total scores was significant, $F_{(7, 96)} = 3.67$, $p < .001$, with an adjusted $R^2$ of .154.

### *3.5.2 Predictors of SIB severity.*

The first block significantly predicted SIB severity $F_{(3,100)} = 3.71$, $p < .05$, with an adjusted $R^2$ of .073. The presence of an ID was the only significant contributor to the variance explained. The addition of total GI symptoms and ASD symptoms in block two and three did not contribute significantly to the model. The fourth block, with the total sleep problems scores significantly contributed to the model $F_{(5,98)} = 5.37$, $p < .001$, with an adjusted $R^2$ of .167. The fifth block with the addition of total CBCL scores was significant $F_{(7, 96)} = 5.32$, $p < .001$ with an adjusted $R^2$ of .227.

*3.5.3 Predictors of aggressive/destructive behavior frequency.*

The first and second block did not contribute to the model significantly. The addition of total GI symptoms scores in the third block did not contribute significantly. In block four, sleep problems and ASD symptom scores significantly contributes to the model $F_{(6,97)} = 2.46$, $p < .05$, with an adjusted $R^2$ of .078. The addition of the total CBCL scores in the fifth block significantly predicted aggressive/destructive behavior frequency, $F_{(7, 96)} = 7.31$, $p < .001$ with an adjusted $R^2$ of .30.

*3.5.4 Predictors of aggressive/destructive behavior severity.*

The first and second block within this model did not contribute significantly as predictors of aggressive/destructive behavior severity. In block three, the addition of total sleep problem scores significantly contributed to this model $F_{(5, 98)} = 3.18$, $p < .05$ with an adjusted $R^2$ of .096. In block four, with the addition of total SCQ scores, the model remained significant $F_{(6, 97)} = 3.70$, $p < .05$ with an adjusted $R^2$ of .136. The addition of the total CBCL scores in the fifth block significantly contributed to this model, $F_{(7, 96)} = 9.97$, $p < .001$ with an adjusted $R^2$ of .379.

*3.5.5 Predictors of stereotyped behavior frequency.*

Block one and block two did not contribute significantly to this model. The addition of the total sleep problem scores in block three was found to contribute significantly to this

model $F_{(5, 98)} = 2.76$, $p < .05$ with an adjusted $R^2$ of .079. Block four remained statistically significant however total scores of SCQ did not contribute to the model significantly. The addition of the total CBCL scores in the fifth block significantly predicted stereotyped behavior frequency, $F_{(7, 96)} = 6.02$, $p < .001$ with an adjusted $R^2$ of .254.

## 4. Discussion

The current study examined children and adolescents with a diagnosis of CP and the associated comorbidities that can develop throughout their life cycle. The primary aim of the current study was to examine the prevalence and relationships between GI symptoms, sleep problems, internalizing and externalizing symptoms, behaviour problems, and ASD symptoms as predictor variables in children and adolescents with CP. Secondly, the current study investigated the relationship between these predictor variables and behaviour problems. This current study is the first to explore the relationship of these comorbidities in children and adolescents with CP simultaneously.

Findings from the current study revealed high rates of comorbid disorders in CP. Behaviour problems was the most prevalent comorbidity in this sample with 88.5% exhibiting at least one topography, which is much higher than previous estimates of 25.5%.[48] In the current study, stereotyped behavior (74%) was the most common topography, followed by SIB (70.2%) and aggressive/destructive behavior (57.7%). Further analysis revealed that the presence of ID, sleep problems, internalizing and externalizing symptoms, and ASD symptoms predicted behaviour problems. Sleep problems was one of the most influential factors in predicting behaviour problems as it predicted both the frequency and severity of stereotyped behavior, SIB and

aggressive/destructive behavior in the current study. This novel finding may indicate that sleep problems exacerbate behaviour problems in CP. From this finding, it may by hypothesized that sleep problems play a role in exacerbating other comorbidities. Research literature suggests that sleep disturbances in children with CP increases the risk of cognitive impairments, emotional problems and health problems.[80] It is important to consider that other comorbidities may be provoked due to the nature of sleep disturbances in children and adolescents with CP.

Sleep disturbances are common following traumatic brain injury and CP is renowned for damage to the brain before or after birth.[81] Sleep regulating brain regions may be affected given the structural injuries to the brain in CP, thus having an impact on sleep quality in CP. The relationship between head injury and sleep disturbance has not been extensively researched and requires more attention in future research on sleep problems in CP.

