THE QUALITY OF LIFE OF MOTHERS WHO HAVE CHILDREN WITH DEVELOPMENTAL DISORDERS

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A research report (in the format of a “submissible” paper) submitted to the Faculty of Health Sciences, University of the Witwatersrand, Johannesburg, in partial fulfilment of the requirements for the degree of Master of Science in Medicine (Genetic Counselling).

Cape Town, 2020

Declaration

I, **Antonetta Malan**, declare that this research report (in the format of a “submissible” paper) is my own, unaided work. It is being submitted for the Degree of Master of Science in Medicine at the University of the Witwatersrand, Johannesburg. It has not been submitted before for any degree or examination at any other university.

AMalan

(signature of candidate)

28th day of February 2020, in Cape Town

Contribution of the candidate to the paper

Declaration: Student's contribution to article and agreement of co-authors

I, Antonetta Malan, student number 792831, declare that this Research Report is my own work and that I contributed significantly towards research findings presented in the paper intended for publication.

Signature of student AMalan

Date 11/10/2019

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Agreement by co-authors: By signing this declaration, the co-authors listed below agree to the use of the article(s) by the student as part of her Research Report.

Article Title: The Quality of Life of Mothers who have Children with Developmental Disorders

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Dedication

This research report is dedicated to my parents, Frans and Antonetta Malan, who always support and encourage me and taught me that I can make a difference in this world.

Presentations arising from this research project

1. South African Society of Human Genetics Biennial Congress 2019, 3 – 6 August, Century City, Cape Town, Western Cape (poster presentation)
2. School of Clinical Medicine Biennial Research Day 2019, 23 October, University of the Witwatersrand, Johannesburg, Gauteng (poster presentation)
3. Division of Human Genetics Departmental Seminar, 30 October, National Health Laboratory Service, Braamfontein, Johannesburg, Gauteng (oral presentation)

Abstract

Limited research has been performed to establish how having a child with a developmental disorder affects the quality of life (QOL) of mothers in South Africa. Developmental disorders encompass developmental delay (DD) and intellectual disability (ID) and refer to significant limitations in different facets of development. They occur during childhood and often persist into adulthood and have a severe impact on psychological and social well-being. Quality of life is a universal concept that refers to an individual's sense of overall well-being. Having a child with a developmental disorder can have an impact on the QOL of mothers caring for them. Limited research has been performed to establish how having a child with a developmental disorder affects the quality of life (QOL) of mothers in South Africa.

The aim of this study was to establish whether having a child with a developmental disorder significantly influences the QOL of South African mothers.

This was a descriptive case-control study that used quantitative research methods. It was nested within two larger studies: Deciphering Developmental Delay (DDD) in Africa and the Long-term Effects of Gestational Diabetes. The World Health Organization Quality of Life Abbreviated Version (WHOQOL-BREF) questionnaire was used as a researcher administered tool to assess the QOL of 20 mothers of children with developmental disorders (cases) and 20 mothers of typically developing children (controls). The tool consists of 26 questions and assesses QOL in four domains: physical, psychological, social and environmental.

The QOL measures in the cases were statistically significantly lower than those for the controls in the physical ($p=0.01$), psychological ($p=0.004$) and environmental ($p<0.001$) domains. The QOL measure in the social domain showed no significant difference between cases and controls ($p=0.077$). No demographic and socio-economic factors were associated with low or high QOL in any one of the four domains, and no significant differences were in demographics or socio-economic status seen between cases and controls.

This study has shown that having a child with a developmental disorder significantly lowers the QOL of mothers in South Africa. Healthcare providers need to be aware of this. Interventions are needed, such as referral for further assistance, including support groups and psychological care.

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To my parents, Frans and Antonetta, I cannot begin to express my gratitude. I thank you for your endless love and support and for raising me to be a woman who believes she can one day change the world through kindness.

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List of abbreviations

$	Dollar
DD	Developmental delay
DDD	Deciphering developmental delay
DPHRU	Developmental Pathways of Health Research Unit
etc.	Et Cetera
ID	Intellectual disability
IQR	Interquartile range
LMIC	Low-to-middle-income countries
MRC	Medical Research Counsel
NHLS	National Health Laboratory Service
p-value	Probability value
QOL	Quality of life
R	Rand
SD	Standard deviation
WHO	World health organization
WHOQOL-BREF	World Health Organization Quality of Life Abbreviated Version Questionnaire
Wits	University of the Witwatersrand

Research report in the format of a “submissible” paper

The Quality of Life of Mothers who have Children with Developmental Disorders

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This work has been written up as a paper for submission to the Journal of Applied Research in Intellectual Disabilities. The author guidelines (Appendix A) have been followed, however figures were embedded in the document and not attached as a separate file.

Abstract

Running title: The Quality of Life of Mothers who have Children with Developmental Disorders

Keywords: Quality of Life, Mothers, Developmental Disorders

Abstract

Background: Limited research has been performed on how having a child with a developmental disorder affects the quality of life (QOL) of mothers in South Africa. This study aimed to establish whether having a child with a developmental disorder significantly influences the QOL of their mothers.

Method: The World Health Organization Quality of Life Abbreviated Version questionnaire (WHOQOL-BREF) was used to assess the QOL of 20 mothers of children with developmental disorders (cases) and 20 mothers of typically developing children (controls). Four domains were assessed: physical, psychological, social and environmental.

Results: The QOL measures of the cases were statistically significantly lower than those of the controls in the physical (p=0.010), psychosocial (p=0.004) and environmental (p<0.001) domains. The social domain showed no significant difference (p=0.077).

Conclusions: Having a child with a developmental disorder significantly lowers the QOL of mothers in South Africa. Awareness and interventions, such as support groups and psychological care, are needed.

[149 words]

Introduction

Developmental disorders encompass developmental delay (DD) and intellectual disability (ID), which refer to significant limitations in intellectual functioning, adaptive behaviour and age-related development. They include neurodevelopmental, emotional, and behavioural disorders that occur during childhood, often persisting into adulthood, and have a severe impact on psychological and social well-being. Children with these disorders often require additional support from families and educational systems (Wehmeyer & Thompson, 2016).

Developmental delay is diagnosed in children when they fail to reach two or more age-related developmental milestones in specific areas of development, such as gross or fine motor skills, language or speech, cognitive development, social or personal achievements, as well as activities of daily living. Developmental delay could lead to a future diagnosis of ID. Intellectual disability is defined by noticeable, and often quantifiable, limitations in intellectual performance and conceptual, social, and practical adaptive skills. (Katz & Lazcano-Ponce, 2008). Clinically, ID is diagnosed using standardized measures of developmental skills in a child with DD. These measures include intelligence quotient (IQ), adaptive behaviour and developmental skills and are normally measured at approximately six years of age. The prevalence of ID is estimated to be between 1–3% in developed countries and presumed to be higher in developing countries, making it the largest global disability group (Moeschler & Shevell, 2014).

