
Parental Stress, Familial Burden and Quality of Life in Parents of Children with Down Syndrome

Areej Mahmood¹, Afsheen Gul^{2*}**Abstract**

The current study was conducted to explore the severity of parental stress, familial burden and how it effects on quality of life of those parents having children with Down Syndrome in Pakistan. Correlational research design was used to recruit the sample. The participants $N=110$ ($M=55$; $F=55$) were recruited through purposive sampling from different institutes of special education and outpatient departments of psychiatry hospitals. Those who completed the inclusion criteria participated in the study by signing written informed consent. Parental Stress Scale (Berry & Jones, 1995) was used to determine the stress level. Burden Scale for Family Caregivers (Grau et al., 2014) was used to analyze the burden severity, and Family Quality of Life Scale (Hoffman et al., 2006) was used to determine their effected life due to having children with Down Syndrome. The results found a negative relationship between parental stress ($r=-.20$, $p<.05$), and familial burden against quality of life among parents having children with Down syndrome. Also, there is significant positive relationship among parental stress and familial burden ($r=.37$, $p<.001$), that shows the parental stress increases with the increase in familial burden. However, independent samples t test showed fathers to have high stress and burden as compared to mothers. The study has implications in the field of clinical psychology. The results will be able to help practitioners to consider the proliferating factors of stress and burden among the parents of children having Down Syndrome.

Keywords: Burden, Down Syndrome, Family Caregivers, Parental Stress, Quality of Life

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Introduction

Down Syndrome known as a chromosomal disorder usually result because of the existence of an extra or duplicate chromosome (Bittles et al., 2007). The prevalent of Down Syndrome is high among all inhabitants across the globe (Fuca et

al.,2022). The center for Disease Control Prevention (CDCP), described that the Down Syndrome (DS) is the most common chromosomal disorder, with occurrence of 1 in 700 births (CDCP, 2020) in High Income Countries (HICs), although the data regarding the incidence and prevalence of this disorder among Low and Middle Income Countries (LMICs) are limited. A study conducted in Lahore, Pakistan, indicates that one among 300 babies diagnosed with Down Syndrome (Siddiqui et al., 2021), although highest number of children with Down Syndrome prevailing in the rural areas of Pakistan (Rahman & Obaid-ur-Rahman., 2005),

Different researches suggested that the parental stress in not being deleterious towards parents but also effects the children and overall functioning of the family, which

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eventually increases the burden towards the family (Daundasekara et al., 2021). A systematic review indicates that, quality of life significantly affected due to parental stress either father or mother which can be determined as a reduction towards the satisfaction of life (Martin et al., 2019). It has been showed that the parental stress became the cause of less stimulation of parents with their children (Miragoli et al., 2018). A study based on cross disability approach indicated that the emotion manifestation, particularly the countenance of high criticism was associated with the parents who have children with Down Syndrome, ultimately effects negatively and increased burden of family (De Clercq et al., 2021).

In developed countries, life expectancy and parental stress had a progressive impact both on individual or family. On the other hand, in developing countries due to lack of resources and other factors, individuals with Down Syndrome and their families are felt burdensome and associated with the feeling of stigmatization (Glasson et al., 2002). In a longitudinal study, it has been revealed that economically and culturally diverse variates in HICs and LMICs (Bryant et al. 2011; Hsiao & Riper, 2009; Selway & Ashman, 1998). Studies conducted in different Islamic countries also showed that although DS was considered due to the GOD's will, but a lot of stress, familial stigma and societal burden were experienced by the parents (Bryant et al., 2011; Latzer et al., 2021). Conversely, there is almost no research was conducted to explore about parental perspective in Pakistan. Thus, the aim of the present study is to explore the level of parental stress and familial burden, how it effects the quality of life of those parents having kids with Down Syndrome in Pakistan. This study also provided suggestions to develop support group for practitioners, physicians and mental health professionals based on identified needs from

the parents and caregivers that might help them to increase the tendency of the parents to help their child grow well in the society and keep pace with the world by availing their current abilities. Thus, the study was beneficial for the parents to get support in the betterment of their children's psychological and physical health, by interacting with other parents whose children were suffering from such disability.

