School-aged Children with Down Syndrome in Galle, Sri Lanka: Relationship between Level of Disability, Resource Use and Caregiver Burden

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ABSTRACT

Purpose: *The study assessed the relationship between the level of disability amongst school-aged children with Down Syndrome and overall caregiver burden, and the potential moderating effects of social support and external service access on the caregiver/child relationship in Sri Lanka.*

Method: *Caregivers were recruited to complete the Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD) assessment, the Caregiver Difficulties Scale (CDS), and to answer questions regarding resource use. A subset of 15 surveyed caregivers were also invited to participate in semi-structured interviews. Regression analyses were used to investigate the impact of resource usage on the association between level of disability and caregiver burden.*

Results: *Lower perceived levels of child disability were related to less caregiver burden. Thirty-seven percent of caregivers reported receiving assistance from external sources. The relationship between the child's level of disability and caregiver burden was attenuated by family support for caregiving and by school attendance in a general class in a mainstream public primary school.*

Conclusion: *External sources of support reduce caregiver burden but may not be available to many caregivers.*

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INTRODUCTION

Down Syndrome (DS) is caused by the presence of an extra chromosome (known as trisomy 21) and affects approximately 1 in 1,000 to 1 in 1,100 new-borns (World Health Organisation, n.d.). Individuals with DS have a characteristic set of physical features including hypotonia, small mouth and ears, and a flat nasal bridge, which are often compounded with gastrointestinal tract anomalies and obstructive sleep apnea (Korenberg et al, 1994; Pediatrics, 2001). Children with DS are often prone to physical conditions including ligamentous laxity, decreased strength, and shortened arms and legs, that can inhibit the rate of gross motor development (Fish, 2010). As children with DS reach school-age, verbal, functional, and attention deficits become more pronounced (Grieco et al, 2015).

Caring for Children with Down Syndrome

Caregivers and families of children with DS are disproportionately burdened when compared to caregivers of children and youth with other special health care needs (Phelps et al, 2012).Parents of children with DS are significantly more likely to cut back their working hours or stop working due to their child's health (Phelps et al, 2012). Children with Down Syndrome have more pronounced unmet needs and, consequently, parents of these children feel more stress, which is attributed to caregiving (Hauser-Cram & Shonkoff, n.d.; McGrath et al, 2011; Skotko et al, 2011). Parents and caregivers of children with DS have also cited important personal lessons such as developing increased patience, acceptance, and flexibility (Skotko et al, 2011).

Children with DS stand to benefit from involvement in peer support groups to overcome limitations in creating and maintaining peer social networks during key developmental periods (Fish, 2010). Children with DS have generally been found to have lower physical activity levels when compared with their peers (Wentz et al, 2021). Subsequently, physical therapy has also often been recommended to minimise abnormal compensatory movement patterns (Fish, 2010). Globally, paediatric physical therapists have recommended more physical activity in children with DS, beginning in infancy through to adolescence (Wentz et al, 2021).

Support for Caregivers

Given the difference in stress between varying neurodevelopmental disorders, interventions that promote support to children and their families should be syndrome-specific (Ashworth et al, 2019). Presently, many low- and middle-income countries lack institutionalised support systems for children with neurodevelopmental disabilities. This places an added burden on caregivers (Al-Kandari & Al-Qashan, 2010). Subsequently, many parents turn to the community and/or family resources for support, but many others lack the knowledge of which support systems exist and/or feel uncomfortable soliciting such support (Al-Kandari & Al-Qashan, 2010).

Generally, parents of children with DS have recommended seeking out resources and support groups such as early intervention programmes, seminars or workshops to others in similar circumstances (Skotko et al, 2011). Other recommendations to caregivers of children with DS have included finding a good physician and learning how to advocate on behalf of their child's needs (Skotko et al, 2011). Some low- and middle-income countries, such as Zambia, have introduced new programmes to train caregivers to be interventionists, using empirically supported and parent-mediated interventions for developmental differences such as DS (Pierucci et al, 2023). These interventions aim to target improvements in children's language skills and their ability to play (Pierucci et al, 2023).

Support for Children with Disabilities and their Caregivers in Sri Lanka

Within Sri Lanka, DS is the most commonly identified aneuploidy or abnormality in the number of chromosomes in children, with a prevalence of 76.3% amongst clients with chromosomal anomalies (Thillainathan et al, 2015). While some demographic studies on the prevalence of generalised chromosomal anomalies have taken place in Sri Lanka, little research has been conducted regarding the availability, utilisation and effectiveness of support systems and resources specifically for children with DS and their caregivers.