Developmental disorders can be caused by exogenous factors, such as maternal alcohol exposure during pregnancy, infections, birth complications and extreme malnutrition, but genetics are known to have an important role in its aetiology. Developmental disorders have become the most frequent reason for referral to paediatric genetic clinics (Vissers *et al*., 2017). A significant proportion of individuals who comes to the attention of medical geneticists and genetic counsellors for having a developmental disorder, have a specific genetic condition or syndrome. These can be single gene disorders, such as in Fragile X syndrome, or chromosomal disorders, such as Down syndrome (Kuczynski & Udwin, 2016). The clinical heterogeneity of developmental disorders is reflected with extreme genetic heterogeneity, which means that a genetic diagnosis is still lacking in most cases. A genetic diagnosis could provide valuable information on the type of developmental disorder, the prognosis, recurrence risk, possible complications, treatment options and relevant information on inheritance which could affect the rest of the family. Additionally, having a child with a developmental disorder impacts the QOL of parents caring for them (Vissers *et al.*, 2017).

Quality of life is a universal concept that has different political, philosophical, and health related definitions. It refers to an individual's sense of overall well-being, and includes physical, psychological, emotional, social, and spiritual dimensions (Cohen & Biesecker, 2010). Quality of life has been noted and discussed in academic literature since the 1960s and has since become increasingly important in healthcare, as modern treatment is able to extend length of life, sometimes at the expense of QOL, not only of those affected, but also of those caring for them (Karimi & Brazier, 2016).

Quality of life can be defined in many ways, which makes its measurement and incorporation into scientific research difficult (Donald, 2009). Quality of life is not a single measurement, but rather a composition of several smaller aspects influencing an individual's satisfaction with life. Expectations regarding health and the ability to cope with limitations (physical, personal, financial etc.) and disability can greatly affect a person's perception of their own health and satisfaction with life. Multiple individuals with the same health status may have very different qualities of life (Testa & Simonson, 1996). This speaks to the presence of other factors that influence QOL. Factors such as health and healthcare, welfare, human rights, availability of shelter, education facilities and income must all be considered as having an influence when determining QOL (Kehler, 2013).

A child with a developmental disorder presents multiple parenting challenges. In a previous study, parents reported heightened levels of stress in domains related to caring for children with developmental disorders. The stress might not be directly caused by the child's developmental disorder, but rather due to the increased demands resulting from their support needs, such as feeding, transport, toileting, and bathing, as well as the presence of related behavioural problems (Baker *et al.*, 2003). These heightened levels of stress place parents at an increased risk for developing depression and anxiety, which adversely affects their QOL. Chronic psychological distress could also place parents at an increased risk of marital disruption, family dysfunction, and various mental and physical conditions (McConnell & Savage, 2015). Some parents may view caring for a child with a developmental disorder as uncontrollably threatening, while others view the added pressure as a challenge and become stronger in the process. Coping strategies employed by parents, and the decisions they make, will influence their QOL (Pritzlaff, 2001).

Lower socio-economic status in low-to-middle-income countries (LMIC), such as South Africa, is related to higher parental depression and lower QOL in parents of children with developmental disorders (Havercamp & Scott, 2015). South Africa, as a multicultural LMIC, has a unique set of factors that must be considered when assessing the QOL of its citizens. Factors and challenges such as poverty and social inequality, urbanisation and the housing backlog (inadequate provision of government funded housing), welfare challenges, crime, HIV/AIDS and poor health, migration and housing, race, class, and gender-based discrimination must all be considered (Møller, 2006). Black rural women are faced with an even greater lack of access to resources and prosperity and therefore live under immense poverty. Kehler (2013) showed that poor, black women's access to resources, opportunities and education, as well as to growth and wealth of the country, is severely limited. Kehler (2013) also stated that historically cultural norms classified women as less important members of society who mainly have a domestic role of bearing and taking care of children. Herring *et al*. (2006) found that mothers of children with autism generally report greater distress than fathers. This could be because traditionally it is the role of the woman to look after the children and therefore interact with them more. South African mothers of children with developmental disorders are of interest, because there is a worldwide trend of women becoming the primary breadwinners in their households. According to African Bank, a growing number of women in South Africa are becoming financially independent (Botha, 2017). A Group Executive from African Bank, Melloy Ramalho, who was interviewed by Botha (2017) also stated that these women are breaking past glass ceilings and making things happen for themselves, but there is no denying they still come up against many challenges in the process, like making their own financial future a priority. Mothers of children with developmental disorders in South Africa thus face additional challenges, as they are trying to become more financially independent, whilst having a child with special needs to care for.

Limited research has been performed to establish how having a child with a developmental disorder affects the QOL of mothers in South Africa. As South Africa is a country with constrained resources, establishing the QOL of these mothers could highlight where intervention and support may be needed. Interventions surrounding resource allocation and healthcare policy can be made as all healthcare systems have to confront the reality of a finite budget and infinite demands (Donald, 2009). Interventions aimed at modifying psychosocial and contextual factors that influence QOL could also be implemented (Cohen & Biesecker, 2010).

The aim of this study was therefore to establish whether having a child with a developmental disorder influences the QOL of mothers caring for them. The objectives were to compare the QOL scores of mothers who have children with developmental disorders to those of mothers of typically developing children; to determine whether any demographic or socio-economic factors were associated with a high or low QOL in any of the four domains; and to identify areas where interventions can be implemented to improve QOL.

Methodology

Study design

This was a descriptive, case-control study that used quantitative research methods. It was nested within two larger studies: Deciphering Developmental Delay (DDD) in Africa (Division of Human Genetics, the National Health Laboratory Service, and the University of the Witwatersrand (Wits)) and the Long-term Effects of Gestational Diabetes (The MRC/Wits Developmental Pathways of Health Research Unit (DPHRU)).

Sampling

Convenience sampling was used. Twenty mothers of children with developmental disorders were recruited from the DDD Africa study and formed the case group. A moderate to severe development disorder was diagnosed in a child, through a developmental assessment by a medical doctor, before the child was recruited into the DDD Africa study. Twenty mothers of typically developing children were recruited from the DPHRU and formed the control group. Some of the mothers in the control group previously had gestational diabetes mellitus during their pregnancies. However, all the children from the control group received a cognitive assessment and were considered as typically developing. The women included in this study were all of African ancestry.

Inclusion and exclusion criteria

English literate South African mothers who were over the age of 18 years and had a child over the age of one year were recruited into the study. In the case group, mothers who had more than one child with a developmental disorder were excluded, and in the control group, mothers who had a critically or acutely ill child were excluded. All the participants were of African ancestry.

Data collection instrument

The World Health Organization Quality of Life Measure Abbreviated version (WHOQOL-BREF), which is a shortened version of the WHOQOL-100, was used to measure the QOL of the participants (Appendix B). This international, cross-culturally validated questionnaire consists of 26 questions and evaluates the following domains: physical health, psychological health, social relationships, and the environment. Facets incorporated within the domains can be seen in Table 1. The four domain scores denote a participant's perception of QOL in each particular domain. Scores were scaled in a positive direction (higher scores denote higher QOL). As per instructions from the WHO, the mean score of items within each domain was used to calculate the domain score. Mean scores were then multiplied by four to give domain scores out of 100, which is comparable with the scores used in the WHOQOL-100. There were also two items that were examined separately: Question one asked about the participant's overall perception of their QOL, and Question two asked about the participant's overall perception of their health (World Health Organization, 1996). The questionnaire was researcher administered. Demographic and socio-economic questions were included. A household asset score was calculated out of 11, by giving the participant a mark for each item present in their household. This has been validated in the South African context as a proxy for household socio-economic status (Feeley *et al.*, 2013) Furthermore, a question asking whether the participants had any suggestions to improve the QOL of mothers in South Africa was added.

