Physical and mental wellbeing of parents of children with Down Syndrome in Ireland

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CARL Research Project

in collaboration with

DOWN SYNDROME IRELAND

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What is Community-Academic Research Links?

Community Academic Research Links (CARL) is a community engagement initiative provided by University College Cork to support the research needs of community and voluntary groups/ Civil Society Organisations (CSOs). These groups can be grassroots groups, single-issue temporary groups, but also structured community and voluntary organisations. Research for the CSO is carried out free of financial cost by student researchers.

CARL seeks to:

- provide civil society with knowledge and skills through research and education;
- provide their services on an affordable basis;
- promote and support public access to and influence on science and technology;
- create equitable and supportive partnerships with civil society organisations;
- enhance understanding among policymakers and education and research institutions of the research and education needs of civil society, and
- enhance the transferrable skills and knowledge of students, community representatives and researchers (Living Knowledge Network).

What is a CSO?

We define CSOs as groups who are non-governmental, non-profit, not representing commercial interests, and/or pursuing a common purpose in the public interest. These groups include: trade unions, NGOs, professional associations, charities, grass-roots organisations, organisations that involve citizens in local and municipal life, churches and religious committees, and so on.

Why is this report on the UCC website?

The research agreement between the CSO, student and CARL/University states that the results of the study must be made public through the publication of the final research report on the CARL (UCC) website. CARL is committed to open access, and the free and public dissemination of research results.

How do I reference this report?

Author (year) *Dissertation/Project Title*, [online], Community-Academic Research Links/University College Cork, Ireland, Available from: https://www.ucc.ie/en/scishop/rr/ [Accessed: date].

How can I find out more about the Community-Academic Research Links and the Living Knowledge Network?

The UCC CARL website has further information on the background and operation of Community-Academic Research Links at University College Cork, Ireland, http://carl.ucc.ie. You can follow CARL on Twitter at @UCC_CARL. All of our research reports are accessible free online here.

CARL is part of an international network of Science Shops called the Living Knowledge Network – website and on Twitter @ScienceShops. CARL is also a contributor to Campus Engage, which is the Irish Universities Association engagement initiative to promote community-based research, community-based learning and volunteering amongst Higher Education students and staff.

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Student Declaration

I confirm that this project submission is my own, original work. All sources and publications have been recognised and referenced appropriately.

Signed: ______________________________

Abbreviations

Please refer below for a list of abbreviations used throughout this study.

ADHD	Attention Deficit Hyperactivity Disorder
ASD	Autism Spectrum Disorder
CUES	Carers' and Users' Expectations of Services Questionnaire
CWS	Carer Wellbeing and Support Questionnaire
DASS-21	Depression Anxiety Stress Scales 21-point Questionnaire
DD	Developmental Disability
DS	Down Syndrome
DSM	Diagnostic and Statistical Manual of Mental Disorders
DT-P	Distress Thermometer for Parents
GHQ-12	General Health 12-point Questionnaire
GSSS	Genetic Syndromes Stressors Scale
HADS	Hospital Anxiety and Depression Scale
HRQOL	Health Related Quality of Life
ICD	International Statistical Classification of Diseases and Related Health Problems
K6	Kessler-6 Psychological Distress Battery
MOS	Medical Outcomes Study
ODD	Oppositional Defiant Disorder
PS	Parental Stress
PSS	Parental Stress Scale
PSI	Parenting Stress Index
QRS	Questionnaire of Resources and Stress
SES	Socioeconomic Status
SF-20	Short Form 20-point Patient Health Questionnaire
SPSS	Statistical Product and Service Solutions
TD	Typically Developing
UCC	University College Cork

Abstract

Introduction

At a prevalence of 1 in 700 births, Down Syndrome (DS) is the most common chromosomal birth defect, with Down Syndrome Ireland (DSI) supporting 3,500 people with DS. Having a child with an intellectual disability is associated with increased levels of depression, anxiety, stress, and poorer reported general health. Interventions to support parents should focus on specific factors thought to exacerbate parental stress. To date, no studies were found in the literature examining these parameters in an Irish population.

Objectives

This study aimed to determine the current stress and wellbeing levels of parents of children with DS living in Ireland, to establish predictors for these wellbeing parameters, to identify supports desired by parents for their health and wellbeing, and to make recommendations to DSI regarding the issue of parental wellbeing and where to allocate resources.

Methods

An online self-reported questionnaire (n=226) was created containing the Medical Outcomes Study Short Form (SF-20), the Depression Anxiety Stress Scales (DASS-21), a subset of the Carer Wellbeing and Support Questionnaire (CWS), and demographics. The questionnaire was distributed via DSI by email and through social media sites.

Analysis

All analysis was computed via SPSS 26.0. Total and subcomponent scores were compared to normative data from user manuals via Independent Sample T Tests. Predictive factors were established via Multiple Regression Analysis.

Results

Parents of children with DS scored significantly higher on the DASS-21 Depression ($p<0.001$), Anxiety ($p<0.001$), and Stress ($p<0.001$) subscales. Almost 50% reported depressive symptoms, and almost 17% had severe-extremely severe symptoms. Just over

35% reported anxiety, and 10% had severe anxiety. Stress levels were above normal in 44% and severe in 13%. Physical health scores were not negatively impacted. Employment status and medical status of the parent were the most important negative predictors of Depression, Anxiety, and Physical Health scores. Medical conditions in the parent and young age of the child were the most predictive factors of Stress.

Conclusion

Parents of children with DS experience higher levels of depression, anxiety, and stress than average, but report better physical wellbeing. Unemployed parents experience the highest levels of depression and anxiety. Parents of children aged 0-5 years experience the highest levels of stress. Parents rate respite, speech therapy and psychological support as their top three priorities when seeking support.

Introduction

Down Syndrome

Down Syndrome is a genetic condition characterised by one of three chromosomal abnormalities: full trisomy 21, mosaicism, or translocations. At a prevalence of 1 in 700 births, it is the most common chromosomal birth defect (1), with Down Syndrome Ireland supporting 3,500 individuals with DS and their families (2).

Down Syndrome has a number of physical and intellectual characteristics, with multi-organ system involvement. Congenitally, individuals with Down Syndrome have an increased rate of Heart Disease (3), Hirschsprung's disease (4), and Hypothyroidism (5), among other anomalies mainly affecting the Cardiovascular and Gastrointestinal systems (6). Throughout life, having Down Syndrome increases one's risk of developing Coeliac disease (7), thyroid dysfunction (5), Diabetes Mellitus, haematological abnormalities (8), Juvenile Idiopathic Arthritis (9), Atlantoaxial Instability (10), Obstructive Sleep Apnoea (11), ophthalmic disorders (12), hearing loss (13), Autism Spectrum Disorder (14), and – later in life – Dementia (15).