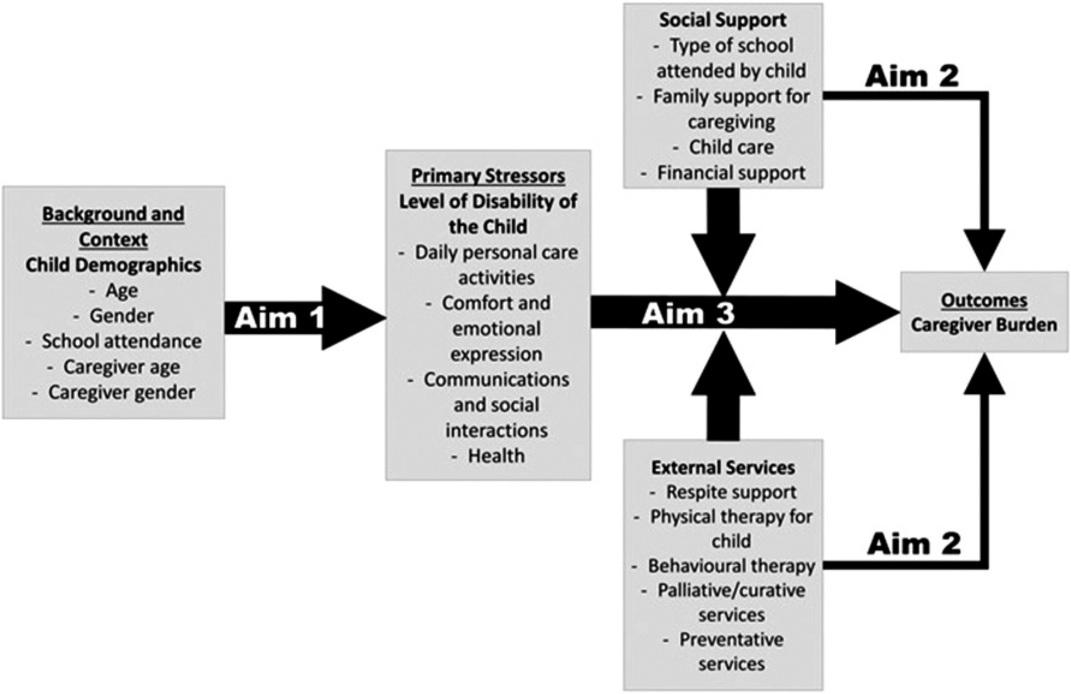
Children with disabilities in Sri Lanka, like children elsewhere, have been found to benefit from inclusive education models (Furuta, 2006). Examples of such models include special programmes in formal education settings, special schools within resource centres, and/or non-formal educational activities that could function as alternative educational opportunities (Furuta, 2006). Moreover, family, community, spiritual, cultural, and rehabilitative outcomes have been found to positively influence mothers' perceptions of their child's disability in Sri Lanka (Landry et al, 2015). Access to medical, social, rehabilitative, and/or public health infrastructure have also been suggested to be critical in alleviating the burden that caregivers face (Landry et al, 2015).

Objective

This study had three aims focused on understanding the experiences of children with Down Syndrome, aged 5-12 years, and their caregivers in Galle, Sri Lanka (see Figure 1):

1. To describe the general level of disability amongst school-aged children with DS and examine possible socioeconomic predictors of the level of disability;
2. To assess the need for and use of DS-related external services and social support; and,
3. To analyse whether primary stressors, social support and external services moderate the relationship between level of child disability and overall caregiver burden.

Figure 1: Description of the Study Aims



METHOD

Study Setting

This study took place in the Galle District on Sri Lanka's south-western coast. The study consisted of interviews and surveys with caregivers of children with Down Syndrome, and was conducted through the Faculty of Medicine at the University of Ruhuna.

Study Participants

Caregivers of children with DS between the ages of 5 and 12 years were selected through direct contact at standard schools with standard classes, standard schools with special needs classes, and schools designed specifically for children with varying disabilities. Through word-of-mouth, attending special needs drop-in centres, and connecting with local social services, researchers also approached caregivers of children who did not attend school.

Inclusion criteria for caregivers:

- They should have lived within the Galle District for at least six months;
- Be able to speak, read, and understand Sinhala or English; and
- Be a primary caregiver for a child with DS (Wijesinghe et al, 2015).

Verbal and written consent was obtained from all the participants.

Data Collection

This study was based upon an explanatory mixed-methods design comprised of a two-phased qualitative and quantitative approach consisting of a 75-question survey and a 10-question semi-structured interview with a smaller subset of the sample. The survey consisted of questions from two pre-existing scales: the Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD) and the Caregiver Difficulties Scale (CDS). Additional questions about demographics and the availability of medical-, rehabilitative-, respite-, counselling- and other services were also included. Participants were asked to elaborate on key points of interest from the survey. Both the survey and interview questions were written in English and translated into Sinhala. A researcher conducted both the surveys and interviews in Sinhala.

Participants were compensated with 700 Sri Lankan rupees (LKR) for filling out the survey and another 700 LKR for participating in the qualitative interview.

Measures

CPCHILD Assessment

The CPCHILD questionnaire was developed by healthcare professionals from the Hospital for Sick Kids and the Bloorview Research Institute in Toronto (Narayanan et al, 2006). The measure assesses children's levels of disability from the perspective of their caregiver. Caregivers are asked to rank their child's abilities, emotions and health across four domains: Personal Care/Activities of Daily Living; Comfort and Emotions; Communication and Social Interaction; and Health (Narayanan et al, 2006, 2007). Generally, caregivers were asked to assess the level of difficulty or discomfort in the child when trying to perform certain functions of daily living, such as eating, bathing, getting in and out of vehicles, and communicating with different groups of people over the prior two weeks. Measures of level of difficulty varied and were based on the set of questions. Some questions use a 3-point Likert scale ranging from "substantial" difficulty to "none", while others use a 7-point Likert scale ranging from "not possible" to "no problem." For each question, raw scores were later converted into a scale ranging from '0' (worst outcome) to '100' (best outcome) (Narayanan et al, 2007). CPCHILD provides a summary of five continuous variables: one overall mean score and four subscale mean scores ranging from '0' to '100', with higher values associated with lower levels of childhood disability.