Table 1: WHOQOL-BREF Domains (World Health Organization, 1996)

Domain	**Facets incorporated within domains**
Physical Health	Activities of daily living Dependence on medical substances and medical aids Energy and fatigue Mobility Pain and discomfort Sleep and rest Work capacity
Psychological	Bodily image and appearance Negative feelings Positive feelings Self-esteem Spirituality/Religion/Personal beliefs Thinking, learning, memory and concentration
Social Relationships	Personal relationships Social support Sexual activity
Environment	Financial resources Freedom, physical safety and security Health and social care: accessibility and quality Home environment Opportunities for acquiring new information and skills Participation in opportunities for recreation Physical environment Transport

Data analysis

Data were captured into a REDCap (Research Electronic Data Capture) database (Harris *et al.*, 2009) and descriptive statistical analyses were performed using the RealStats package in Excel. For continuous data, normally distributed data were presented as means and standard deviations and non-normally distributed data were presented as medians and interquartile ranges. Categorical data were presented as frequencies and percentages. The chi-square test was used to assess the difference between categorical variables of cases and controls, and the student t-test and Mann-Whitney test was used to assess the difference between continuous variables of cases and controls. Multiple linear regression analysis was used to determine predictors for higher or lower QOL scores. Predictors were variables that were shown to be statistically significantly different between cases and controls, and the QOL score was used as the outcome variable and was kept continuous. Significance was assumed at a two-tailed p-value of <0.05 and a large effect size was assumed at effect r > 0.5.

Ethical considerations

Ethical clearance was granted by the University of the Witwatersrand's Human Research Ethics Committee (Medical) (Certificate reference: M190257; Appendix C). Study participants gave informed, written consent to participate. Participants who scored lower than the average in the psychological domain received referrals to psychologists.

Results

Characteristics of participants

A total of 40 participants were recruited into the study: 20 cases (mothers of children with developmental disorders) and 20 controls (mothers of typically developing children). The sociodemographic data of the participants are shown in Table 2. Cases did not differ demographically or socio-economically from controls.

Table 2: Characteristics of the study participants

	Mean ± SD / n (%) / Median (IQR)				
Characteristics	**Total (n=40)**	**Cases (n=20)**	**Controls (n=20)**	**p-value**	**Effect r**
Age (years)	34.98 ± 6.40	35.55 ± 6.24	34.40 ± 6.62	0.575	0.091
Marital status				0.323	
Single	17.00 (42.50%)	7.00 (35.00%)	10.00 (50.00%)		
Married/Cohabiting	22.00 (55.00%)	13.00 (65.00%)	9.00 (45.00%)		
Separated/Divorced	1.00 (2.50%)	0.00 (0.00%)	1.00 (5.00%)		
Education				0.303	
Primary school	1.00 (2.50%)	1.00 (5.00%)	0.00 (0.00%)		
Secondary school	25.00 (62.50%)	13.00 (65.00%)	12.00 (60.00%)		
Professional training	10.00 (25.00%)	3.00 (15.00%)	7.00 (35.00%)		
University	4.00 (10.00%)	3.00 (15.00%)	1.00 (5.00%)		
Household asset score (/11)	6.00 (5.00-7.00)	6.00 (5.00-7.00)	6.10 (5.00-7.00)	0.820	0.037
Age of index child (years)	4.00 (3.00-6.30)	5.00 (4.00-8.30)	4.00 (3.00-4.00)	0.120	0.400
Parity	2.00 (1.80-3.00)	2.50 (1.80-3.00)	2.00 (1.00-3.00)	0.174	0.222

SD = Standard deviation; IQR = Interquartile range

Comparison of the QOL scores of cases and controls

Participants' mean QOL in all four domains and perceptions of own QOL and health can be seen in Table 3. As compared to mothers in the control group, mothers of children with developmental disorders scored significantly lower in the physical, psychological and environmental domains. Having a child with a developmental disorder had a large effect size in the environmental domain (effect r >0.5) (Table 3). Cases did not perceive their own QOL or health to be significantly lower than controls. No significant differences were seen between cases and controls in the social domain.

Table 3: QOL scores and perceived QOL and health scores of participants

WHOQOL-BREF domain (score /100)	**Mean ± SD / Median (IQR)**			
	Cases	**Controls**	**p-value**	**Effect r**
Physical score	54.05 ± 21.36	69.20 ± 12.99	0.010*	0.402
Psychological score	50.95 ± 20.55	68.55 ± 18.79	0.004*	0.444
Social score	59.40 ± 20.54	70.30 ± 17.17	0.077	0.283
Environmental score	42.20 ± 16.53	65.05 ± 11.91	< 0.001*	0.631**
Perception of own QOL	60.00 (40.00-80.00)	80.00 (60.00-80.00)	0.141	0.233
Perception of own health	80.00 (40.00-80.00)	60.00 (40.00-80.00)	0.620	0.083

*Significant difference (p<0.05); **Large effect size (>0.50); SD = Standard deviation; IQR = Interquartile range: QOL = Quality of life

The linear regression model for the four domains can be seen in Table 4. It showed that none of the demographic or socio-economic variables (participant age, marital status, education, household asset score, age of index child or parity) were significantly associated with QOL score in either one of the four domains. The only variable that significantly lowered QOL in the physical, psychological and environmental domains was having a child with a developmental disorder. None of the variables significantly influenced the QOL scores in the social domain.

When participants were asked what they thought would improve overall QOL of women in South Africa, most of them answered that they would like more support groups and improved access to educational resources to help them understand their children's needs better.