As shown in previous studies, children with a diagnosis of CP tend to report a higher incidence of sleep problems than their typically developing peers.[46] Results from the current study found that 81% of children and adolescents with CP presented with a sleep problem, with daytime sleepiness, parasomnias and bedtime resistance being the most common problems. Previous research identified epilepsy comorbid with CP as a main cause for sleep disorders in children with CP.[44] A total of 38.5% percent of participants in this sample reported a diagnosis of epilepsy which may have contributed to the high levels of sleep problems in the current study. A reported 19.2% of the sample reported taking melatonin, which confirms that sleep problems are a common problem for this population, with some seeking medical aids to assist at night. Results from the current study

highlighted the importance of considering sleep problems when addressing the needs of children and adolescents with CP and their associated comorbidities.

The current study reported one of the highest incidences of GI symptoms (81.9%) in a sample of children and adolescents with CP. Previous research has identified the high prevalence of GI symptoms in CP as a result of the neural implications of the condition affecting the correct functioning of the digestive tract. [2,32,33] The current study reported constipation as the most common symptom with 63.5% of the sample being affected. As suggested by previous research, children with CP lack mobility which indirectly contributes to high levels of constipation.[32] Results from the current study indicated that GI symptoms, specifically constipation, are common in children and adolescents with CP.

The relationship between comorbidities in CP was examined in the current study and a significant relationship was found between GI symptoms and sleep problems. This finding suggests that high levels of sleep problems were associated with high levels of GI symptoms. As previously mentioned, bowel problems, particularly constipation have been found to occur frequently for individuals with CP. Previous research acknowledged that sleep problems commonly co-occur in CP, while abdominal discomfort and bloating can disrupt quality of sleep. [80] Although there are many factors that can interfere with sleep in CP, pain is a factor which can arise from GI symptoms. It is a common factor which is known to be underreported and undertreated in this population, thus having a negative impact on sleep duration for those with CP.[80] This coincides with the current study's findings and therefore, it is an area that warrants further attention when researching sleep problems in CP.

Results from the current study found that 31.7% of participants scored between the borderline and clinical range of the CBCL. According to previous research, one in two children with CP met the criteria for a comorbid psychopathological disorder, with AD/HD being the most common.[52] In the current study, 15.4% of the sample had a diagnosis of AD/HD. Based on findings from the CBCL, 32.7% met the cutoff for borderline and clinical scores for internalizing problems which further confirms the prevalence of internalizing and externalizing symptoms in CP.

Similar to sleep problems, the presence of internalizing and externalizing symptoms in the sample predicted the frequency and severity of behaviour problems, specifically stereotyped, SIB and aggressive/destructive behavior. Previous research has identified high levels of behaviour problems in AD/HD.[82] Therefore, the prevalence of AD/HD in this sample may contribute to the higher levels of behaviour problems in the current study. Additionally, research has shown that children with comorbid ASD and AD/HD exhibit higher rates of behaviour problems.[82] The high levels of participants with ASD symptoms may also be an influential factor in this finding. The current study also found that internalizing and externalizing symptoms correlated with GI symptoms, sleep problems and ASD symptoms. There is an under-researched link between these comorbid disorders and future investigation is required in order to determine the nature of this relationship. This is a novel finding and these comorbidities should be considered during diagnosis and treatment of children and adolescents with CP.

A total of 48% of the sample presented with symptoms of ASD. As suggested by previous literature, this high prevalence may be indicative of the overlap of symptoms in ASD and CP.[1, 28, 29] Findings from the current study also found that ASD symptoms

predicted behaviour problems, specifically the frequency and severity of aggressive/destructive behavior. Aggression is one of the most common behaviour problems for individuals with CP and ASD.[48,83] Therefore, this highlights the overlap between CP and ASD. Although participants were not diagnosed with ASD, these findings highlight the need for screening of ASD when a child is diagnosed with CP. It is imperative that symptoms of ASD are detected early as early identification and intervention are the key elements in managing this disorder, which in turn may also help manage CP.

The main findings of the study are helpful in identifying an up-to-date prevalence rate of some of the most common factors affecting the day to day life of children and adolescents with CP. This shows how important it is for practitioners and clinicians that work with such individuals in an applied setting to consider the influence that these factors may be having on the individual and consider the treatment that best fits all their needs. Previous research has discussed that the treatment of CP should be targeted and tailored to the individual and should be introduced promptly, with careful consideration taken into the overall diagnosis.[2] However, considering findings of the present study, careful consideration should be taken into the initial diagnosis as well as the various comorbid conditions that the individual may possess to allow for the early intervention of treatment. Identifying comorbid conditions are important for many reasons. For example, many comorbid conditions are treatable, and if they are detected and managed it will ensure a greater sense of well-being and quality of life for the individual[37].