Rationale

Family life depends on the health of each and every family member. Literature suggested that parents of those children who have any intellectual and developmental delays are high to prone to suffer in stress and emotional disturbance which directly or indirectly affect the marital life as well. Still, the current study aimed to explore the level of parental stress and familial burden, how it effects the quality of life of those parents having children with Down Syndrome in Pakistan.

Hypotheses

- There would likely to be a significant relationship between parental stress, familial burden and family quality of life among parents of children with Down Syndrome.
- Parental stress and Familial Burden would likely to predict family quality of life among parents of children with Down's Syndrome.
- There would likely to be a significant difference between father and mother of the child in scores of parental stress, familial burden and family quality of life.

Method

Research Design

The correlational research design was used to explore the level of parental stress, familial burden and how it effects the quality of life of Down Syndrome parents in Pakistan

Sampling Strategy & Sample

The Correlational research study was conducted through purposive sampling by selecting those parents having children with Down Syndrome in Lahore, Pakistan

($N=110$) aged between 1 year to 10 years ($M = 6.59$, $SD = 1.88$). Total 110 participants participated in the research in which fifty-five ($N=55$) were fathers and fifty-five ($N=55$) were mothers as the formal sample size recommendations by Andy Field were $N=100$ (Field, 2017) for good practice. The sample was recruited by administering the checklist of inclusion criteria and after completion of the written and consent both from parents and institute.

Inclusion Criteria

- Both father and mother having diagnosed child with Down's Syndrome.
- Parents whose child with Down Syndrome lies within the age range of birth to 10 years.
- Parents having at least primary level of education.

Exclusion Criteria

- Both father and mother having child diagnosed with any comorbid disorder along with Down's Syndrome.
- Parents having Child with Down Syndrome with age above 10 years.
- Parents having less than primary level of education.

Research Tools

The demographic form was filled by the researcher for those who fulfil the inclusion criteria or given consent to participate in the study. The demographic form includes; age, socioeconomic status, mother's occupation, family system, age of diagnosis, reaction of family upon diagnosis, changing reaction, future concerns (inclusive schooling, Mainstream Education, Vocational training) was asked by parents before starting the interview.

Different psychological assessment form was completed from parents by researchers these were;

a) Parental Stress Scale (PSS)

The Parental Stress Scale was used to measure the stress level of parents with Down Syndrome. It is 18-item five point Likert scale scoring from strongly disagree to

strongly agree in order to score the level of stress among parents. The 8 items of the scale were reverse scored. The maximum possible scores for this scale ranged from 18 to 90. The scale was administered on both fathers and mothers of the children. The scale is relatively short and easy to administer which is completed in less than 10 minutes. The instrument internal reliability was .83, whereas the test retest reliability was .81 (Berry & Jones, 1995).

b) The Burden Scale for Family Caregivers (BSFC)

Burden Scale for Family Caregivers was used to measure the burden among the caregivers or parents who are having the children with Down Syndrome. This is 10-item scale consisted of 4 point Likert scale scoring from strongly disagree to strongly agree ranging from 0 to 3. The maximum score of the BSFC is from 27 to 30. The higher score indicates the high burden of the caregivers and parents. The internal reliability of the instrument is .90 whereas, the test retest reliability was .9 (Grau et al., 2014).

c) Family Quality of Life Scale (FQOL)

The instrument was used to measure the quality of life of the families who have any illness or problem in any of the family member. It is 25-item consisted of 5 point Likert scale on the basis of their satisfaction level from life stretching from "very dissatisfied" till "satisfied". The maximum score of the quality of life among family is up to 100. The more the participant got score, the more satisfaction he or she have about the quality of life. The reliability of the Cronbach's alpha was .94 whereas, the inter-rater reliability was .77 (Hoffman et al., 2006).