Table 4: Linear regression analysis for factors associated with QOL

Characteristic	Physical domain					Psychological domain				
	Coefficient	95% confidence interval	Standard error	t statistic	p-value	Coefficient	95% confidence interval	Standard error	t statistic	p-value
Intercept	78.70	[33.98;123.41]	21.95	3.59	0.001*	32.37	[-19.11;83.86]	25.28	1.28	0.21
Age (years)	-0.96	[-2.11;0.19]	0.57	-1.69	0.10	0.08	[-1.25;1.40]	0.65	0.12	0.91
Marital status	-4.11	[-16.32;8.10]	6.00	-0.69	0.50	2.10	[-11.08;17.05]	6.90	0.43	0.67
Education	2.44	[-5.55;10.43]	3.92	0.62	0.54	7.24	[-1.96;16.45]	4.52	1.60	0.12
HAS	0.98	[-3.39;5.34]	2.14	0.46	0.65	1.35	[-3.67;6.38]	2.47	0.55	0.59
Index child's age	0.95	[-1.36;3.26]	1.14	0.84	0.41	-0.46	[-3.12;2.20]	1.31	-0.35	0.73
Parity	-2.16	[-9.52;5.19]	3.61	-0.60	0.55	-3.42	[-11.89;5.05]	4.16	-0.83	0.42
Control^{x}	14.43	[2.51;26.36]	5.90	2.47	0.02*	15.05	[1.31;28.77]	6.74	2.23	0.03*
Adjusted R^2	0.28					0.14				
F-statistic	3.19 (df=39); p = 0.01*					1.92 (df=39); p= 0.10				
	Social domain					Environmental domain				
Intercept	62.44	[9.84;115.05]	25.82	2.41	0.02*	38.45	[-1.06;77.96]	19.40	1.98	0.06
Age (years)	-0.40	[-1.75;0.95]	0.66	-0.60	0.55	-0.37	[-1.39;0.64]	0.50	-0.75	0.46
Marital status	7.70	[-6.66;22.07]	7.05	1.09	0.28	-9.97	[-20.58;1.00]	5.60	-1.85	0.08
Education	1.65	[-7.75;11.05]	4.61	0.36	0.72	-2.37	[-9.43;4.69]	3.47	-0.68	0.50
HAS	3.04	[-2.09;8.17]	2.52	1.21	0.23	3.15	[-0.71;7.00]	1.89	1.66	0.11
Index child's age	-1.21	[-3.92;1.51]	1.33	-0.91	0.37	0.49	[-1.55;2.54]	1.00	0.49	0.63
Parity	-3.42	[-12.07;5.23]	4.25	0.81	0.43	2.64	[-3.86;9.14]	3.19	0.83	0.41
Control^{x}	7.10	[-6.94;21.12]	6.89	1.03	0.31	24.05	[13.51;34.59]	5.17	4.65	<0.001**
Adjusted R^2	0.05					0.39				
F-statistic	1.29 (df=39); p= 0.29					4.63 (df=39); p=0.001*				
HAS = household asset score; *significant difference (p<0.05); **significant difference (p<0.001); Control^{x} = mothers who have typically developing children; df = degrees of freedom										

Discussion

The results of this study demonstrate that, overall, the QOL of mothers who have children with developmental disorders is lower than mothers who have typically developing children. This was particularly seen across the physical, psychological and environmental domains, but there was no significant difference between cases and controls in the social domain. The lack of a significant difference in the social domain was an unexpected finding as other similar studies have found that parents of children with developmental disabilities, compared to parents of typically developing children, have a lower QOL across all four domains (Brown *et al*., 2006; Alhazmi *et al*., 2018).

The facets incorporated within the social domain assess personal relationships, social support and sexual activity (World Health Organization, 1996). The lack of finding a significant difference of QOL scores between cases and controls in the social domain in this study could be explained by the possible positive social impact having a child with a developmental disorder could have on their parents, how adversity unites individuals, and community support (Pritzlaff, 2001). In a Brighton & Wills (2016) study, parents reported better social relations, they recounted how having more meaningful relationships with other family members and with professionals have become more important to them since a diagnosis of a developmental disorder in their child. They reported that it strengthened their marriage by making them work better together and support one another. Parents described themselves as being more patient, accepting, empathic, and compassionate individuals. In addition, they reported having better relationships with other members of the community. Africa has an indigenous philosophy of "Ubuntu", which means that community strength comes from community support. "Ubuntu" is essentially linked to the ideas of cooperation and working together, and in essence, it is instilled principles that compel individuals and families to care for each other out of moral obligation (Twikirizwe & Spitzer, 2019). Given that all the participants were of African ancestry, the concept of "Ubuntu" could explain why no significant difference was seen between cases and controls in the social domain, as they have a form of social support and security not necessarily seen in other cultures. "Ubuntu" has relevance in the field of informal social security in South Africa. Social protection is rooted in principles of justice and equality. "Ubuntu" stresses the fact that the fundamental human need is to make life meaningful for everybody (Tshoose, 2009). This would mean that it would be the responsibility of the community to provide social support and security for mothers of children with developmental disorders.

The lowest QOL scores, of both cases and controls, in this study were reported in the environmental domain. Cases scored significantly lower than controls ($p<0.001$). The facets incorporated within this domain measured financial resources, accessibility to health and social care, opportunity for leisure activities, security, and safety (World Health Organization, 1996). Considering the socio-economic context of South Africa, this finding is not unexpected.

Poverty, crime, the HIV epidemic, difficulties accessing health services, overburdened schools, poor physical safety, and a poor home environment are all contributing factors to participants' experiences in this domain (Tomlinson, 2013), evidently even more so for mothers who have children with developmental disorders. In South Africa, parents/guardians of children who have developmental disorders qualify for a government Care Dependency Grant, which is R1780.00 ($118.18) per month (SASSA, 2019). This money is intended for special schooling, specialised health services, and transport. Considering the cost of living in South Africa and the fact that the parents have themselves and other family members to support, in addition to their child with special needs, this is unlikely to be enough money. Parents taking care of children with disabilities have a lower employment potential. This could be due to having to assist their child during daily living, often resulting in the fact that these parents cannot work and therefore have no income (Parish *et al*., 2004). In addition, women are more likely to be impoverished in South Africa, with a poverty headcount of 58.6% as compared to 54.9 % in men (Statistics South Africa, 2019).

The physical domain measured participants' energy levels, capacity for work, daily living activities and sleep (World Health Organization, 1996). The significantly lower QOL score of the case group, as compared to the controls, can be explained by the added physical demands on mothers who have children with developmental disorders. A study performed in Hong Kong reported that some parents of children with developmental disabilities developed chronic pain due to repetitive strain helping their children with their daily living activities, such as transfer, toileting and bathing. Parents reported that they were physically exhausted in the provision of intensive care and attention to their child (Leung & Li-Tsang, 2003). South African homes are not equipped to accommodate a child with a developmental disorder and some households do not even have basic amenities like indoor plumbing or running water (Statistics South Africa, 2019). This places an increased burden on South African mothers of children with special needs. Homes need to be equipped to accommodate children with developmental disorders, such as hand railings to help with mobility, toileting and bathing, and at the very least running water and indoor plumbing.

The psychological domain measured participants’ feelings, self-esteem, and beliefs (religious, cultural and personal) (World Health Organization, 1996). It was expected that cases would score lower than controls in this domain. Parents who have children with developmental disorders have reported feelings of depression and increased levels of stress and anxiety. This is even more than true for mothers who are considered to be the primary caregivers and spend more time with their children (McConnell & Savage, 2015). This finding emphasises that mothers of children with developmental disorders need psychological care and support, such as referrals to social workers, counsellors and psychologists. This could also explain why many participants suggested support groups.

A study performed in Croatia found a positive association between lower QOL scores and lower educational levels in parents (Misura & Memisevic, 2017). The current study, however, found no association between QOL score in any of the four domains or in those of the demographic or socio-economic factors. Furthermore, the cases and controls did not differ significantly in socio-economic status or demographics, suggesting a fairly homogenous group. The only variable associated with a lower QOL score, was having a child with a developmental disorder. This was supported by the linear regression analysis.