CPCHILD has been confirmed to have sufficient reliability and validity as a measure in multiple studies for applications with cerebral palsy (Narayanan et al, 2006; Zalmstra et al, 2015). The section on mobility was removed in this study since it was less applicable to children with DS, and this was accounted for when tabulating overall scores.

CDS Assessment

The Caregiver Difficulties Scale (CDS) was created by researchers at the University of Ruhuna, Galle (Wijesinghe et al, 2015). This assessment measures caregiver burden by asking 25 questions associated with "concern for the child", "impact on self", "support for caregiving", and "social and economic strain" using a 5-point Likert scale (Wijesinghe et al, 2015). The survey consisted of a series of caregiving-related questions including asking whether caregivers feared for their child's present or future state of being; whether caregivers felt their personal health was affected by their child's condition; and whether specific people (e.g., spouse, neighbours, other family members) help caregivers with their physical and emotional caregiving responsibilities. Caregivers' responses to each question were assessed on a 6-point scale ranging from "always" to "not applicable." A final aggregate score was calculated for overall caregiver burden in the sample by calculating the mean of all CDS scores ranging from '0' to '100'. The face-, construct-, content-, and consensual validity of the CDS have been established through item generation in previous studies (Wijesinghe et al, 2013). Previous tests have also established satisfactory internal consistency and reliability (Wijesinghe et al, 2013).

Data Analysis

STATA version SE/15 (StataCorp., 2017) was used to analyse the data and generate descriptive statistics. For categorical variables, raw data frequencies and percentages were tabulated. CPCHILD and CDS scores were calculated for each participant and aggregated to estimate subsection and overall means and standard deviations. Child and caregiver demographic information were then related to CPCHILD and CDS scores. Relationship and demographic indicators of level of child disability and caregiver burden were estimated through simple linear regression analysis. Significant variables were entered into multivariable regression models to assess the effect on CPCHILD and CDS scores. The associations between CPCHILD and CDS scores were also assessed, using a significance level of 0.05. Variables that yielded significant associations were included in further multivariable regression models to estimate possible moderator effects of specific social supports and external services on the relationship between level of disability of the child and caregiver burden.

Qualitative data was analysed separately. A thematic analysis was conducted using NVIVO to identify common phrases, trends, and themes. Per-question summaries were drafted and revisited to determine commonly recurring cross-sectional themes across questions.

Ethics Approval

This study was approved by the Institutional Reviews Board at the University of Ruhuna, Sri Lanka, and Duke University, USA.

RESULTS

Participant Characteristics

One hundred and twenty-five caregivers of children with Down Syndrome participated in the study. The mean age of the children was 8.4 ± 2.2 years with a range from 5 to 12 years (see Table 1), and 50.4% of children were male. Among them, 18.4% had never attended school, 5.6% were currently not attending school and 76% were currently attending school. The majority (88%) did not follow the regular curriculum, and most of them attended either a special class in a mainstream public school (38.5%) or a special school (32.8%) for children with disabilities.

Table 1: Child Characteristics

Variable	Frequency	%	Mean (SD)	(Min, Max)
Child Sex (n=125)				
Male	63	50.40		
Female	62	49.60		
Child Age (n=125)			8.4 (2.2)	(5, 12)
Child Educational Status (n=125)				
Never schooled	23	18.40		
Currently non-schooling	7	5.60		
Attending preschool	5	4.00		
Attending school	90	72.00		
Child Grade (n=125)				
1st grade	8	6.40		
2nd grade	3	2.40		
3rd grade	2	1.60		
4th grade	1	0.80		
5th grade	1	0.80		
Not applicable	110	88.00		
School Type (n=122)				
Not applicable	29	23.77		
Mainstream school – general class	6	4.92		
Mainstream school – special class	47	38.52		
Special school	40	32.79		
Number of Years of Schooling (n=122)			2.0 (1.8)	(0, 8)
Cardiac Comorbidities (n=125)	36	28.80		
Respiratory Comorbidities (n=125)	7	5.60		
Gastrointestinal Comorbidities (n=125)	32	25.60		
Other Comorbidities (n=125)	62	49.60		

Caregiver characteristics are presented in Table 2. Most of the caregivers (83.2%) were female and were on average 46.7 ± 8.8 years old at the time of the survey. Most of them (90.4%) were married and almost all (96%) were the biological parent of the child with DS. Approximately one-third (27.2%) worked in either full-or part-time jobs, with another third (32%) reporting that they were unemployed.