Regarding intellectual disability, impairments in learning, memory, and language range from mild to profound (16). Down Syndrome is associated with impaired verbal short term memory and explicit long-term memory, as well as weaknesses in expressive language and syntax (17). In addition to this, neurodevelopmental and behavioural disorders such as Attention Deficit Hyperactivity Disorder (ADHD) and Oppositional Defiant Disorder (ODD) are found more frequently in children with DS (18).

Parental Stress

Having a child with an intellectual disability can bring several challenges to a parent and is associated with increased levels of parental stress (19)(20). Parental stress is defined as having two components: a child domain and a parent domain. The former arises directly from the child's own characteristics, the latter from parental functioning. Abidin's Parenting Stress Index, created in 1995, serves to identify high levels of stress in the parent child interaction (21). The former involves qualities of the child "'that make it difficult for parents to fulfil their parenting roles", comprised of six components – distractability/hyperactivity, adaptability, reinforcement of parent, demandingness, mood, and acceptability. The parent domain consists of a further seven components – competence, isolation, attachment, health, role restriction, depression, and spouse. This study's focus is on the latter, specifically investigating physical and mental health levels.

Interest and research in parental stress related to learning disabilities emerged 60 years ago. For the first two and a half decades, researchers adopted a pathology-based approach. The rates of depression (22) and "neurotic-like" constriction (23) were studied, as well as family life factors such as "marital integration" and "sibling tension"(24). By the mid-1980s, research undertook a shift to focus upon the stress and coping mechanisms of parents. Early predictors such as spousal support, perceived control, and child characteristics were established (25). Similar efforts were made to understand the ability of the family to adapt, both in positive and negative ways, to having a child with a learning disability (26).

Physical Wellbeing

Wellbeing is a challenging concept to define given its multi-faceted nature and subjection to cultural practices (27). This study will consider three dimensions of physical wellbeing: Physical Functioning, Pain, and Current Health.

Physical Functioning is described as the extent to which health interferes with a variety of activities (28). Activities assessed include walking, climbing stairs, activities of daily living (e.g. eating, bathing, dressing), moderate activities (e.g. carrying groceries), and vigorous activities (e.g. involvement in sports). It is important to distinguish physical functioning from role restriction, which describes limitation to work or other regular activities such as parenting or caring.

From a medical perspective, pain is defined as "an unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage" (29). In the context of physical wellbeing, pain is explored through its level of severity, and through its effects on behaviour and mood (28). For these purposes, pain is considered to be the severity of "bodily pain" generally experienced by an individual in a certain timespan.

Current Health, lastly, is defined as an individual's overall rating of their current health (28). Again, it is important to distinguish this from Health Distress, which pertains to distress, worry, and discouragement regarding health. In the study of wellbeing, the perception of health is examined through rating one's own health (e.g. agreement or disagreement with the statements “I am somewhat ill”, “My health is excellent”) and comparing one’s health to others (e.g. “I am as healthy as anybody I know”).

Mental Wellbeing

Conventionally defined emotional states of clinical interest include Depression, Anxiety, and Stress.

Depression can be defined as “a state characterised principally by a loss of self-esteem and incentive, and associated with a very low perceived probability of attaining personal life goals of significance to the individual” (30). Anxiety is characterised by long term anticipation “of negative events which typically, but not exclusively, are psychological in character” (30). This is in contrast to fear, which is a response to an immediate threat of harm, particularly physical harm. Stress, lastly, can be conceived as “a persistent state of over-arousal which reflects continuing difficulty in meeting taxing life demands” (30).

These concepts overlap considerably. Research indicates that there is an irreducible minimum correlation of up to 0.5 between self-reported scales that measure depression, anxiety, and stress (31)(32). This may reflect intercorrelation between the concepts themselves, or an overlap of the underlying causes of these affective states. Of note, Anxiety and Depression are often comorbid (33).

Parental Wellbeing in DS and other Developmental Disabilities (DDs)

Comparison of wellbeing levels across parent groups is difficult due to two main factors: heterogeneity of assessment and a lack of a clear control.

Studies have investigated parental wellbeing via general health/mental health questionnaires such as the Medical Outcomes Study Short Form (SF-12) (34), the General Health Questionnaire (GHQ-12) (35), the Hospital Anxiety and Depression Scale (HADS) (36), the Depression, Anxiety, and Stress Scale (DASS-21) (37), or the Kessler-6 Psychological Distress Battery (K6) (38), but also through more specific psychometric tools such as the Questionnaire of Resources and Stress (QRS) (39), the Genetic Syndromes Stressors Scale (GSSS) (40), the Parenting Stress Scale (PSS) (37), and the Parenting Stress Index (PSI) (41)(42). It is notable that the majority of these have concentrated on mental health rather than physical health. From the current literature, it is apparent that parents of children with DS report high levels of physical health (34) and poor levels of mental health (34)(39). 21%

of mothers and 14.3% of fathers with DS children report significant depression and anxiety symptoms on the GHQ-12 (35).

The question of controls has also been ambiguous – many studies do not compare parent groups, but some have compared to parents of children with ASD, other DDs, non-DD conditions such as Type 1 Diabetes, or TD children. Ultimately, we can conclude that the parental burden of DS is above that of TD children (and some non-DD conditions) but below ASD (35)(43) and some other DDs (36)(44).

Predictors of Mental Wellbeing

Linear Regression studies have explored factors that predict PS levels. Results from these are diverse, with predictors coming from demographics, parent characteristics, and child characteristics. Marital status (37), financial stressors (41), parent coping styles (39), and antisocial/disruptive child behaviour (34) have all emerged as the strongest predictors in different studies.

Aims and Objectives

The primary aim of this study is to assess the wellbeing of parents of children with Down Syndrome in the Republic of Ireland.

The objectives of this study are to:

1. Determine the current physical and mental wellbeing levels of parents of children with DS.
2. Establish predictors of parental stress and wellbeing.
3. Identify the supports desired by parents for their health and wellbeing.
4. Make recommendations to Down Syndrome Ireland (DSI) regarding the issue of parental wellbeing and where to allocate resources.

Methods

Study Design

This study was a cross-sectional quantitative self-reported questionnaire, distributed by Down Syndrome Ireland.

The initial planning of this project was completed through a joint meeting involving the research team, representatives from DSI, and a member of CARL. CARL is a community engagement initiative that seeks to link academic researchers with community and voluntary groups. The overall goal of CARL's work is to facilitate research that is desired by these groups by pairing projects with student researchers. The resulting collaboration allows for research by groups that normally would not have the resources to pay for or carry out their own scientific research.

Study Sample

The target population of this study was parents of children with Down Syndrome living in the Republic of Ireland. A total of 226 parents answered the questionnaire. Participants were invited to take part in this study via email from DSI, and via posts on DSI's social media accounts.