This study provides valuable information and highlights the importance of physical, psychological and environmental support for mothers who have children with developmental disorders. This support can be in the form of a support group or psychology referrals, made by genetic counsellors, medical geneticists and other healthcare professionals. The diagnosis of a developmental disorder not only has a major impact on the individual diagnosed, but also on the main caregiver and the family as a whole. Support groups have been used for family intervention to improve care, as well as family and patient functioning (Chien *et al*., 2005). The objectives of caregiver support groups are to provide education to increase knowledge and understanding, to explore different coping strategies, and provide emotional support (Chou *et al*., 2002). In the current study mothers of children with developmental disorders expressed their needs and desires for these services. Genetic Clinics should move towards a multidisciplinary approach, where medical geneticists, genetic counsellors, psychologists, and social workers work together towards improving the QOL of caregivers and parents of children with developmental disorders. Hospital-based support groups could form a regular component of Genetic Clinics and could be run by genetic counsellors and genetic counselling trainees, with the purpose of providing education and emotional support. They could also serve to empower women to facilitate their own parent-based support groups. Recommendations have been made to establish a support group within the DDD Africa study.

This study was limited by sample size due to time constraints inherent in doing research for degree purposes. Furthermore, the WHOQOL-BREF used in this study, although internationally validated, has not been specifically validated in the South African population, and lastly, this study only looked at the QOL of mothers and is not representative of all caregivers.

Future research could include qualitative studies, to gain better insight into how having a child with special needs affects the QOL of mothers caring for them. Qualitative studies could determine what coping strategies mothers who have children with developmental disorders use. They could also investigate the exact challenges and burdens these mothers face daily. The concept of “Ubuntu” as a form of social security and support could be studied further. This could be done by interviewing mothers of children with developmental disorders on their perception of the social support they get from their community. While this study looked at mothers, it would be interesting to see how having a child with a developmental disorder impacts the quality of life of fathers and other caregivers of children with developmental disorders.

The findings in this study highlights that mothers of children with developmental disorders in South Africa are struggling and need additional support. This speaks to the need for policy changes in South Africa to provide improved assistance and healthcare services for this group of women.

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The topics it covers include community living, quality of life, challenging behaviour, communication, sexuality, medication, ageing, supported employment, family issues, mental health, physical health, autism, economic issues, social networks, staff stress, staff training, epidemiology and service provision. Theoretical papers are also considered provided the implications for therapeutic action or enhancing quality of life are clear. Both quantitative and qualitative methodologies are welcomed. All original and review articles continue to undergo a rigorous, peer-refereeing process.

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It is a requirement that all authors have been accredited as appropriate under submission of the manuscript. Contributors who do not qualify as authors should be mentioned under Acknowledgements.

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Under Acknowledgements please specify contributors to the article other than the authors accredited. Please also include specifications of the source of funding for the study and any potential conflict of interest if appropriate. Suppliers of materials should be named and their location (town, state/county, country) included.

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Appendix B: WHOQOL-BREF questionnaire

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Demographic and Socio-Economic Characteristics

1) What is your age (years): ☐

2) What is your marital status: (cross only one box)

Single	☐	Widowed	☐
Married/living with partner	☐	Separated/Divorced	☐

3) What is your highest level of education attended: (cross only one box)

No school attended	☐	Professional/technical training	☐
Primary school	☐	Secondary school	☐
University	☐		

4) Which of the following assets are present in your household? (cross the boxes that apply)

Electricity	☐	Cell Phone	☐	Bicycle	☐
Radio	☐	Personal computer	☐	Motorcycle/Scooter	☐
Television	☐	Farm animals	☐	Car/Truck/Tractor	☐
Refrigerator	☐	Agricultural land	☐		

5) What is the age of your child who has come to clinic with you today? ☐

6) How many children do you have in total? ☐

7) Do any of your children have medical problems? Y/N ☐

8) Have you personally ever been formally/officially been diagnosed by a medical professional with depression or any other psychiatric/psychological disorder? Y/N ☐

If yes, what? ____

The World Health Organization: Quality of Life (WHOQOL) -BREF

The following questions ask how you feel about your quality of life, health, or other areas of your life. I will ask the questions. **Please choose the answer that appears most appropriate.** If you are unsure about which response to give to a question, the first response you think of is often the best one.

Please keep in mind your standards, hopes, pleasures and concerns. We ask that you think about your life **in the last four weeks.**

		Very poor	Poor	Neither poor nor good	Good	Very good
1.	How would you rate your quality of life?	1	2	3	4	5

		Very dissatisfied	Dissatisfied	Neither satisfied nor dissatisfied	Satisfied	Very satisfied
2.	How satisfied are you with your health?	1	2	3	4	5

The following questions ask about **how much** you have experienced certain things in the last four weeks.

		Not at all	A little	A moderate amount	Very much	An extreme amount

3.	To what extent do you feel that physical pain prevents you from doing what you need to do?	5	4	3	2	1
4.	How much do you need any medical treatment to function in your daily life?	5	4	3	2	1
5.	How much do you enjoy life?	1	2	3	4	5
6.	To what extent do you feel your life to be meaningful?	1	2	3	4	5

		Not at all	A little	A moderate amount	Very much	Extremely
7.	How well are you able to concentrate?	1	2	3	4	5
8.	How safe do you feel in your daily life?	1	2	3	4	5
9.	How healthy is your physical environment?	1	2	3	4	5

The following questions ask about how completely you experience or were able to do certain things in the last four weeks.

		Not at all	A little	Moderately	Mostly	Completely
10.	Do you have enough energy for everyday life?	1	2	3	4	5
11.	Are you able to accept your bodily appearance?	1	2	3	4	5
12.	Have you enough money to meet your needs?	1	2	3	4	5
13.	How available to you is the information that you need in your day-to-day life?	1	2	3	4	5

14.	To what extent do you have the opportunity for leisure activities?	1	2	3	4	5

		Very poor	Poor	Neither poor nor good	Good	Very good
15.	How well are you able to get around?	1	2	3	4	5

		Very dissatisfied	Dissatisfied	Neither satisfied nor dissatisfied	Satisfied	Very satisfied
16.	How satisfied are you with your sleep?	1	2	3	4	5
17.	How satisfied are you with your ability to perform your daily living activities?	1	2	3	4	5
18.	How satisfied are you with your capacity for work?	1	2	3	4	5
19.	How satisfied are you with yourself?	1	2	3	4	5

20.	How satisfied are you with your personal relationships?	1	2	3	4	5
21.	How satisfied are you with your sex life?	1	2	3	4	5
22.	How satisfied are you with the support you get from your friends?	1	2	3	4	5
23.	How satisfied are you with the conditions of your living place?	1	2	3	4	5

24.	How satisfied are you with your access to health services?	1	2	3	4	5
25.	How satisfied are you with your transport?	1	2	3	4	5

The following question refers to how often you have felt or experienced certain things in the last four weeks.

		Never	Seldom	Quite often	Very often	Always
26.	How often do you have negative feelings such as blue mood, despair, anxiety, depression?	5	4	3	2	1

Do you have any suggestions to improve the quality of life of mothers in South Africa?