Table 2: Caregiver Characteristics

Variable	Frequency	%	Mean (SD)	(Min, Max)
Caregiver Sex (n=125)				
Male	21	16.80		
Female	104	83.20		
Age at Birth of Child (n=125)			37.9 (8.9)	(9.0, 64.4)
<30 years	29	23.20		
31-40 years	53	42.40		
>40 years	43	34.40		
Caregiver Work Status (n=125)				
Unemployed (due to child's health)	40	32.00		
Unemployed (for other reasons)	0	0.00		
Searching for a job	0	0.00		
Working full- or part-time (outside the house)	26	20.80		
Working full- or part-time (at a home-based business)	9	7.20		
Homemaker	50	40.00		
Caregiver Marital Status (n=125)				
Single	2	1.60		
Married	113	90.40		
Widowed	5	4.00		
Divorced/Separated	5	4.00		
Caregiver Education Level (n=125)				
No school	2	1.60		
Below grade 5	7	5.60		
Fifth to tenth grade	21	16.80		
Up to ordinal level	51	40.80		
Post-high school diploma or up to advanced level	37	29.60		
Degree or diploma	7	5.60		
Monthly Income (in LKR) (n=125)				
Less than 5,000	17	13.60		
5,001 - 20,000	63	50.40		
20,001 - 35,000	24	19.20		
35,001 - 50,000	19	15.20		
Over 50,000	2	1.60		

Aim 1: Child and Caregiver Demographics and Level of Disability (CPCHILD)

The overall mean level of child disability was 64.8, with sub-scores of 59.6 for personal care and activities of daily living; 68.4 for comfort and emotions; 68.4 for communication and social interaction; and 47.8 for health.

All demographic variables were regressed against CPCHILD in simple models to assess for significant associations. Significant associations are listed in Table 3 and were later included in the multivariable model. The type of school that children attended was significantly associated with overall CPCHILD scores. Caregivers who had children attending general classes in mainstream schools reported lower levels of disability in their children (β=17.7, $p<0.05$), as did those whose children attended special classes in mainstream schools (β=13.4, $p<0.05$) and in special schools (β=12.8, $p<0.05$), in comparison to children who were not attending school. Female caregivers reported lower levels of disability in their children (β=7.5, $p<0.05$) as compared to their male counterparts, as did caregivers who worked full- or part-time at home-based businesses (β=10.3, $p<0.05$) and those who self-identified as homemakers (β=5.6, $p<0.05$). Caregivers' education level was also significantly associated with CPCHILD scores. Caregivers who completed secondary school reported their children had lower levels of disability (β=12.9, $p<0.05$), as did those who had a degree or diploma (β=21.5, $p<0.05$), in comparison to those with less than a 10th-grade education.

Variables which had significant associations with the CPCHILD assessment were included in linear multivariable regression models (Table 3). In the multivariable model, female caregivers had a significant association with reporting lower levels of child disability (β=9.3; $p<0.05$). Moreover, caregivers of children who attended general classes in mainstream schools (β=18.1, $p<0.05$), special classes in mainstream schools (β=11.6, $p<0.05$) and special schools (β=12.0, $p<0.05$) were also associated with lower levels of child disability.

Table 3: Child and Caregiver Demographics with Level of Disability of Child (CPCHILD scores): Simple and Multivariable Linear Regression

	CPCHILD Simple Regression	**CPCHILD Multivariable Regression**
	Coefficient (95% CI)	**Coefficient (95% CI)**
Model Intercept		**29.1 (14.3, 43.9)**
Educational Status		
Never schooled (REF)	53.2 (48.2, 58.2)	
Currently non-schooling AND/OR Attending preschool	6.1 (-2.5, 14.6)	5.6 (-5.2, 16.4)
Attending school	15.3 (9.7, 20.9)**	-13.7 (-28.0, 0.6)
School Type		
Not applicable (REF)	54.7 (50.1, 59.2)	
Mainstream school - general class	17.7 (6.6, 28.7)**	18.1 (6.3, 29.9)**
Mainstream school - special class	13.4 (7.6, 19.2)**	11.6 (5.1, 18.2)**
Special school	12.8 (6.8, 18.8)**	12.0 (5.4, 18.6)**
Caregiver Gender		
Male (REF)	58.6 (52.8, 64.3)	
Female	7.5 (1.3, 13.8)**	9.3 (2.6, 16.1)**
Caregiver Work Status		
Unemployed (due to child's health) (REF)	60.6 (56.4, 64.8)	
Working full- or part-time (outside the house)	6.1 (-0.6, 12.7)	5.2 (-2.3, 12.7)
Working full- or part-time (at a home-based business)	10.3 (0.6, 20.0)**	9.1 (-0.7, 18.9)
Homemaker	5.6 (0.0, 11.2)**	1.3 (-4.3, 6.9)
Caregiver Education Level		
Less than grade 5 (REF)	54.5 (45.9, 63.1)	
Fifth to tenth grade	6.1 (-3.h1, 17.3)	-0.3 (-10.7, 10.1)
Up to ordinal level	12.9 (3.6, 22.1)**	8.0 (-1.5, 17.5)
Post-high school diploma or up to advanced level	9.2 (-0.4, 18.7)	5.5 (-4.2, 15.1)
Degree or diploma	21.5 (8.6, 34.5)**	11.7 (-2.5, 25.8)
Caregiver Income Level* **		
Less than 5,000 (REF)	58.0 (51.7, 64.4)	
5,001 - 20,000	5.9 (-1.2, 13.1)	2.3 (-4.8, 9.3)
20,001 - 35,000	11.4 (3.1, 19.7)**	2.8 (-5.7, 11.2)
35,001 and above	9.8 (1.3, 18.3)**	-0.8 (-10.6, 9.0)

Note: CI, confidence interval
**P<0.05
***1 USD = 160 LKR at the time of the survey”

Aim 2: Use of Social Support Systems and External Services

Table 4 outlines the frequencies in responses for social supports used by children with DS and their caregivers. The majority (56%) of caregivers stated that they had more than one co-caregiver, with most naming their spouse as the main co-caregiver (31.2%). Almost all (92.8%) caregivers reported receiving caregiving assistance from family and friends.