Ethical Considerations

To start the study, initial step was to get approval from the ethical board of the university. The research proposal was presented, and some changes were recommended by the board of ethics. After

making appropriate changes, finally the permission was granted by the university board to conduct the research. The second step was to approach the authors of all instruments which were planned to use during the research. An official email was sent with the objective of the study to respective authors of instruments for the permission to use their scales. After getting written permission from the authors, the work was started. At the third level, permission was obtained from different special education schools and psychiatry out patient departments of hospitals to obtain data. The information leaflet had been designed for the participants in hard form so they could understand the objectives of the study. The information leaflet was written in both English and Urdu languages for their understanding. Written consent was obtained before starting the study. It had been

informed to the participants that confidentiality would be maintained during the research.

Results

The current study was conducted to explore the level of parental stress, familial burden, and how it effects the quality of life of Down Syndrome parents living in Pakistan. Descriptive and inferential statistics were used to analyze the data accordingly including; to determine the reliability the Cronbach's alpha of the scales, skewness and kurtosis of the variables and its scales, values of means, standard deviations and percentiles in case of the demographic characteristics of the participants. Pearson's Correlation Coefficient, Multiple Linear Regression, and Independent Samples *t*-test were computed to execute results by using "Statistical Package for the Social Science (SPSS) version 21".

Table 1

Psychometric Properties of Psychological Instruments (N = 110)

Instruments	K	M	SD	α	Skewness	Kurtosis
Parental Stress	110	65.46	4.6	0.83	.28	-.50
Parental Burden	110	32.14	2.5	0.90	-.24	-.63
Family Quality of Life	110	79.05	4.06	0.94	.29	-.29

Note. *k* = No of items; α = Cronbach's alpha; *SD* = Standard Deviation

The result concluded that psychometric properties of variables had significant skewness and kurtosis values i.e., all assessment scales used in the study had good reliability to use in the current study. The

distribution of the assessment ranges was normal. Parental stress, familial burden and family quality of life values were .83, .90 and .94 respectively.

Table 2*Characteristics of the Sample (N=110)*

Variables	M(SD)	f (%)	Variables	M(SD)	f (%)
Age	6.59(1.88)	-----	Reaction upon diagnosis	2.36(.70)	
Socioeconomic Status	2.1(.62)	-----	Denial	-----	15(27.3)
Lower	-----	6(16.9)	Hard to accept	-----	13(23.6)
Middle	-----	38(69.1)	Acceptance	-----	27(49.1)
Upper	-----	11(20.1)	Age at Diagnosis	1.47(.66)	-----
Mother's Occupation	1.67(.48)	-----	At birth	-----	34(61.8)
Working	-----	18(32.7)	Within 1 year	-----	16(29.1)
Housewives	-----	37(67.3)	Within 2 years	-----	5(9.1)
Family System	1.40(.49)	-----	Future Concerns	2.05(.83)	-----
Nuclear	-----	18(32.7)	Inclusive Education	-----	17(30.9)
Joint	-----	37(67.3)	Mainstream	-----	18(32.7)
			Vocational Training	-----	20(36.4)

Note. M=Means, SD= Standard Deviations, f= frequencies, %=Percentages

Results revealed that most of the participants were belonged to middle socioeconomic status with mean of ($M=2.1$, $SD=.62$), and from belonged to nuclear family system ($M=1.40$, $SD=.49$). The percentage of working women were higher ($M=1.67$, $SD=.48$). The acceptance of having special

children i.e. Down Syndrome was very difficult for the parents ($M=2.05$, $SD=.83$). Furthermore, majority of the parents were willing to make their children study at mainstream schools ($M=2.05$, $SD=.83$) rather than inclusive education or vocational training.

Table 3*Correlation among Parental Stress, Familial Burden, and Family Quality of Life (N=110)*

Variables	1	2	3	M	SD
1. Parental Stress	—	.37***	-.20*	65.40	4.61
2. Familial Burden		—	-.06	32.13	2.52
3. Family Quality of Life			—	78.84	4.06

* $p<0.05$, ** $p<0.01$, *** $p<0.001$

Results concluded that, there is significant positive relationship among parental stress with familial burden $r = .37$, $p < .0001$. Thus,

the parental stress increase with enhancement of familial burden among parents of children with Down Syndrome.