____

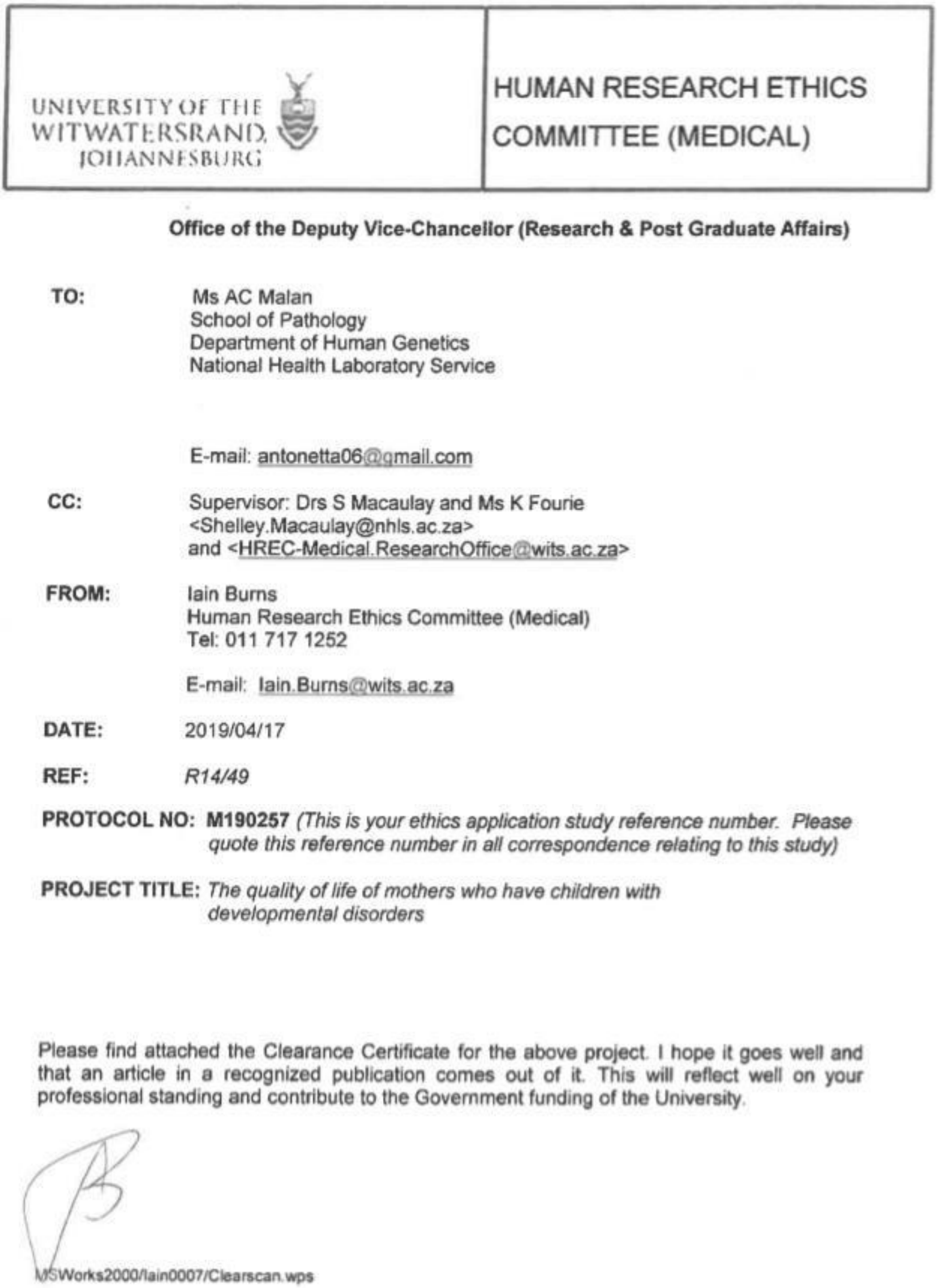
____

____

Please leave your name and contact details should you want us to contact you and arrange counselling with one of the supervisors if we detect a poor QOL in the psychosocial domain.

Name and Surname ____________________________________
Telephone number ____________________________________
Email address ____________________________________

Appendix C: Ethical clearance certificate



UNIVERSITY OF THE
WITWATERSRAND,
JOHANNESBURG

HUMAN RESEARCH ETHICS
COMMITTEE (MEDICAL)

Office of the Deputy Vice-Chancellor (Research & Post Graduate Affairs)

TO: Ms AC Malan
School of Pathology
Department of Human Genetics
National Health Laboratory Service

E-mail: antonetta06@gmail.com

CC: Supervisor: Drs S Macaulay and Ms K Fourie
<Shelley.Macaulay@nhls.ac.za>
and <HREC-Medical.ResearchOffice@wits.ac.za>