Invitation was in the form of a link that brought participants to a Google Form containing the questionnaire. A consent form was provided with the information sheet and questionnaire. Participants were required to read the information sheet and tick a box indicating that they gave their consent in order to complete the questionnaire. After completing the questionnaire, a message appeared thanking the participant for their completion of the survey. It also served to remind them that all data was treated with full anonymity, with no patient identifiers being published. The Google Form accepted answers between November 2020 and March 2021.

No exclusion criteria were applied in this study.

Study Timeline

The total duration of the study was nine months. Ethical approval was granted in July 2020. Data collection began in November 2020 and concluded in March 2021. Data Analysis took place between March 2021 and April 2021.

Instruments

General and Physical Wellbeing was measured by the RAND Organisation's Medical Outcomes Study 20 item questionnaire, the SF-20. Mental wellbeing was measured with the Depression Anxiety Stress Scales 21 item questionnaire (DASS-21) from the University of New South Wales. A subset of the Carer Wellbeing and Support Questionnaire (CWS) from the National Institute for Health Research was added to give qualitative information on desired additional support for carers. Appendix 1 gives a summary and background of each instrument used in this study.

SF-20

The RAND Medical Outcomes Study saw the development of a brief, multidimensional, self-administered, social support survey (45). Originally a 116-item battery, it has since been revised to 36-item, 20-item, and 12-item Short Forms (46). The SF-20 is used to calculate scores on a number of Health Related Quality of Life (HRQOL) concepts, with Physical Functioning, General Health, Pain, Mental Health Index, and Psychological Distress looked at in this study. The SF-20 has high reliability, achieving a Cronbach's Alpha of greater than 0.7 in every scale, with higher values (0.93, 0.86, and 0.83 respectively) for the three scales applied in this study. Reliability is reduced in the SF-20 compared to its longer counterparts, but very modestly so (47). A high SF-20 score indicates good general health.

Sample Questions from the SF-20

Choose one option for each questionnaire item.

Q1. In general, would you say your health is:

- Excellent
- Very good
- Good
- Fair
- Poor

Q4. Does your health keep you from working at a job, doing work around the house, or going to school?

- Yes, for more than 3 months
- Yes, for 3 months or less
- No

Q6. How much of the time, during the past month, has you health limited your social activities (like visiting with friends or close relatives)?

- 1 - All of the time

- 2 - Most of the time
- 3 - A good bit of the time
- 4 - Some of the time
- 5 - A little of the time
- 6 - None of the time

DASS-21

The DASS-21 is comprised of three 7-item subscales – Depression, Anxiety, and Stress – with answers given on a 4-point Likert scale. All subscales are rated from 0 (never) to 3 (almost always). Scores are classified into five ranges: normal, mild, moderate severe, and extremely severe. The intended purpose is to measure the current state of the three emotional dimensions, but not explicitly to aid a DSM or ICD diagnosis (30). However, it has been shown that using a cut-off score of 33 (out of 42) screens for DSM-IV depression and anxiety disorders (Major Depressive Disorder, Dysthymia, Generalised Anxiety Disorder and Panic Disorder) with a sensitivity of 79.1% and a specificity of 77.0% (48). Regarding internal consistency the DASS-21 performs well, with estimates of Cronbach's Alpha ranging from 0.88 (48) to 0.93 (49) The DASS-21 has been shown to have adequate construct validity, with doubling of scores yielding similar values to that of its longer DASS-42 form (49). A high DASS-21 score indicates poor mental health.

Sample Questions from the DASS-21

Please read each statement and select the number 0, 1, 2 or 3 which indicates how much the statement applied to you over the PAST WEEK.

Q3. I couldn't seem to experience any positive feeling at all:

- 0 - Did not apply to me at all
- 1 - Applied to me to some degree, or some of the time
- 2 - Applied to me to a considerable degree, or a good part of time
- 3 - Applied to me very much, or most of the time

Q4. I experienced breathing difficulty (e.g. excessively rapid breathing, breathlessness in the absence of physical exertion):

- 0 - Did not apply to me at all
- 1 - Applied to me to some degree, or some of the time
- 2 - Applied to me to a considerable degree, or a good part of time
- 3 - Applied to me very much, or most of the time

Q12. I found it difficult to relax:

- 0 - Did not apply to me at all
- 1 - Applied to me to some degree, or some of the time
- 2 - Applied to me to a considerable degree, or a good part of time
- 3 - Applied to me very much, or most of the time

CWS

Developed by the National Institute for Health Research, the Carer Wellbeing and Support Questionnaire aims to evaluate a variety of factors related to carer physical and mental wellbeing, as well as their current level of support. The section of interest to this study is Subset C, which involves a qualitative investigation of carers' desired supports through open-ended questions. Overall, the CWS has been shown to have adequate reliability and validity, being a further developed form of the CUES-C (Carers' and Users' Expectations of Services – Carers' version) questionnaire (50).

Sample Questions from the CWS Questionnaire, Subset C

Q1. Would you like more support to help you in your role as a carer?

- No, not at all
- Yes, a little
- Yes, a lot

Q2. What types of additional support would you most like to receive?

____

Q3. Is there anything else that's important to your well-being that you'd like help with or would like to change?

____

Statistical Analyses

Anonymised data from the Google Form was compiled on a Microsoft Excel file on a password-protected computer. All statistical analyses were carried out by SPSS 26.0. Statistical significance was designated at the conventional level of $p < 0.05$. Descriptive statistics were used for demographic data. Missing data was accounted for through pairwise deletion.

Independent T tests were used to compare mean scores of the questionnaire subscales with normative data. A total of eight tests were ran: five subscales from the SF-20 (General Health, Physical Functioning, Pain, Mental Health Index, and Psychological Distress) and three from the DASS-21 (Depression, Anxiety, and Stress).

To establish predictors, ANOVAs were performed to compare total DASS-21 scores across different socio-demographic groups. If a socio-demographic factor had more than two subgroups, Tukey's Post Hoc Multiple Comparisons was further used to compare scores. The impact of these predictors on the variance of both total DASS-21 scores and Depression, Anxiety, and Stress scores was evaluated by Multilinear Regression models.

To assess for the possibility of multicollinearity, Pearson Correlations and Collinearity Statistics were calculated. Significance was designated at the conventional level of >0.7 for Pearson Correlation, <0.1 for Tolerance, and >10 for Variance Inflation Factor (VIF). Distribution of data was plotted on frequency charts, with Kolmogorov-Smirnov and Shapiro-Wilk tests employed to assess for normal distribution. Internal Consistency was assessed using Cronbach's Alpha, which was calculated for each questionnaire subscale.

Study Ethics

Approval was given by the School of Medicine Sub-Committee of the UCC Social Research Ethics Committee, which provides an evaluation of research proposals in alignment with the European Code of Conduct for Research Integrity (2017) and the revised National Policy Statement on Ensuring Research Integrity in Ireland (2019).