Table 4: Social Supports for Children with Down Syndrome and their Caregivers

Variable	Frequency	%
Main Co-Caregiver (n=125)		
Spouse	39	31.20
Grandparent caregivers	1	0.80
Siblings of the child	9	7.20
Other relatives	4	3.20
Neighbours	1	0.80
None	1	0.80
Other – More than one co-caregiver	70	56.00
Receive Government Assistance (n=125)		
Yes	25	20.00
Receive Social Services (n=125)		
Yes	25	20.00
Receive NGO Assistance (n=125)		
Yes	0	0.00
Religious Observances (n=125)		
Yes	97	77.60
Type of Religious Observance (n=36)		
Buddhist rituals	34	94.44
Islamic rituals	1	2.78
Going to temple (unspecified religion)	1	2.78
Participate in Recreational Activities (n=125)		
Yes	4	3.20
Family and Friends Support (n=125)		
Yes	116	92.80
Provider of Support (n=28)		
Spouse	3	10.71
Other family member	25	89.29
Support Level Provided by Family/Friends (n=116)		
Full	37	31.90
Almost full	49	42.24
Partial	25	21.55
Minimal	5	4.31

Table 5 describes the use of external services by caregivers for themselves and their child. Of those surveyed, 7.2% said they had special facilities in their home to support their child. Approximately one-third (36.8%) utilised external support for caregiving. Very few reported using behavioural therapy (16.8%) for their children.

Table 5: External Service Use for Children with Down Syndrome and their Caregivers

Variable	Frequency	%
Special Facilities at Home (n=125)	9	7.20
External Support (n=125)	46	36.80
Behaviour Therapy (n=125)	21	16.80
Physical Therapy (n=125)	15	12.00
Palliative Therapy (n=125)	8	6.40
Preventive Therapy (n=125)	15	12.00

Most interviewed caregivers had minimal knowledge of Down Syndrome prior to being tasked with caregiving responsibilities. They reported that they obtained information about DS from medical professionals following the delivery of their child. Many learned about DS from attending clinics conducted by the hospital, reading existing literature, and conducting internet searches. Caregivers used a variety of strategies including speaking to doctors and other parents, and engaging with television, newspaper, and internet sources to fill gaps in their understanding.

There were broad variations in the caregivers' experiences of finding external resources such as physical, behavioural, and speech-language therapies for their child. Approximately half (46.7%) of those interviewed reported having no trouble finding therapies for their child or not requiring any therapy for their child. Almost half (42.8%) of the participants had trouble accessing resources such as physiotherapy and speech-language therapy because of transportation or timing issues, such as not being able to receive sufficient time off work.

When speaking of challenges, the most prominent difficulties reported were related to family finances, managing the child's comorbidities, and finding adequate schooling. Many caregivers stated that their income was not satisfactory to meet expenses and that they did not have sufficient time to take on additional economic opportunities.

Moreover, caregivers struggled to find ways to cope with their child's comorbidities, such as learning how to interact with their child given the speech impediments, or navigating medical problems and challenges. When facing both financial issues and difficulty in supporting their child's comorbidities, one caregiver reported using the internet to find materials to conduct speech therapy to assist her son by herself.

Caregivers also reported difficulties in finding teachers and classrooms that could support their child's development, with most reporting that they sent their children to special schools or special units in mainstream schools. Factors that influenced these decisions included proximity to the school, rejection of the child by teaching staff in mainstream schools, referrals from other parents, and the opportunity for increased individual attention. One caregiver stated that her child had studied in a mainstream school up until the fifth grade but began struggling to keep up with the class as the curriculum progressed, prompting them to transition the child into a special education unit. Some caregivers reported taking part in advocacy and awareness initiatives by supporting other children with DS in their spare time, and advocating to local government authorities to develop improved facilities that would be more accessible to children with DS. For instance, one caregiver, who also worked as a teacher, said that she tries to encourage parents of children with DS to enrol their children in school.