The current study had some limitations. The data obtained was by parental report, as none of the participants were approached or evaluated directly to respond for themselves. However, previous research has found that parental reports of GI symptoms

are highly correlated with the findings of the evaluations of clinicians.[84] In addition, there is a potential for sampling bias in the current study. It may have been the case that mainly parents of children and adolescents with CP presenting with GI symptoms, sleep problems, behaviour problems, internalizing and externalizing symptoms, and ASD symptoms decided to participate rather than parents of children and adolescents with CP without these issues. Future research should explore the relationship between these variables in a larger population sample to better understand how they affect and predict each other by establishing causal relationships in more detail or exploring other associations between them.

In this study there was a large age range in the sample, with different typical developmental problems. For example, the severity of aggressive/destructive behaviour may be very different across the different age groups in the sample. Future research should incorporate a smaller age range sample in order to get a more accurate account of the severity of behaviour problems presented by the participants in that age group. A number of analyses were conducted in this study. Lastly, the measures used in this study were quite broad. Future research should include multi-method assessments of each comorbidity so that interventions can be more specific and tailored towards children and adolescents with CP.

In conclusion, this is the first study of its kind to investigate GI symptoms, sleep problems, behaviour problems, internalizing and externalizing symptoms, and ASD symptoms in children and adolescents with CP. The current study highlighted the prevalence of these comorbidities in CP as well as identifying the presence of ID, sleep

problems, internalizing and externalizing symptoms, and ASD symptoms as predictors of behaviour problems in CP.

## Compliance with Ethical Standards

### *Disclosure of Interest*

The authors declare no conflict of interest.

### *Ethical Approval*

All procedures performed in studies involving human participants were in accordance with the ethical standards of National University of Ireland Galway and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

### *Informed Consent*

Informed consent was obtained from all individual participants included in the study.

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Table 1.

*Percentage and frequencies of GI symptoms, sleep problems, ASD symptoms, and internalizing and externalizing symptoms in the total sample and across types of CP within the sample.*

| | Total (*N*=104) | | Spastic (*n*=64) | | Mixed (*n*=27) | | Ataxic (*n*=5) | | Other (*n*=8) | |
|---|---|---|---|---|---|---|---|---|---|---|
| | *n* | % | *n* | % | *n* | % | *n* | % | *n* | % |
| Presence of Gastrointestinal Symptoms [1] | 85 | 81.7 | 53 | 82.8 | 22 | 81.5 | 3 | 60 | 7 | 87.5 |
| Presence of Sleep Problems [2] | 84 | 80.8 | 49 | 76.6 | 25 | 92.6 | 2 | 40 | 8 | 100 |
| Presence of ASD symptoms [3] | 50 | 48.1 | 36 | 56.3 | 9 | 33.3 | 2 | 40 | 3 | 37.5 |
| Presence of Internalizing and Externalizing symptoms [4] | 33 | 31.7 | 17 | 26.6 | 13 | 48.2 | 1 | 20 | 2 | 25 |
| Internalizing Borderline Range | 5 | 4.8 | 3 | 4.7 | 2 | 7.4 | 0 | 0 | 0 | 0 |
| Internalizing Clinical Range | 29 | 27.9 | 15 | 23.4 | 12 | 44.4 | 1 | 20 | 1 | 12.5 |
| Externalizing Borderline Range | 8 | 7.7 | 3 | 4.7 | 5 | 18.5 | 0 | 0 | 0 | 0 |
| Externalizing Clinical Range | 16 | 15.4 | 10 | 15.6 | 3 | 11.1 | 1 | 20 | 2 | 25 |

1. Presence of at least one gastrointestinal symptom within the previous three months.
2. Scored above 41 on the Children's Sleep Habits Questionnaire (CSHQ).
3. Scored between the borderline and clinical range on the Child Behavior Checklist (CBCL).
4. Scored above 15 on the Social Communication Questionnaire (SCQ).

Table 2.