Table 4

Linear Regression Analysis for Predicting Family Quality of Life among Parents of Children with Down Syndrome (N=110)

Variables	B	SE(B)	β	R ²	ΔR^2
				.33	.32
Parental Stress	.20	.04	.43***		
Familial Burden	.11	.04	.25**		

Note. ΔR^2 = Adjusted R^2 ; B= Unstandardized Coefficients; SE(B) = Standard error of unstandardized coefficients; β =Standardized Coefficient Beta. * $p<0.05$, ** $p<0.01$, *** $p<0.001$

The results showed two predictors 33.48% of the adjustment $R=.58$, $F(2,107)=26.81$, $p<0.005$. Parental stress significantly predicts quality of life in family ($\beta = .43$, $p<.001$). Furthermore, familial burden also shows significant prediction for family quality of life ($\beta = .25$, $p<.005$) among the parents of children with Down Syndrome. Both two

predictors, parental stress and familial burden were tested against family quality of life among the parents of the children with Down Syndrome, both of them significantly predicted family quality of life which shows that parental stress and familial burden influence the family quality of life among the parents of the children with Down Syndrome.

Figure 1

Predictors of Family Quality of Life among the Parents of Children with Downs Syndrome

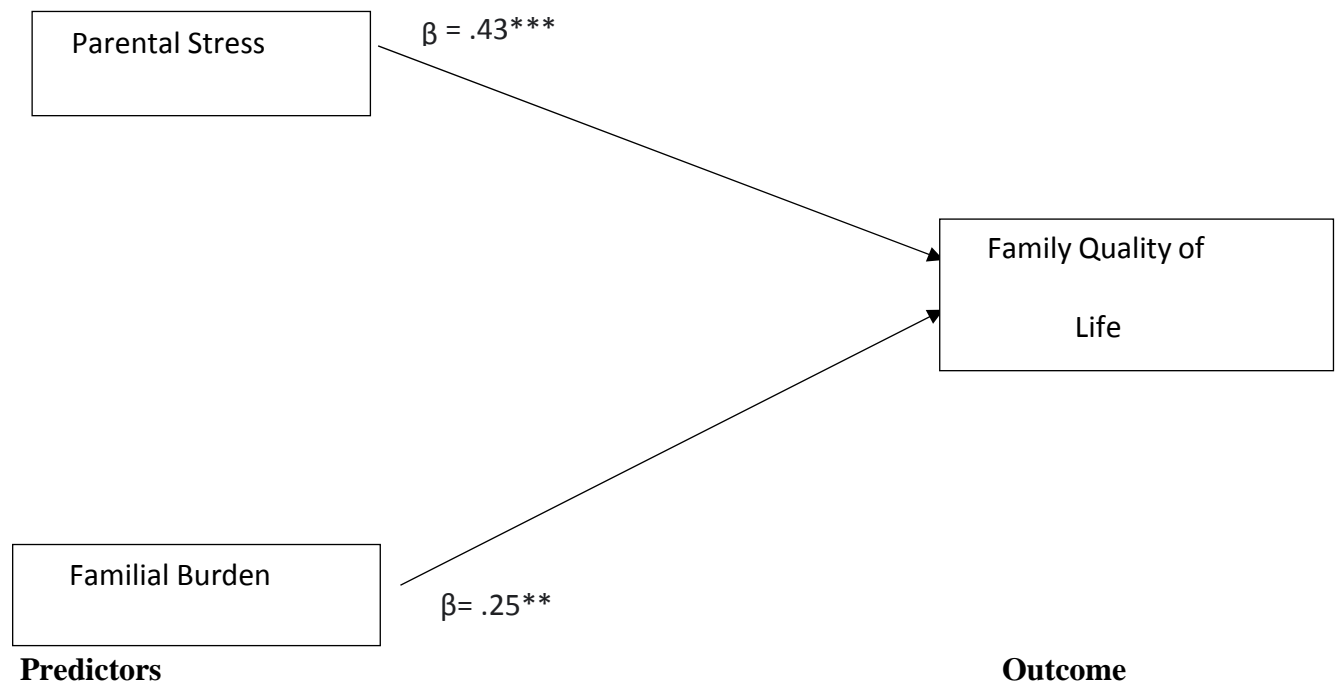


Table 5

Independent Samples t-test for Gender Differences in Parental Stress, Familial Burden, and Family Quality of Life in Parents of Children with Down Syndrome (N=110)

Variables	Mothers		Fathers		<i>t(df)</i>	<i>p</i>	95% CI		Cohen's <i>d</i>
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>			<i>LL</i>	<i>UL</i>	
PS	3.56	.24	3.71	.24	-3.32(108)	.001	-.24	-.06	.06
FB	3.12	.23	3.31	.23	-3.81(108)	.000	-.26	-.08	1.08
FQOL	4.20	.12	4.30	.12	-4.54(108)	.000	-.13	-.05	0.94

Note: Mothers = 55; Fathers = 55; *M* = mean; *SD* = standard deviation; CI = confidence interval; *LL* = lower limit; *UL* = upper limit; PS= Parental Stress; FB= Familial Burden

Results indicated that there is significant variance for the scores of mothers in parental stress ($M=3.56, SD=.24$) whereas for father ($M=3.71, SD=.24$), where $t(108) = -3.32, p=.001$. Fathers experienced more parental stress instead of mothers. Furthermore, familial burden among mothers ($M=3.12, SD=.23$) while for fathers ($M=3.31, SD=$

$.23$), where $t(108) = 3.81, p=.000$. In case of familial burden, the higher mean score shows that the fathers experience more familial burden as compared to mothers. It has been also showed that fathers have better quality of life in comparison of mothers who have children with Down Syndrome.

Discussion