Results

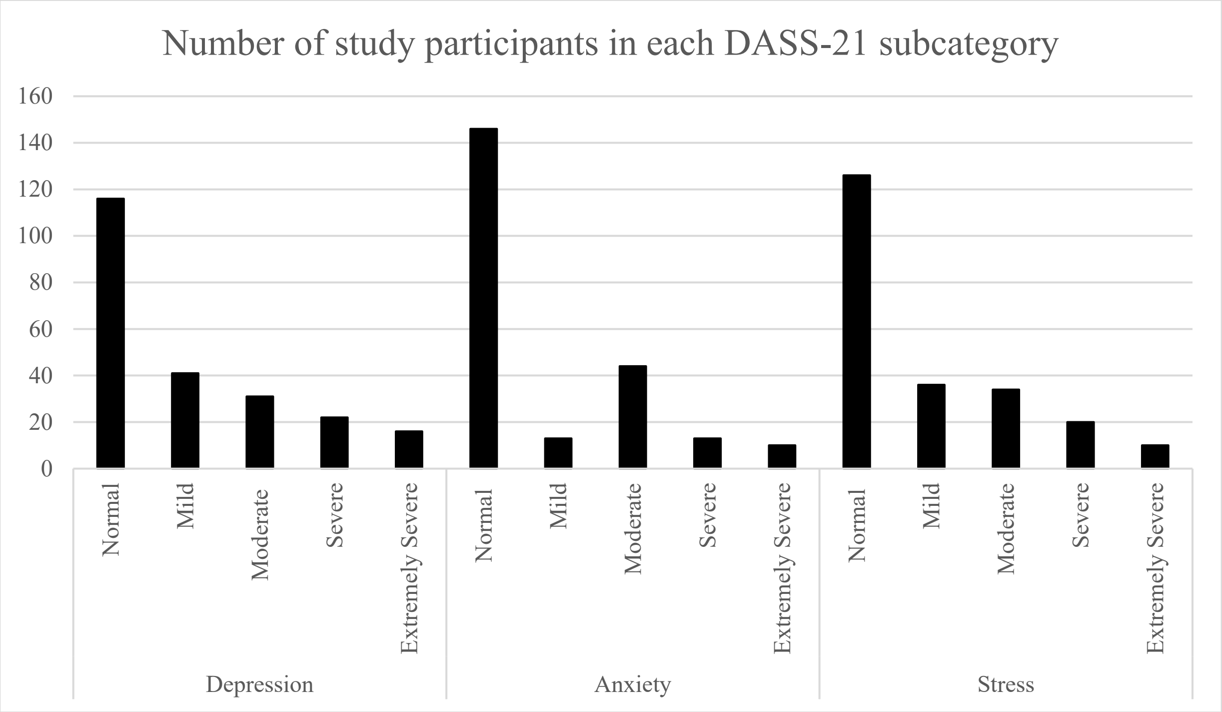
Participants

Appendix 2 shows the socio-demographic characteristics of the 226 parents who took part in this study. 93.4% of participants were female. Just over half (52.2%) were aged 41-50. There was an approximately even distribution between urban, suburban, and rural participants. The majority of parents, at 88.4%, were married. 69.9% had obtained either an undergraduate or postgraduate degree. Just over one quarter (25.8%) had a diagnosed medical condition themselves, with a greater proportion of children with DS having one (60.2%).

Descriptive Data for DASS-21 scores

The mean, standard deviation, minimum, and maximum scores for each DASS-21 subscale were calculated. Following this, the number of participants in each classification (normal, mild, moderate, severe, extremely severe) was totalled, as shown in Table 1. In each subscale, over half of parents were categorised into the "normal" group. The subscale with the greatest number of parents in a moderate to severe category was Depression, at 30.6%. The corresponding numbers for Anxiety and Stress were 29.6% and 28.3% respectively.

Table 1: Descriptive Statistics of DASS-21 subscales



Comparison to Normative Data

Mean scores were calculated for all subscales on both the SF-20 and DASS-21 and compared to normative data from the user manuals. Table 2 summarises the eight Independent T Tests performed in this study. Regarding the SF-20, the study sample's mean scores for General Health ($p<0.001$), Physical Functioning ($p<0.001$), and Pain ($p<0.001$) are all significantly higher than that of the normative sample. In the mental health battery of the SF-20, the study sample's mean is significantly lower than that of normative data in the case of both the Mental Health Index ($p<0.001$) and Psychological Distress ($p<0.001$). Similar findings came from the DASS-21, where the study participants had a higher mean score on all three of the Depression ($p<0.001$), Anxiety ($p<0.001$), and Stress ($p<0.001$) subscales.

Table 2: Independent T-tests for group comparisons of study variables

Subscale	Sample	µ	SE Difference	Mean Difference	Sig. (2-tailed)
General Health (SF-20)	Study	67.00	1.79	12.20	**<0.001*****
	Normative	54.80			
Physical Functioning (SF-20)	Study	86.71	1.57	15.80	**<0.001*****
	Normative	70.90			
Pain (SF-20)	Study	80.77	1.59	9.97	**<0.001*****
	Normative	70.80			
Mental Health Index (SF-20)	Study	61.77	1.39	-10.93	**<0.001*****
	Normative	72.70			
Psychological Distress (SF-20)	Study	71.68	1.53	-5.62	**<0.001*****
	Normative	77.30			
Depression (DASS-21)	Study	10.81	0.66	4.47	**<0.001*****
	Normative	6.34			
Anxiety (DASS-21)	Study	6.10	0.46	1.40	**<0.001*****
	Normative	4.70			
Stress (DASS-21)	Study	14.85	0.61	4.74	**<0.001*****
	Normative	10.11			

$^{*}p \leq 0.05$ $^{**}p \leq 0.01$ $^{***}p \leq 0.001$

Predictive Factors

The relationship of socio-demographic factors with the study variables was initially investigated via ANOVA, comparing the mean scores across demographic subgroups. This analysis (Table 3) yielded a statistically significant difference across employment status subgroups, as well as across parental medical status. Further analysis with Tukey's Post Hoc Multiple Comparison showed that unemployed parents scored higher on all three DASS-21 subscales than parents in either full or part time employment, with a significant difference occurring in almost all subgroup comparisons (Table 4). The greatest difference was seen on the Depression subscale.