*Means and standard deviations for BPI-S, CSHQ, CBCL, GSI and SCQ based on the whole sample and across types of cerebral palsy.*

| | | Total (*N*=104) | | Spastic (*n*=64) | | Mixed (*n*=27) | | Ataxic (*n*=5) | | Other (*n*=8) | |
|---|---|---|---|---|---|---|---|---|---|---|---|
| | | *M* | *SD* | *M* | *SD* | *M* | *SD* | *M* | *SD* | *M* | *SD* |
| **BPI-S** | SIB frequency | 3.46 | 4.66 | 3.28 | 4.84 | 3.56 | 5.04 | 4.80 | 3.96 | 3.75 | 1.83 |
| | SIB severity | 2.39 | 2.84 | 2.33 | 3.13 | 2.30 | 2.42 | 3.80 | 3.03 | 2.25 | 1.49 |
| | Aggressive/destructive behavior frequency | 4.62 | 6.46 | 4.66 | 6.85 | 4.37 | 5.71 | 3.60 | 3.91 | 5.75 | 7.70 |
| | Aggressive/destructive behavior severity | 3.35 | 4.54 | 3.34 | 5.06 | 3.22 | 3.24 | 3.20 | 3.27 | 3.88 | 5.19 |
| | Stereotyped behavior frequency | 8.30 | 9.48 | 6.80 | 8.83 | 10.74 | 9.44 | 11.60 | 5.55 | 10.00 | 14.79 |
| **CSHQ** | Bedtime resistance | 10.20 | 3.61 | 10.02 | 3.80 | 10.93 | 3.04 | 9.00 | 3.54 | 10.00 | 4.11 |
| | Sleep onset delay | 1.87 | .80 | 1.78 | .83 | 1.89 | .75 | 1.80 | 084 | 2.50 | .53 |
| | Sleep duration | 5.41 | 1.95 | 5.33 | 1.93 | 5.15 | 1.75 | 4.80 | 2.68 | 7.38 | 1.41 |
| | Sleep anxiety | 3.16 | 1.26 | 3.13 | 1.29 | 3.15 | 1.06 | 3.20 | 1.78 | 3.50 | 1.51 |
| | Night wakings | 5.12 | 1.79 | 5.23 | 1.86 | 4.63 | 1.69 | 5.00 | 1.58 | 5.88 | 1.55 |
| | Parasomnias | 10.89 | 2.80 | 10.84 | 3.12 | 10.82 | 1.90 | 10.60 | 2.51 | 11.63 | 3.20 |
| | Sleep disordered breathing | 4.45 | 1.82 | 4.42 | 1.85 | 4.48 | 1.74 | 4.40 | 2.19 | 4.63 | 2.00 |
| | Daytime sleepiness | 11.19 | 3.39 | 11.36 | 3.46 | 11.26 | 3.12 | 8.40 | 3.21 | 11.38 | 3.62 |

| | | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|
| | Total CSHQ score | 52.29 | 11.48 | 52.12 | 12.55 | 52.30 | 8.45 | 57.20 | 11.39 | 56.88 | 11.85 |
| **CBCL** | Internalizing problems | 13.49 | 9.89 | 12.44 | 8.40 | 17.44 | 11.94 | 11.00 | 14.70 | 10.13 | 7.97 |
| | Externalizing problems | 11.41 | 10.53 | 10.63 | 10.60 | 13.37 | 10.21 | 11.00 | 11.05 | 11.38 | 12.04 |
| | Total CBCL score | 45.91 | 29.37 | 42.07 | 27.10 | 56.41 | 31.64 | 45.80 | 37.46 | 41.38 | 31.48 |
| **GSI** | Total GSI score | 2.44 | 1.64 | 2.44 | 1.59 | 2.67 | 1.73 | 1.00 | 1.23 | 2.63 | 1.77 |
| **SCQ** | Total SCQ score | 12.55 | 13.27 | 14.13 | 12.66 | 10.07 | 14.64 | 10.60 | 14.52 | 9.50 | 13.14 |

Note: BPI-S=Behavior Problems Inventory-Short Form; CSHQ=Children's Sleep Habits Questionnaire; CBCL=Child Behavior Checklist; GSI=Gastrointestinal Symptom Inventory; SCQ=Social Communication Questionnaire

Table 3.

*Pearson's correlation between BPI-S subscale scores and GSI total scores, CSHQ total scores, CBCL total scores, SCQ total scores.*

| | SIB Frequency | SIB Severity | Aggressive/ destructive behavior Frequency | Aggressive/ destructive behavior Severity | Stereotyped behavior Frequency |
|---|---|---|---|---|---|
| Total GSI | .11 | .16 | .03 | .04 | .16 |
| Total CSHQ | .30** | .37** | .27** | .34** | .29** |
| Total CBCL | .36** | .40** | .57** | .62** | .53** |
| Total SCQ | -.08 | -.08 | .19 | .21* | .10 |

* $p<.05$

** $p<.01$

Note: BPI-S=Behavior Problems Inventory-Short Form; GSI=Gastrointestinal Symptom Inventory; CSHQ=Children's Sleep Habits Questionnaire; CBCL=Child Behavior Checklist; SCQ=Social Communication Questionnaire