FROM: Iain Burns
Human Research Ethics Committee (Medical)
Tel: 011 717 1252

E-mail: Iain.Burns@wits.ac.za

DATE: 2019/04/17

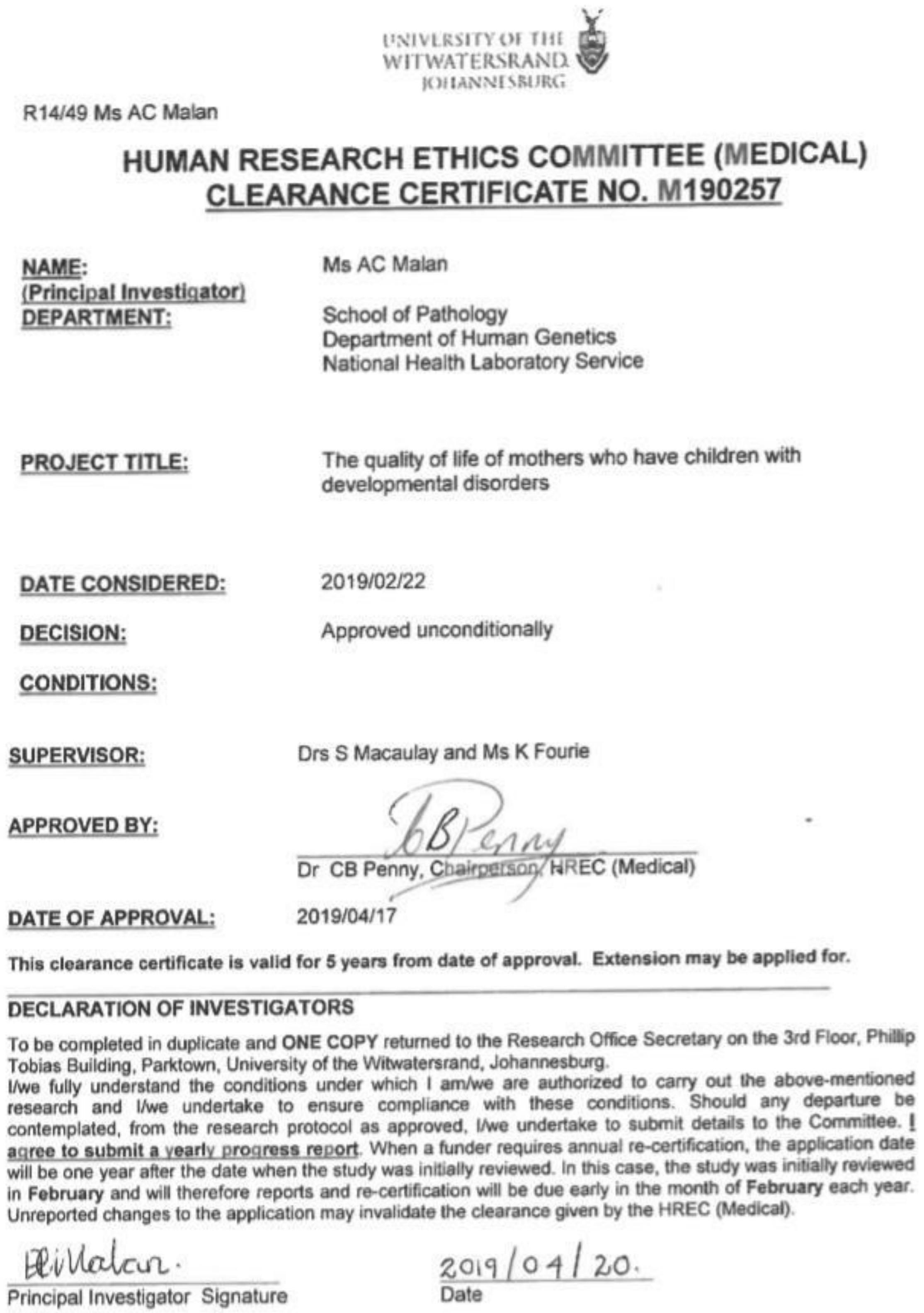
REF: *R14/49*

PROTOCOL NO: **M190257** *(This is your ethics application study reference number. Please quote this reference number in all correspondence relating to this study)*

PROJECT TITLE: *The quality of life of mothers who have children with developmental disorders*

Please find attached the Clearance Certificate for the above project. I hope it goes well and that an article in a recognized publication comes out of it. This will reflect well on your professional standing and contribute to the Government funding of the University.

MSWorks2000/Iain0007/Clearscan.wps



UNIVERSITY OF THE
WITWATERSRAND,
JOHANNESBURG

R14/49 Ms AC Malan

HUMAN RESEARCH ETHICS COMMITTEE (MEDICAL)
CLEARANCE CERTIFICATE NO. M190257

NAME: **(Principal Investigator)** Ms AC Malan

DEPARTMENT: School of Pathology
Department of Human Genetics
National Health Laboratory Service

PROJECT TITLE: The quality of life of mothers who have children with developmental disorders

DATE CONSIDERED: 2019/02/22

DECISION: Approved unconditionally

CONDITIONS:

SUPERVISOR: Drs S Macaulay and Ms K Fourie

APPROVED BY: CB Penny
Dr CB Penny, Chairperson, HREC (Medical)

DATE OF APPROVAL: 2019/04/17

This clearance certificate is valid for 5 years from date of approval. Extension may be applied for.

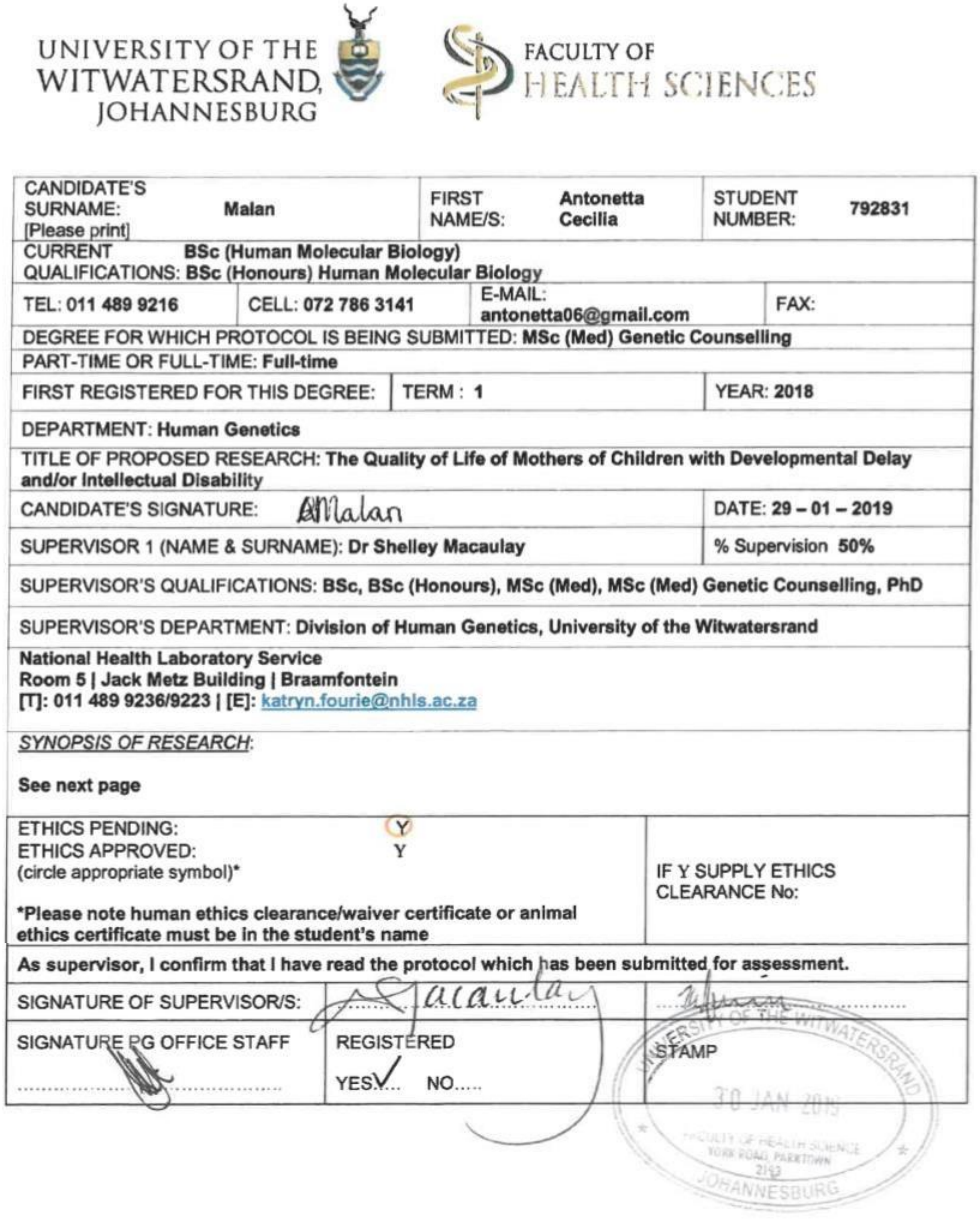
DECLARATION OF INVESTIGATORS

To be completed in duplicate and **ONE COPY** returned to the Research Office Secretary on the 3rd Floor, Phillip Tobias Building, Parktown, University of the Witwatersrand, Johannesburg.
I/we fully understand the conditions under which I am/we are authorized to carry out the above-mentioned research and I/we undertake to ensure compliance with these conditions. Should any departure be contemplated, from the research protocol as approved, I/we undertake to submit details to the Committee. **I agree to submit a yearly progress report**. When a funder requires annual re-certification, the application date will be one year after the date when the study was initially reviewed. In this case, the study was initially reviewed in **February** and will therefore reports and re-certification will be due early in the month of **February** each year. Unreported changes to the application may invalidate the clearance given by the HREC (Medical).