Table 3: ANOVAs comparing DASS-21 scores across socio-demographic groups

Factor	Statistic	Depression	Anxiety	Stress
Age of Child	F	1.436	1.274	2.206
	sig.	0.223	0.281	0.069
Age of Parent	F	0.319	0.09	1.516
	sig.	0.865	0.985	0.199
Education	F	1.476	2.276	1.021
	sig.	0.211	0.062	0.398
Employment Status	F	8.939	6.42	3.783
	sig.	**<0.001*****	**0.002****	**0.024***
Gender	F	0.148	1.314	0.76
	sig.	0.7	0.253	0.384
Marital Status	F	1.423	0.717	0.32
	sig.	0.197	0.658	0.944
Medical Status of Child	F	0.56	2.519	0.463
	sig.	0.642	0.059	0.709
Medical Status of Parent	F	5.937	16.291	10.392
	sig.	**0.016***	**<0.001*****	**0.001*****
Number of Children	F	1.315	0.544	0.882
	sig.	0.265	0.704	0.476
Position of Child in Family	F	0.56	2.519	0.463
	sig.	0.642	0.059	0.709
Residence	F	3.143	2.099	1.969
	sig.	**0.045***	0.125	0.142

$^{*}p \le 0.05$ $^{**}p \le 0.01$ $^{***}p \le 0.001$

Table 4: Tukey's Post Hoc Multiple Comparisons of DASS-21 scores by employment status

Dependent Variable	Employment Status	Comparison Group	Mean Difference	Sig.
Depression	Unemployed	In full time employment	4.71181	**0.010****
		In part time employment	6.17581	**<0.0001*****
Anxiety	Unemployed	In full time employment	3.63542	**0.004****
		In part time employment	3.04958	**0.012***
Stress	Unemployed	In full time employment	2.41667	0.241
		In part time employment	3.87553	**0.019***

$^{*}p \leq 0.05$ $^{**}p \leq 0.01$ $^{***}p \leq 0.001$

Predictive factors for both total and subscale DASS-21 scores were further evaluated via Multilinear Regression. The analysis for total score (Table 5) showed that medical status of the parent, age of child with DS, and employment status are the three socio-demographic factors that account for the most variation in score. Model 1, including just medical status of the parent, accounts for 6.5% of the variation in total score. Models 2 and 3 explain 9.4% and 12.0% of the variance respectively.

Table 5: Stepwise Regression of total DASS-21 score and socio-demographic factors

Model Summary[d]

Model	R	R Square	Adjusted R Square	Std. Error of the Estimate
1	.255[a]	.065	.060	21.40458
2	.307[b]	.094	.085	21.11801
3	.347[c]	.120	.107	20.86152

a. Predictors: (Constant), Medical status of parent

b. Predictors: (Constant), Medical status of parent, Age of child

c. Predictors: (Constant), Medical status of parent, Age of child, Employment status

d. Dependent Variable: Total DASS-21 Score

ANOVA[a]

	Model	**Sum of Squares**	**Df**	**Mean Square**	**F**	**Sig.**
1	Regression	6570.668	1	6570.668	14.342	**.000**[b]***
	Residual	94838.327	207	458.156		
	Total	101408.995	208			
2	Regression	9539.116	2	4769.558	10.695	**.000**[c]***
	Residual	91869.879	206	445.970		
	Total	101408.995	208			
3	Regression	12192.360	3	4064.120	9.338	**.000**[d]***
	Residual	89216.635	205	435.203		
	Total	101408.995	208			

a. Dependent Variable: Total DASS-21 Score

b. Predictors: (Constant), Medical status of parent

c. Predictors: (Constant), Medical status of parent, Age of child

d. Predictors: (Constant), Medical status of parent, Age of child, Employment status

$^{*}p \leq 0.05$ $^{**}p \leq 0.01$ $^{***}p \leq 0.001$

When repeating this analysis with different subscales as the dependent variable, a similar relationship was found for both the Anxiety and Stress scores, but not for Depression. Again, medical status of the parent was the greatest predictor of Anxiety and Stress, accounting for 7.9% and 5.6% of the variance respectively. Differing from this, the regression model for Depression scores revealed employment status to have the greatest predictive value. However, this model was less predictive, with employment status accounting for only 3.4% of the variance.

Multicollinearity

To assess for potential confounding, multicollinearity statistics were employed. Appendix 3 details the Pearson Correlations of the predictive socio-demographic factors established from previous Linear Regression. No two factors had a correlation >0.7. Furthermore, When Collinearity Diagnostics were performed on DASS-21 subscale scores, no Tolerance was found to be <0.1, and no VIF was found to be >10. Therefore, no significant degree of collinearity is present between the predictors.

Distribution, Internal Consistency

Appendix 4 shows the frequency plots of DASS-21 subscale scores and statistical assessment for normal distribution. With all six Kolmogorov-Smirnov and Shapiro-Wilk tests being significant ($p < 0.01$), it can be concluded that this data does not follow a normal distribution curve.

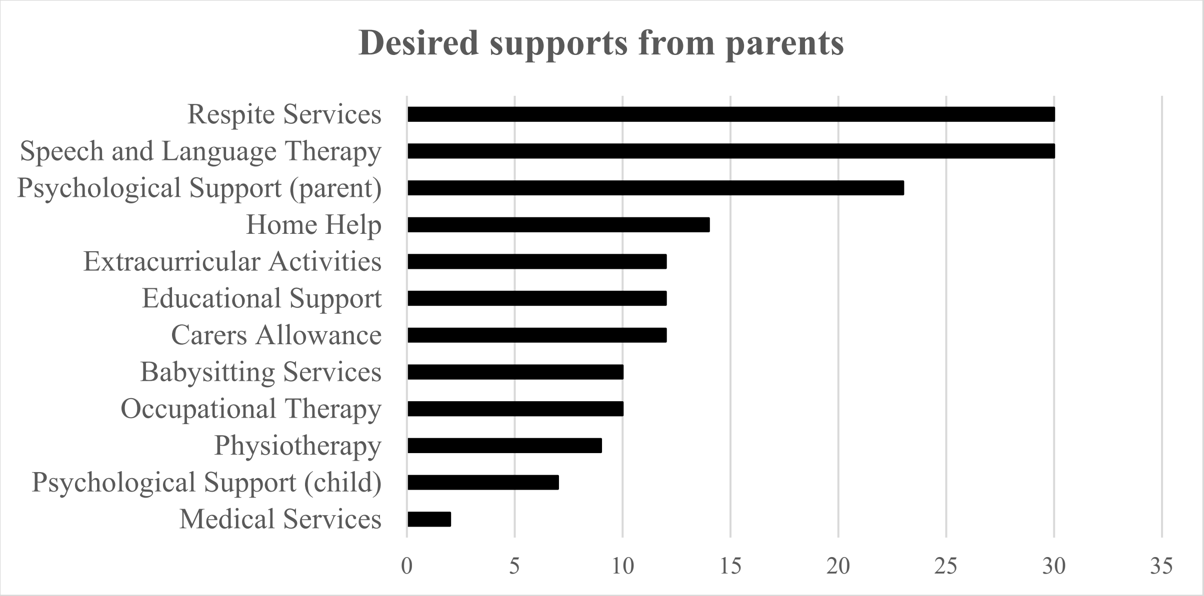
Cronbach's Alpha scores were high across all subscales: 0.86 for General Health, 0.93 for Physical Functioning, 0.83 for Pain, 0.89 for Mental Health Index, 0.85 for Psychological Distress, 0.81 for Depression, 0.73 for Anxiety, and 0.81 for Stress.