Aim 3: Relationship of Child's Level of Disability on Overall Caregiver Burden (CDS score)

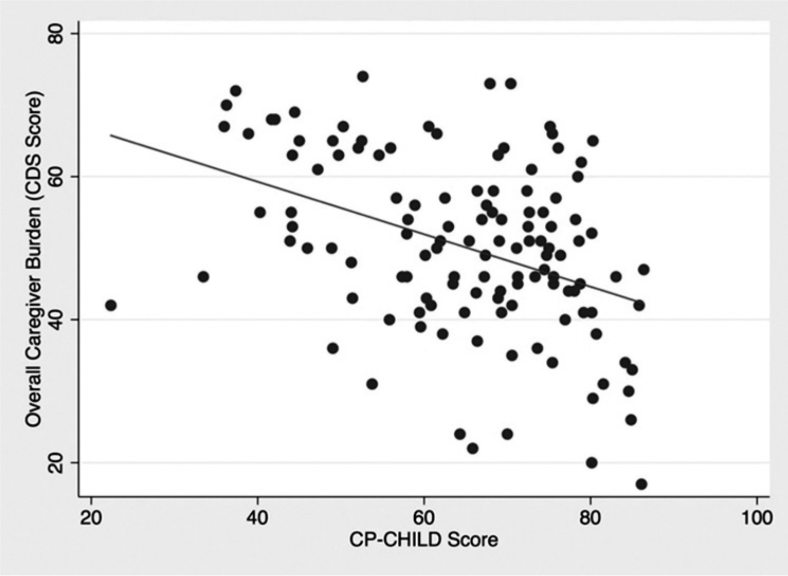
Overall, the mean CDS score for caregiver burden was 50.2. A simple linear regression model for mean CDS score and the level of disability (CPCHILD) was estimated (see Table 6). Lower caregiver burden scores were associated with lower levels of child disability for personal care and activities of daily living ($\beta=-0.2$, $p<0.05$), comfort and emotions ($\beta=-0.3$, $p<0.05$), communication and social interaction ($\beta=-0.2$, $p<0.05$), and health ($\beta=-0.5$, $p<0.05$), but the association for overall child disability scores ($\beta=-0.4$) was not statistically significant (see Figure 2).

Table 6: Relationship between Level of Disability of Child (CPCHILD score) and Overall Caregiver burden (CDS score): Simple Linear Regression

	CDS Mean Coefficient (95% CI)
CPCHILD Personal Care/Activities of Daily Living	
Model intercept	59.7 (54.8, 64.6)
Coefficient	-0.2 (-0.2, -0.1)**
CPCHILD Comfort and Emotions	
Model intercept	71.5 (58.1, 84.9)
Coefficient	-0.3 (-0.4, -0.1)**
CPCHILD Communication and Social Interaction	
Model intercept	67.1 (53.7, 80.5)
Coefficient	-0.2 (-0.4, -0.1)**
CPCHILD Health	
Model intercept	71.9 (60.8, 82.9)
Coefficient	-0.5 (-0.7, -0.2)**
CPCHILD Overall	
Model intercept	74.0 (64.1, 83.7)
Coefficient	-0.4 (-0.5, -0.2)

Note: CI, confidence interval
**P<0.05

Figure 2: Relationship between Child Disability (higher CPCHILD score indicates lower level of disability) and overall Caregiver Burden (CDS score)



Aim 3: Relationship of Child’s Level of Disability, Social Support Use and External Service Use on Overall Caregiver Burden (CDS score)

Table 7 outlines simple linear regressions of external service use and social support use against the child’s level of disability (CPCHILD score) and overall caregiver burden (CDS score). Family support for caregiving ($\beta=-14.2$, $p<0.05$) and school attendance in a general class in a standard school ($\beta=-15.1$, $p<0.05$) yielded significant negative associations.

Multivariable regressions were developed based on significant associations from the simple linear regressions for social supports and external services with overall caregiver burden. The multivariable models regressed the CPCHILD score, child’s school type, family support for caregiving, use of government assistance, and assistance from external sources against caregiver burden scores. Since none of the external service variables were found to be significant in the simple regression models, only the assistance from external sources variable was further analysed. The child’s level of disability ($\beta=-0.4$, $p<0.05$) became significantly associated with caregiver burden when controlling for the effects of other variables. Family support for caregiving ($\beta=-10.1$, $p<0.05$) was significantly associated with lower caregiver burden.

Table 7: Relationships between Child’s Level of Disability, Social Support, and External Service Use with Caregiver Burden (CDS score): Simple Regression and Multivariable Regression with and without interaction terms

	CDS Simple Regression	**CDS Multivariable Regression**	**CDS Multivariable Regression (incl. Social Support as moderator)**	**CDS Multivariable Regression (incl. External Services as moderator)**
	Coefficient (95% CI)	**Coefficient (95% CI)**	**Coefficient (95% CI)**	**Coefficient (95% CI)**
Model Intercept	**-**	**80.4 (69.6, 91.2)**	**81.1 (41.2, 121.0)**	**67.4 (51.8, 83.0)**
CPCHILD Score	74.0 (64.1, 83.7)	-0.4 (-0.6, -0.3)**	-0.3 (-1.0, 0.3)	-0.3 (-0.5, -0.0)**
Coefficient	-0.4 (-0.5, -0.2)			
School Type (n=122)				
No school (REF)	50.4 (46.0, 54.8)	REF	REF	
Mainstream school – general class	-15.1 (-25.8,-4.5)**	-4.7 (-14.4, 4.9)	-1.0 (-79.4, 77.5)	
Mainstream school – special class	1.4 (-4.2, 7.0)	8.9 (3.5, 14.3)**	10.6 (-13.0, 34.2)	
Special school	0.1 (-5.7, 5.9)	6.0 (0.5, 11.5)**	10.6 (-13.0, 38.9)	
Family Support for Caregiving	63.3 (55.6, 71.1)	-10.1 (-17.3,-2.9)**	-10.3 (-47.5, 26.9)	
Coefficient	-14.2 (-22.2, -6.1)**			