The study was conducted to examine the severity of parental stress, familial burden, and how it effects the quality of life of those parents having children with Down Syndrome in Pakistan. The parental stress has deleterious effects on the whole family including children and overall family system (Staunton et al., 2020). In the present study, parental stress was found among the parents having children with Down Syndrome and also effects the wellbeing of the family both physical and psychological. Different researches proved there is a correlation between stress that effects individual's physical and psychological health although it also disturbed quality of life (Lappe et al., 2018).

The literature identified that parents of children with special abilities had stress as well as neurodevelopmental disorders (Lohaus et al., 2019), thus, our current study was also indicated that fathers and mother were in high stress but fathers were probably

in greater stress due to responsibility of children and whole family. Mothers have their own sense of responsibility, but fathers have comparatively more burdened due to multiple responsibilities, such as, financial, social, psychological, lagging work, excessive dues, lagging professional support of his children and cultural stigmatization. The study concluded that the fathers of the children diagnosed with Down Syndrome had a better quality of life as compared to mothers. As mothers and females have less tendency to tolerate stress and burden and are comparatively weak to bear emotional burden than males. There is a concealed gap of psychological stress with respect to the cultural context among the mothers of children with common psychiatric or developmental problems. The literature showed focused on the maternal stress and management problems, due to cultural sensitivity, it has been observed that male aspects is less explored (Nordstrom et al., 2020).

The result of a study showed that the parents of children with no neurodevelopment problem had higher marital intimacy than the parents having children with Down Syndrome (Carrada et al., 2019). The research also revealed high quality of life among parents of healthy children instead of parents of children with Down Syndrome (Sheridan et al., 2019). It has been also indicated in our findings that quality of life has been disturbed in the parents having children with Down Syndrome.

Conclusion

The study concludes that parents of the children with Down Syndrome has to face many factors that proliferate stress among them including, social pressure, lagging abilities of their children, excessive dependence of children on them, financial crisis, lack of professional support and stigmatization. These factors not only cause stress among parents but also influence the physical and mental health.