Desired Support from Parents

All 226 participants answered the first question of the CWS subset of the questionnaire, "Would you like more support to help you in your role as a carer?". 85% reported yes, with 65% of these answering "yes, a little", and 35% answering "yes, a lot".

171 parents answered the following question, "What types of additional support would you most like to receive?", giving a response rate of 76%. The results from Content Analysis of their answers are summarised in Table 6. The three supports most desired by parents are respite care, speech and language therapy services, and psychological support for parents. Other desired supports include help at home, educational support, and other allied healthcare supports including occupational therapy and physiotherapy.

Table 6: Results from CWS-C



Parental Views of Supports and Services

Many parents voiced a need for additional services, particularly speech and language therapy. The irregularity and infrequency of services was noted by several parents, with one describing how they “would like to receive the therapies that my son is entitled to such as speech and language, only 2 sessions in 3 years”. The shortage of services had a clearly described impact on parental wellbeing, with one participant explaining: ”as a parent with a child with additional needs you are very much left on your own and you have to be the speech therapist, occupational therapist, etc and sometimes you can feel that you are failing your child as you don’t have the tools to help them and guide them”.

For some parents, there was considerable overlap between help at home and their own mental wellbeing. One parent spoke about the need for “someone to come to house to entertain, play, interact in a meaningful way with my child with Down syndrome for a time (a couple of hours a week split up into different days) so that I could spend some quality time with my 3 other kids doing homework, games or just chatting that can be difficult to find time for otherwise”. This sentiment was echoed for respite care, which another parent saying: “as my son grows up he needs more interaction with people, he does not always want to be with his mum. He needs other outlets to meet people, but they are not available. A buddy system to link in with another family or adult or a respite day or night would be very beneficial for both him and me”.

Psychological support for parents was raised by 23 participants. The first years of raising their child – described as a “steep learning curve” – were particularly noted as stressful: “(I) never got help with mental health especially when I needed it the most, when the kids were younger. (It) sounds like there is a lot more awareness now... I wish I had therapy back then.” Typically, counselling services were cited by parents as viable supports for parents. However, some specifically mentioned more interaction with other parents of children with DS, and pointed to parent support groups. It would appear that several parents were already involved in self-organised groups, but others have struggled: “I have reached out to other parents of kids with DS when I have been experiencing challenges with school, expectations, behaviour or slumps myself. But these parents are busy and sometimes are not always able to say it… as they don’t want to be judged”.

Summary of Key Findings

1. 30.6% of parents received a moderate to severe score on the Depression DASS-21 subscale.
2. Parents of children with DS scored significantly higher than normative data controls on all three DASS-21 subscales ($p < 0.001$ for all).
3. Parents of children with DS scored significantly higher on all physical health SF-20 scales, and lower on mental health scales ($p < 0.001$ for all).
4. Employment status and medical status of the parent were the most predictive factors of Depression, Anxiety, and Physical Health scores.
5. 85% of parents report a need for more support in their role as a carer.
6. The two supports most desired by parents are respite care and speech and language therapy services.

Discussion

Physical Wellbeing

The results of this study demonstrate that, compared to normative data controls, parents of children with DS report better physical health. This was seen across all three subscales of physical wellbeing – Physical Functioning, Pain, and General Health – and was highly significant ($p < 0.001$). Though physical health has been studied in this population comparatively less than mental health, these findings are in line with previous studies (34). However, a brief report from 2020 (51) indicates that parents of children with DS are more likely to be inactive than parents of TD children or children with DD. It is possible that selection bias in this study may have led to an overestimation of physical health.

Mental Wellbeing

From both the SF-20 and DASS-21 findings, this study strongly points to increased mental health symptoms in the sample population. Parents scored higher than normative data controls on all three of the Depression, Anxiety, and Stress subscales ($p < 0.001$). This adds to the existing literature which shows poorer mental health in both parents of children with DS (34)(35)(39) and children with other DDs (38)(52).

Predictors of Mental Wellbeing

This study sought to establish predictors of mental wellbeing. Predictors of physical health were not explored given the higher performance of the sample over controls. In the ANOVA analysis, employment status and medical status of the parent emerged as factors with significant differences in their subgroups. In further Multilinear Regression of total DASS-21 scores, parental medical status emerged as the most predictive factor, followed by child's age and employment status. No other parent factors (parent's age, parent's gender, level of education, marital status, residence) or child factors (medical status of child, position of child with DS in family, number of children in family) had a significant relationship in either analysis.

The role of parental medical status has not been observed by other studies, but likely speaks of a larger relationship between physical and mental health (53). The impact of employment status has not explicitly been investigated by other researchers, but the financial strain (34) and time commitments (35) of raising a child with DS are well documented. The results from this study echo those of another which found family financial hardship and lower parent empowerment to be predictors of parental distress in parents of children with DD (41). It is also relevant to note that this study did not distinguish unemployed (and job seeking) from parents that cannot work due to carer commitments. As these two groups face distinct challenges and stressors, this may have obscured causal relationships.

Lastly, numerous studies have demonstrated the influence of child factors on parental wellbeing, such as behavioural issues (34)(37)(42), milestone development (40), and global ability (36). This is particularly relevant given the higher prevalence of ASD, ADHD, and ODD in children with DS (18) and the association of these conditions with higher levels of parental stress (54)(55). By contrast, this study indicated comparatively little impact of child factors. A potential explanation for this is the limited way in which child behaviour or ability was assessed, whereas other studies with positive findings have investigated these factors with full questionnaires – such as the Developmental Behaviour Checklist (34) or Children's Global Assessment Scale (CGAS) (35).

Intervention

One of the more important reasons for this study is to point towards future intervention to reduce parental distress.

The most desired supports by parents are increased respite care and speech and language therapy. Services in Ireland are heavily limited, with recent evidence showing that only 44% of parents were offered public speech and language therapy in 2019 (56). In this study, out of the parents who listed speech and language therapy as a desired support, 60% of them had a child aged 6-12. DSI's See and Learn programme provides an excellent, evidence-based, and structured teaching programme for children up to age 6 (57)(58). Campaigning for allocation of resources to children aged over 6 would address this need identified by parents.