	CDS Simple Regression	**CDS Multivariable Regression**	**CDS Multivariable Regression (incl. Social Support as moderator)**	**CDS Multivariable Regression (incl. External Services as moderator)**
	Coefficient (95% CI)	**Coefficient (95% CI)**	**Coefficient (95% CI)**	**Coefficient (95% CI)**
Model Intercept	**-**	**80.4 (69.6, 91.2)**	**81.1 (41.2, 121.0)**	**67.4 (51.8, 83.0)**
Government Assistance	49.0 (46.6, 51.4)	5.0 (0.2, 9.8)**	-17.6 (-44.8, 9.7)	
Coefficient	5.7 (0.4, 11.1)			
At-Home Special Families	49.9 (47.7, 52.2)			
Coefficient	3.2 (-5.2, 11.6)			16.9 (-14.0, 47.8)
Assistance from External Sources	51.6 (48.9, 54.3)	-2.4 (-6.3, 1.6)		
Coefficient	-3.8 (-8.3, 0.7)			21.6 (-0.9, 44.0)
Behaviour Therapy	50.1 (47.7, 52.5)			
Coefficient	0.4 (-5.4, 6.3)			38.4 (8.9, 67.8)**
Physical Therapy	50.6 (48.3, 52.9)			
Coefficient	-3.8 (-10.5, 2.8)			-11.5 (-40.9, 17.9)
Palliative/Curative Therapy	49.9 (47.7, 52.2)			
Coefficient	3.4 (-5.5, 12.3)			-27.3 (-70.6, 16.1)
Preventive Care Services	50.1 (47.8, 52.4)			
Coefficient	0.6 (-6.1, 7.3)			-34.6 (-61.3, -7.9)
Interaction Terms				
CPCHILD Score*No school			REF	
CPCHILD Score*Mainstream school – general class			-0.1 (-1.2, 1.0)	
CPCHILD Score*Mainstream school – special class			-0.1 (-0.4, 0.3)	
CPCHILD Score*Special school			-0.1 (0.6, 0.3)	
CPCHILD Score*Family Support for Caregiving			-0.0 (-0.6, 0.6)	
CPCHILD Score*Government Assistance			0.4 (-0.1, 0.8)	
CPCHILD Score*At-Home Special Facilities				-0.3 (-0.8, 0.2)
CPCHILD Score*Assistance from External Sources				-0.3 (-0.7, 0.0)**
CPCHILD Score*Behaviour Therapy				-0.7 (-1.2, -0.2)**
CPCHILD Score*Physical Therapy				0.2 (-0.3, 0.6)
CPCHILD Score*Palliative/ Curative Therapy				-0.5 (-0.2, 1.3)
CPCHILD Score*Preventive Care Services				0.6 (0.1, 1.1)**

Note: CI, confidence interval
**P<0.05

Aim 3: Social Supports as a Moderator of Level of Child's Disability (CPCHILD score) on Overall Caregiver Burden (CDS score)

The moderator effects of specific social supports on the relationship between the child's level of disability and caregiver burden were analysed using multivariable regressions. For these models, three variables with significant associations in the simple models were selected: school type, family support for caregiving, and government assistance (see Table 7). No significant associations were observed for any of the tested variables within this multivariable model.

Aim 3: External Services as a Moderator of Level of Child's Disability (CPCHILD score) on Overall Caregiver Burden (CDS score)

The moderator effects of external services on the relationship between the child's level of disability and caregiver burden were assessed, using all external service variables (see Table 7). In simple regressions, the overall CPCHILD score (β=-0.3; $p<0.05$) and receiving behavioural therapy (β=38.4, $p<0.05$) were significantly associated with caregiver burden. When included as interaction terms in a multivariable model, three of the variables: assistance from external sources (β=-0.3, $p<0.05$), behavioural therapy (β=-0.7, $p<0.05$), and preventive care (β=0.6, $p<0.05$) displayed significant associations.

DISCUSSION

Socioeconomic Predictors of the Level of Disability

CPCHILD has not been previously used to measure disability levels for DS, making it difficult to draw comparisons with populations elsewhere. In earlier CPCHILD studies of children with cerebral palsy, severity scores were reported to range between 52.0 and 56.2. This study's adjusted score of 64.8 demonstrates lower levels of disability than studies for children with cerebral palsy (Narayanan et al, 2006; Zalmstra et al, 2015).

Significant predictors of the caregivers' perception of their child's level of disability included school type, caregiver sex, caregiver work status, caregiver education level and family income level. Significant relationships were found between enrolments in general classes and lower caregiver burden scores. Levels of perceived disability were lower amongst children who attended general classes in mainstream schools than among children who attended special classes or special schools. The decision of which school type to enrol children in is ultimately at the discretion of the caregiver in Sri Lanka (Padmani, 2003), with previous studies indicating that the mental age scores of children with DS are strongly related to the type of school attended (Sloper et al, 1990). It is likely that children attending general classes can do so because they have lower levels of disability.