Implications

This study contributed to the existing literature in the field of Down Syndrome and factors which cause stress among parents and how the stress leads to various difficulties in life of the parents and leads to the poor quality of life among the families living in Pakistan. This research will support the clinical psychologist to devise the management plan to tackle the problems at very early stage while keeping in view the explored factors according to the cultural context.

Contribution of Authors

Areej Mahmood: Investigation, Data Curation, Formal Analysis, Writing - Original draft

Afsheen Gul: Conceptualization, Methodology, Writing- Reviewing & Editing, Supervision

Conflict of Interest

There is no conflict of interest declared by authors.

Source of Funding

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Data Availability Statement

The datasets of the current study are not available publicly due to ethical reasons but are available from the corresponding author [A.G.] upon the reasonable request.

References

- Berry, J. O., & Jones, W. H. (1995). The Parental Stress Scale: Initial Psychometric Evidence. *Journal of Social and Personal Relationships*, 12(3), 463–472. <https://doi.org/10.1177/0265407595123009>
- Bittles, A. H., Bower, C., Hussain, R., & Glasson, E. J. (2007). The four ages of Down syndrome. *The European Journal of Public Health*, 17(2), 221–225. <https://doi.org/10.1093/eurpub/ckl103>
- Bryant, L. D., Ahmed, S., Ahmed, M., Jafri, H., & Raashid, Y. (2011). “All is done by Allah”. Understandings of Down syndrome and prenatal testing in Pakistan. *Social Science & Medicine*, 72(8), 1393–1399. <https://doi.org/10.1016/j.socscimed.2011.02.036>
- Carrada, C. F., Scalioni, F. A. R., Abreu, L. G., Ribeiro, R. A., & Paiva, S. M. (2019). Impact of oral conditions of children/adolescents with Down syndrome on their families’ quality of life. *Special Care in Dentistry*, 40(2), 175–183. <https://doi.org/10.1111/scd.12444>
- Center for Disease Control and Prevention (2020, October 23). *Data and Statistics on Down Syndrome*. Centers for Disease Control and

- Prevention.
<https://www.cdc.gov/ncbddd/birthdefects/downsyndrome/data.html>
- Daundasekara, S. S., Schuler, B. R., Beauchamp, J. E. S., & Hernandez, D. C. (2021). The mediating effect of parenting stress and couple relationship quality on the association between material hardship trajectories and maternal mental health status. *Journal of Affective Disorders*, 290, 31–39. <https://doi.org/10.1016/j.jad.2021.04.058>
- De Clercq, L. E., Dieleman, L. M., van der Kaap-Deeder, J., Soenens, B., Prinzie, P., & De Pauw, S. S. W. (2021). Negative Controlling Parenting and Child Personality as Modifiers of Psychosocial Development in Youth with Autism Spectrum Disorder: A 9-Year Longitudinal Study at the Level of Within-Person Change. *Journal of Autism and Developmental Disorders*, 51(8), 2891–2907. <https://doi.org/10.1007/s10803-020-04761-4>
- Field, A. (2017). *Discovering Statistics Using IBM SPSS Statistics: North American Edition*. SAGE Publications Ltd.
- Fucà, E., Costanzo, F., Ursumando, L., & Vicari, S. (2022). Parenting Stress in Mothers of Children and Adolescents with Down Syndrome. *Journal of Clinical Medicine*, 11(5), 1188. <https://doi.org/10.3390/jcm11051188>
- Glasson, E., Sullivan, S., Hussain, R., Petterson, B., Montgomery, P., & Bittles, A. (2002). The changing survival profile of people with Down's syndrome: implications for genetic counselling. *Clinical Genetics*, 62(5), 390–393. <https://doi.org/10.1034/j.1399-0004.2002.620506.x>
- Grau, H., Graessel, E., & Berth, H. (2014). The subjective burden of informal caregivers of persons with dementia: extended validation of the German language version of the Burden Scale for Family Caregivers (BSFC). *Aging & Mental Health*, 19(2), 159–168. <https://doi.org/10.1080/13607863.2014.920296>
- Hoffman, L., Marquis, J., Poston, D., Summers, J. A., & Turnbull, A. (2006). Assessing Family Outcomes: Psychometric Evaluation of the Beach Center Family Quality of Life Scale. *Journal of Marriage and Family*, 68(4), 1069–1083. <https://doi.org/10.1111/j.1741-3737.2006.00314.x>
- Hsiao, C.-Y., & Riper, M. V. (2009). Individual and family adaptation in Taiwanese families of individuals with severe and persistent mental illness (SPMI). *Research in Nursing & Health*, 32(3), 307–320. <https://doi.org/10.1002/nur.20322>
- Lappé, M., Lau, L., Dudovitz, R. N., Nelson, B. B., Karp, E. A., & Kuo, A. A. (2018). The Diagnostic Odyssey of Autism Spectrum Disorder. *Pediatrics*, 141(Suppl 4), S272–S279. <https://doi.org/10.1542/peds.2016-4300C>
- Latzer, I. T., Leitner, Y., & Karnieli-Miller, O. (2021). Core experiences of parents of children with autism during the COVID-19 pandemic lockdown. *Autism*, 25(4), 136236132098431. <https://doi.org/10.1177/1362361320984317>