From this study, employment status has emerged as a predictor of parental distress. Though further research is needed to distinguish financial burden from work related stress, one issue cited by parents in the CWS questionnaire is Carer's Leave. Carer's Leave allows a carer to temporarily take time off work to provide full-time care. Current Irish legislature requires Carer's Leave to last for a minimum of 13 weeks and a maximum of 104 (59). If leave is less than 13 weeks, employers have the right to refuse the carer's request. Having this requirement may restrict leave from parents whose children require short-term full-time care, such as illness or medical procedures, school holidays, or transitioning between schools. Carer's Benefit – a social insurance payment to those who leave work for full-time care – is another option open to parents of children with DS. Though it has no minimum time requirement, Carer's Benefit has PRSI contribution conditions, thus restricting it from some carers. Campaigning for changes in the requirements of these allowances may allow more parents to take leave from work.

Further Research

Significant differences have emerged from this study when looking across employment status, with unemployed parents reporting more mental health symptoms than fully or partly employed parents. Two potential explanations for this result are the impact of financial strain on this parent group and the inability to work due to carer duties. Both theories are plausible, given that there is evidence for both: 50% of parents of children with DS experience financial difficulty (34), and have a low proportion of both parents in full time employment, at 1 in 4 (35). Further research could explore the impact of financial strain on parents and specify the added financial costs of raising a child with DS.

Child's age had a relationship with parental mental health in certain contexts. Stress scores decreased with increasing age, but anxiety scores peaked in the 6-12 age group. These results could be further investigated to identify specific stressors related to these age groups.

Limitations

Firstly looking at this study's design, there is a question of what parameters should be investigated. Given the multifaceted nature of wellbeing, a study should ideally compare the impact of as many aspects of life as possible. Research into predictive factors been through the lens of biology (physical health (34)), psychology (coping styles (37), family resources (41), child factors (35)), and sociology (financial strain (38), societal stigma (60)), proving the wide range of potential influences. This study primarily focussed on parent factors, with relatively little attention to child factors, and hence does not give a holistic picture of parental wellbeing.

From this study's methods, Volunteer Bias is a potential threat to external validity. Participation in this research was voluntary, without any incentives. Volunteer Bias refers to a specific bias whereby volunteers are different in some way from the general population

(61). Furthermore, the recruitment of these volunteers through email and online media platforms could exclude subgroups of the target population that either cannot access or struggle to use the internet – a phenomenon named the “Digital Divide” (62).

Regarding statistical analysis, the main limitation of this study is the use of short form versions of questionnaires. Research has shown that the validity and reliability of these scales is lower than that of their longer counterparts, but modestly so (49)(63). Furthermore, there is the issue of the validity of normative data. These datasets often come from different populations, cultures, and periods of time than the population of interest – for example, the SF-20 and DASS-21 were both developed in the mid-1990s in the USA and Australia respectively, and thus their normative data is from these populations. If use of these questionnaires is to be optimal, then the sample should be compared to a more similar population group (64). One final consideration for analysis is the distribution of data. Parametric tests assume that the variable of interest has a normal distribution in the population. From our study sample, it is evident that Depression, Anxiety, and Stress scores were not normally distributed (Appendix 4).

Lastly, there is potential influence on mental wellbeing from current world events. The changes brought about by the COVID-19 pandemic have had a major impact on emotional wellbeing. Early cross-sectional research at the beginning of the pandemic demonstrated a high prevalence of psychological distress (65)(66)(67), reinforced by systematic review (68). However, further longitudinal research yielded mixed results when comparing different points since the onset of COVID-19 (69)(70). A very recent meta-analysis and systematic review of both pre- and mid-pandemic longitudinal cohort studies (the only one of its kind at its time of publishing) demonstrated a small but significant increase in mental health symptoms early in the pandemic (71). Mental health symptoms returned to pre-pandemic baseline by mid-2020, at the time when this study began.

Conclusion

In conclusion, this study confirms that parents of children with DS experience higher levels of depression, anxiety, and stress than controls.

Several aspects of both physical and mental wellbeing were examined, with significant differences found on all parameters. Reported levels of physical health were higher in this sample, whereas those for mental health were lower. Subgroup analysis showed differences across employment status and medical status of the parent.

Identifying and understanding factors related to parental distress is vital for the planning of targeted interventions, as is eliciting desired supports from parents themselves. This study identifies the parents most in need of support are those with young children (<5 years), those with underlying medical conditions, and those who do not work outside the home. Most parents identify speech and language therapy as an unmet need for their children but respite and psychological support for themselves.

Future work in this area should centre around intervention-based study. Unlike other DDs, DS has had no observable trials to improve parental wellbeing (72)(73)(74). The development of interventions specifically targeted at predictors of distress has the potential to improve the lives of both parents and children.

Student Declaration

The research topic of this study was jointly decided by the student, academic supervisor, and representatives from CARL and DSI. Student contributions to this research project included designing of the survey (including choice of validated questionnaires), sourcing of user manuals, liaising with DSI to distribute the questionnaire, data storage and analysis, writing of the report, and presentation of findings.

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Conflict of Interest

The authors declare that they have no conflict of interest.

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Appendices

Appendix 1

Description of instruments used in this study

Instrument	Developer	Number of items	Description	Score range	Cohort mean (SD) and range
SF-20	R. Hays, C. Sherbourne et al (*RAND Organisation, USA, 1995*)(28)	20	Widely used measure of general health status, including physical functioning, pain, health, vitality, social functioning, and mental health. A higher score indicates better general health.	0-100 (for each HRQOL parameter)	General Health 67 (23.89): 0-100 Physical Functioning 86.71 (21.85): 0-100 Pain 80.77 (22.27): 10-100
DASS-21	S.H. Lovibond, P.F. Lovibond (*University of New South Wales, Australia, 1995*)(30)	21	Three scales designed to measure the negative emotional states of depression, anxiety, and stress. Assesses the severity of the *core* symptoms of each. For use by researchers and clinicians. A higher score indicates a greater degree of experienced depression, anxiety, or stress.	0-42 (for each of the three scales)	Depression 10.81 (9.53): 0-42 Anxiety 6.1 (6.61): 0-36 Stress 14.85 (8.82): 0-42
CWS (Section C subset)	A. Quirk, S. Smith et al (*National Institute for Health Research, UK, 2012*)(50)	3	Subset of the Carer Wellbeing and Support questionnaire, used to assess supports needed by carer. Enquires about degree of help needed and openly asks about desired additional supports.	N/A qualitative	N/A qualitative