Consistent with existing literature, most caregivers were female – possibly due to historical and cultural traditions that place more caregiving responsibilities on women (Barros et al, 2017). Lower levels of caregiver burden were associated with older children, suggesting that there may be an association between perceived decreases in the level of the child's disability by caregivers as children age. This could possibly stem from caregivers having a better understanding of how to manage their child's disability and learning styles (Fish, 2010).

External Services and Social Support for Children and Caregivers

Despite low uptake of services, caregivers identified needs for external services. For example, caregivers reported a need for speech-language therapy. As children age, disability related to language, particularly the use of expressive language, declines (Chapman, 1997; Grieco et al, 2015). Barriers to accessing speech-language therapy included a lack of knowledge on locating services, access barriers (either through transportation or lack of time), and/or an inability to afford services.

It is of particular significance to note that this study did not specify what participants thought external services were and so it is unclear what services participants were thinking about as they answered questions related to external services. Such issues could be mitigated in future studies by conducting cognitive interviews of survey questions prior to widely conducting the survey with participants, and/or amending questions to call out specific types of external services of interest.

Relationship of External Services, Social Supports, and Primary Stressors with overall Caregiver Burden

Lower levels of disability amongst children with DS were related to lower overall levels of caregiver burden. This is consistent with previous studies where CDS measured caregiver burden for children with cerebral palsy in Sri Lanka and found that a child's functional deficits contributed to caregiver burden (Wijesinghe et al, 2015). It could also be postulated that when there is a higher level of disability in the child, caregiver burden will subsequently be higher as well. This is particularly showcased within the context of how it was found that caregiver burden was reported be higher among caregivers of children attending special schools or special classes in mainstream schools. The causal direction of this relationship warrants further study.

Similar to the previous discussion of the term 'external services', future studies should take care to either more specifically define what is encompassed within government services or conduct cognitive testing of survey items prior to widespread use to ensure participants' understanding of the questions are in line with researcher intents of such probes.

Moderator Effects of Social Supports and External Services on Level of Disability and Caregiver Burden

Mean CDS scores were regressed against mean CPCHILD scores, demographic, and resource access information. No support services were found to have a significant moderating role on the relationship between the level of disability and caregiver burden. This information conflicts with previous studies that have shown social supports to be a moderator of caregiver well-being (Demirtepe-Saygılı & Bozo, 2011). It is possible that the interpretation of social supports within this study was misunderstood by caregivers because it was presented generally, without providing examples within the survey, leaving it open to broad interpretation. This issue could be overcome in future studies by conducting cognitive interviews to test participant interpretations of such questions prior to deployment.

In contrast, certain external services were shown to have significant associations with caregiver burden, and acted as moderators of the relationship between disability level and caregiver burden in this study. Receiving assistance from external sources and attending behavioural therapy were found to be associated with lower levels of caregiver burden. Since these sources are designed to help mitigate the child's level of disability and improve coping mechanisms (for the child and/or caregiver), this association is consistent with prior assumptions.

Study Strengths and Limitations

Previous studies in Sri Lanka have used similar measures to assess other developmental disabilities within the country but this was the first study to apply this measure to better understand DS. A strength of this study was its diverse participant pool in terms of sociodemographic backgrounds and access to disability services. As to limitations, both CPCHILD and CDS were originally designed for children with CP. Moreover, there were limited descriptions of resource availability, external services, and social supports. Given that the section on mobility was removed from the CPCHILD tool that was used in this study, the reliability or validity of the overall tool may have been somewhat affected. While measures were taken to remove that portion from any calculations that were done to account for this loss, future studies would benefit from assessing the reliability and validity of the modified tool. Furthermore, both sets of quantitative and qualitative tools used general language that may have left room for differing interpretations. Future studies could also include objective clinical assessments of children to measure level of disability more and compare those results against caregiver perceptions of their child’s level of disability.

As this was a cross-sectional study, care should be taken in drawing causal inferences about the observed associations. Longitudinal studies would better assess the effects of access and availability on the child’s level of disability or caregiver burden for DS in Sri Lanka over time. Future research is also necessary for further examination of resource availability and usage for children with DS and their caregivers in the region. Studies should further investigate the barriers preventing resource accessibility for caregivers and their children with DS and better understand the impact of comorbidities for children with DS in Sri Lanka.

CONCLUSION

Overall, higher levels of child disability were associated with higher levels of caregiver burden. Despite moderate levels of child disability, approximately one-third of caregivers reported receiving assistance from external supports, with most caregivers relying on assistance from friends and family.

Future research is warranted to further examine resource availability and usage for children with DS and their caregivers within the region. Larger samples and expansion to other regions within Sri Lanka could further improve understanding of the challenges faced by caregivers and their children with DS, especially given the recent economic challenges that Sri Lanka is facing. Future studies should also seek to understand how terms such as external services, government services, and social supports are interpreted by local populations prior to widespread deployment, through cognitive testing of survey questions in the context of caregiving for children with Down Syndrome in Sri Lanka and/or south Asia.

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The authors declare no conflicts of interest.

The first author can be contacted regarding further analyses of the study data.

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