- Lohaus, A., Rueth, J.-E., & Vierhaus, M. (2019). Cross-Informant Discrepancies and their Association with Maternal Depression, Maternal Parenting Stress, and Mother-Child Relationship. *Journal of Child and Family Studies*. <https://doi.org/10.1007/s10826-019-01625-z>
- Martin, C. A., Papadopoulos, N., Chellew, T., Rinehart, N. J., & Sciberras, E. (2019). Associations between parenting stress, parent mental health and child sleep problems for children with ADHD and ASD: Systematic review. *Research in Developmental Disabilities*, 93, 103463. <https://doi.org/10.1016/j.ridd.2019.103463>
- Miragoli, S., Balzarotti, S., Camisasca, E., & Di Blasio, P. (2018). Parents' perception of child behavior, parenting stress, and child abuse potential: Individual and partner influences. *Child Abuse & Neglect*, 84, 146–156. <https://doi.org/10.1016/j.chiabu.2018.07.034>
- Nordstrøm, M., Retterstøl, K., Hope, S., & Kolset, S. O. (2020). Nutritional challenges in children and adolescents with Down syndrome. *The Lancet Child & Adolescent Health*, 4(6), 455–464. [https://doi.org/10.1016/s2352-4642\(19\)30400-6](https://doi.org/10.1016/s2352-4642(19)30400-6)
- Rahman, S., & Obaid-ur-Rahman, M. (2005). Prevalence rate of Down's syndrome in Karachi resident women. *Pakistan Journal of Pharmaceutical Sciences*, 18(2), 61–63. <https://pubmed.ncbi.nlm.nih.gov/16431402/>
- Selway, D., & Ashman, A. F. (1998). Disability, Religion and Health: A literature review in search of the spiritual dimensions of disability. *Disability & Society*, 13(3), 429–439. <https://doi.org/10.1080/09687599826722>
- Sheridan, C., OMalley-Keighran, M., & Carroll, C. (2019). What are the perspectives of adolescents with Down syndrome about their quality of life? A scoping review. *British Journal of Learning Disabilities*. <https://doi.org/10.1111/bld.12299>
- Siddiqui, A., Ladak, L. A., Kazi, A. M., Kaleem, S., Akbar, F., & Kirmani, S. (2021). Assessing Health-Related Quality of Life, Morbidity, and Survival Status for Individuals With Down Syndrome in Pakistan (DS-Pak): Protocol for a Web-Based Collaborative Registry. *JMIR Research Protocols*, 10(6), e24901. <https://doi.org/10.2196/24901>
- Staunton, E., Kehoe, C., & Sharkey, L. (2020). Families under pressure: stress and quality of life in parents of children with an intellectual disability. *Irish Journal of Psychological Medicine*, 1–8. <https://doi.org/10.1017/ipm.2020.4>