Appendix 2

Socio-demographic characteristics

Gender of parent (n, %)	
Female	211 (93.4%)
Male	15 (6.6%)
Age of parent (n, %)	
18-30	2 (0.9%)
31-40	5 (15.5%)
41-50	118 (52.2%)
51-60	66 (29.2%)
61-70	5 (2.2%)
>70	0 (0.0%)
Residence (n, %)^x	
Urban	49 (21.8%)
Suburban	77 (34.2%)
Rural	99 (44.0%)
Current Employment (n, %)^x	
Full Time Employment	67 (30.3%)
Part Time Employment	80 (36.2%)
Unemployed/Carer	74 (33.5%)
Highest Level of Completed Formal Education (n, %)	
Primary School	1 (0.4%)
Secondary School	22 (9.7%)
Post Leaving Cert Course (PLC)	45 (19.9%)
Undergraduate Degree	64 (28.3%)
Postgraduate Degree	94 (41.6%)
Number of Children (n, %)	
1	18 (8.0%)
2	80 (35.4%)
3	91 (40.3%)
4	27 (11.9%)
5	9 (4.0%)
>5	1 (0.4%)
Marital Status (n, %)^x	
Married	198 (88.4%)
Divorced	7 (3.1%)
Other (single, widowed, cohabiting, civil partnership)	19 (8.5%)
Age of child (n, %)	
0-5	49 (21.7%)
6-12	103 (45.6%)
13-18	47 (20.8%)
18-35	26 (11.5%)
>35	1 (0.4%)
Position of child with DS in family (n, %)	
First born	72 (31.9%)
Second born	35 (35.0%)
Third born	49 (21.7%)
Fourth or subsequent born	26 (11.5%)
Medical status of child (n, %)	
Has diagnosed medical condition	136 (60.2%)
Does not have diagnosed medical condition	90 (39.8%)
Medical status of parent (n, %)^x	
Has diagnosed medical condition	58 (25.8%)
Does not have diagnosed medical condition	167 (74.2%)

^{x} Numbers do not add up to total number of participants (N = 226) due to missing values

Appendix 3

Multicollinearity diagnostics

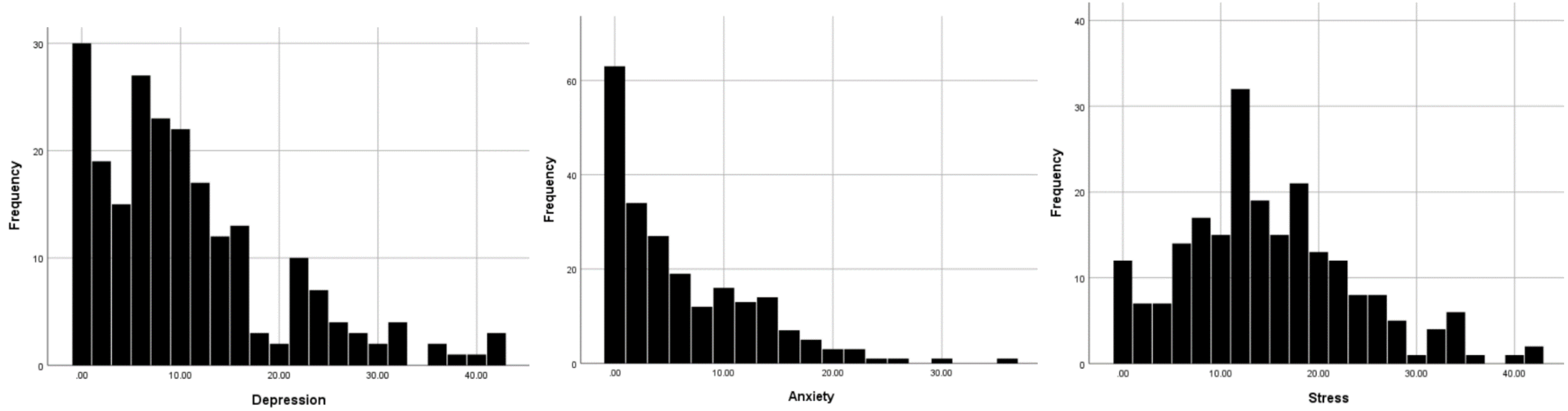
		In full time employment	**In part time employment**	**Unemployed**	**Parent with Medical Dx**	**Parent without Medical Dx**
Pearson Correlation	**Child 0-5**	0.111	0.048	-0.156	-0.027	0.027
	Child 6-12	-0.050	0.118	-0.072	-0.068	0.068
	Child 13-18	-0.042	-0.118	0.161	-0.033	0.033
	Child 19-35	-0.011	-0.097	0.110	0.181	-0.181
	Child >35					
	In full time employment	1.000	-0.494	-0.460	-0.105	0.105
	In part time employment	-0.494	1.000	-0.545	-0.103	0.103
	Unemployed	-0.460	-0.545	1.000	0.206	-0.206

Collinearity Diagnostics; Anxiety		
	Tolerance	**VIF**
Child 6-12	0.992	1.008
Unemployed	0.954	1.048
Parent with Medical Dx	0.955	1.047

Collinearity Diagnostics; Stress		
	Tolerance	**VIF**
Child 0-5	0.976	1.025
Unemployed	0.935	1.07
Parent with Medical Dx	0.958	1.044

Appendix 4

Frequency plots of DASS-21 scores



Statistical assessment of normal distribution

	Kolmogorov-Smirnov			**Shapiro-Wilk**		
	Statistic	df	Sig.	Statistic	df	Sig.
Depression	0.152	220	**<0.001*****	0.888	220	**<0.001*****
Anxiety	0.188	220	**<0.001*****	0.847	220	**<0.001*****
Stress	0.099	220	**<0.001*****	0.967	220	**<0.001*****

$^{*}p \leq 0.05$ $^{**}p \leq 0.01$ $^{***}p \leq 0.001$

STROBE statement for cross-sectional studies

	Item No	Recommendation	Page No
Title and abstract	1	(*a*) Indicate the study's design with a commonly used term in the title or the abstract	4
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found	5
Introduction			
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	6
Objectives	3	State specific objectives, including any prespecified hypotheses	10
Methods			
Study design	4	Present key elements of study design early in the paper	11
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	11
Participants	6	(*a*) Give the eligibility criteria, and the sources and methods of selection of participants	11
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	6-8
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	12-13
Bias	9	Describe any efforts to address potential sources of bias	N/A
Study size	10	Explain how the study size was arrived at	11
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why	14-15
Statistical methods	12	(*a*) Describe all statistical methods, including those used to control for confounding	14-15
		(*b*) Describe any methods used to examine subgroups and interactions	14-15
		(*c*) Explain how missing data were addressed	14-15
		(*d*) If applicable, describe analytical methods taking account of sampling strategy	N/A
		(*e*) Describe any sensitivity analyses	N/A

Results			
Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	N/A
		(b) Give reasons for non-participation at each stage	N/A
		(c) Consider use of a flow diagram	N/A
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	36
		(b) Indicate number of participants with missing data for each variable of interest	36
Outcome data	15*	Report numbers of outcome events or summary measures	
Main results	16	(*a*) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	16-18
		(*b*) Report category boundaries when continuous variables were categorized	16-18
		(*c*) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	N/A
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	19-20
Discussion			
Key results	18	Summarise key results with reference to study objectives	23
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias	26-27
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	28
Generalisability	21	Discuss the generalisability (external validity) of the study results	28
Other information			
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	N/A