ACMalan.
Principal Investigator Signature

2019/04/20.
Date

Appendix D: Approved protocol



UNIVERSITY OF THE WITWATERSRAND, JOHANNESBURG

FACULTY OF HEALTH SCIENCES

CANDIDATE'S SURNAME: [Please print] **Malan**

FIRST NAME/S: **Antonetta Cecilia**

STUDENT NUMBER: **792831**

CURRENT QUALIFICATIONS: **BSc (Human Molecular Biology)**
BSc (Honours) Human Molecular Biology

TEL: **011 489 9216**

CELL: **072 786 3141**

E-MAIL: **antonetta06@gmail.com**

FAX:

DEGREE FOR WHICH PROTOCOL IS BEING SUBMITTED: **MSc (Med) Genetic Counselling**

PART-TIME OR FULL-TIME: **Full-time**

FIRST REGISTERED FOR THIS DEGREE: TERM : **1** YEAR: **2018**

DEPARTMENT: **Human Genetics**

TITLE OF PROPOSED RESEARCH: **The Quality of Life of Mothers of Children with Developmental Delay and/or Intellectual Disability**

CANDIDATE'S SIGNATURE: AMalan

DATE: **29 – 01 – 2019**

SUPERVISOR 1 (NAME & SURNAME): **Dr Shelley Macaulay**

% Supervision **50%**

SUPERVISOR'S QUALIFICATIONS: **BSc, BSc (Honours), MSc (Med), MSc (Med) Genetic Counselling, PhD**

SUPERVISOR'S DEPARTMENT: **Division of Human Genetics, University of the Witwatersrand**

National Health Laboratory Service
Room 5 | Jack Metz Building | Braamfontein
[T]: 011 489 9236/9223 | [E]: katryn.fourie@nhls.ac.za

SYNOPSIS OF RESEARCH:

See next page

ETHICS PENDING: [✓ Y]

ETHICS APPROVED: Y

(circle appropriate symbol)*

***Please note human ethics clearance/waiver certificate or animal ethics certificate must be in the student's name**

IF Y SUPPLY ETHICS CLEARANCE No:

As supervisor, I confirm that I have read the protocol which has been submitted for assessment.

SIGNATURE OF SUPERVISOR/S: Macaulay

SIGNATURE PG OFFICE STAFF

REGISTERED

YES ✓ NO.....

STAMP

UNIVERSITY OF THE WITWATERSRAND
30 JAN 2019
FACULTY OF HEALTH SCIENCE
YORK ROAD, PARKTOWN
2193
JOHANNESBURG

SYNOPSIS OF RESEARCH

Introduction

Limited research has been performed to establish how having a child with a developmental disorder affects the quality of life (QOL) of mothers in South Africa. Developmental disorders encompass developmental delay (DD) and intellectual disability (ID) and refer to significant limitations in intellectual functioning, adaptive behaviour and age-related development. They include neurodevelopmental, emotional, and behavioural disorders that occur during childhood and often persist into adulthood and have a severe impact on psychological and social wellbeing. Quality of life is a universal concept that has different political, philosophical and healthrelated definitions. It refers to an individual's sense of overall well-being, including physical, psychological, emotional, social, and spiritual dimensions. Having a child with a developmental disorder can have an impact on the QOL of the mothers caring for them.

Aim

The aim of the study is to establish whether having a child with a developmental disorder significantly influences the QOL of their mothers.

Participants and methods

This will be a descriptive case-control study using quantitative research methods. It will be nested within two larger studies; Deciphering Developmental Delay (DDD) in Africa, that the Division of Human Genetics, at the National Health Laboratory Service (NHLS) and the University of the Witwatersrand (Wits) is undertaking, and the Long-term Effects of Gestational Diabetes study that the Developmental Pathways of Health Research Unit (DPHRU) within the Department of Paediatrics at Wits is undertaking. Participants will be recruited from the DDD Africa study to form the case group and the control group will be recruited from the DPHRU study. Between 30 and 50 participants will be recruited into each group. The data collection tool is the World Health Organisation QOL Measure abbreviated version questionnaire (WHOQOL-BREF). It consists of 26 questions assessing QOL in four domains: physical health, psychological, social relationships and environment.

Anticipated outcomes

This study will provide valuable insight into the QOL of mothers caring for children with developmental disorders. By assessing the QOL of these mothers, targeted interventions can be recommended to assist them in getting care and support possible to take care of their children.

The Quality of Life of Mothers who have Children with Developmental Disorders

Antonetta Malan
Student Nr : 792831
MSc (Med) Genetic Counselling

Supervisor 1:
Dr Shelley Macaulay
MSc (Med) Genetic counselling, PhD, Genetic Counselling Manager and Lecturer
Clinical & Counselling Section, Division of Human Genetics
National Health Laboratory Service, The University of the Witwatersrand

Supervisor 2:
Katryn Fourie
MSc (Med) Genetic Counselling, Genetic Counsellor and Lecturer
Clinical & Counselling Section, Division of Human Genetics
National Health Laboratory Service, The University of the Witwatersrand

1. Background

1.1 Introduction

Limited research has been performed to establish how having a child with developmental disorders affects the quality of life (QOL) of mothers in South Africa. Considering that South Africa is a country with limited resources, establishing the QOL of these mothers could highlight where intervention and support may be needed.

1.2. Developmental disorders

Developmental disorders encompass developmental delay (DD) and intellectual disability (ID). Developmental delay and ID refer to significant limitations in intellectual functioning, adaptive behaviour and age-related development. They include neurodevelopmental, emotional, and behavioural disorders that occur during childhood and often persist into adulthood and have a severe impact on psychological and social well-being. Children with these disorders often require additional support from families and educational systems (Wehmeyer & Thompson, 2016). Developmental delay can be defined as having a significant delay in two or more developmental domains, such as gross or fine motor, language or speech, cognitive, social or personal, and the activities of daily living, which could predict a future diagnosis of ID. The prevalence of ID is estimated to be between 1–3% (Moeschler & Shevell, 2014), making it the largest global disability group.

Intellectual disability can be caused by exogenous factors, as indicated in Figure 1, such as maternal alcohol exposure during pregnancy, infections, birth complications, and extreme malnutrition, but genetics is known to have an important role in its aetiology. Intellectual disability has become the most frequent referral to paediatric genetic clinics (Vissers *et al*., 2017). Genetic clinics consist of medical geneticists and genetic counsellors who work towards diagnosing the reason for the DD and/or ID and explaining the condition to the family. A significant proportion of individuals who come to the attention of medical geneticists and genetic counsellors as having DD and/or ID can be described as having a specific genetic condition or syndrome (Kuczynski & Udwin, 2016). Developmental delay and/or ID can have an impact on QOL of both the individual and his/her parents.

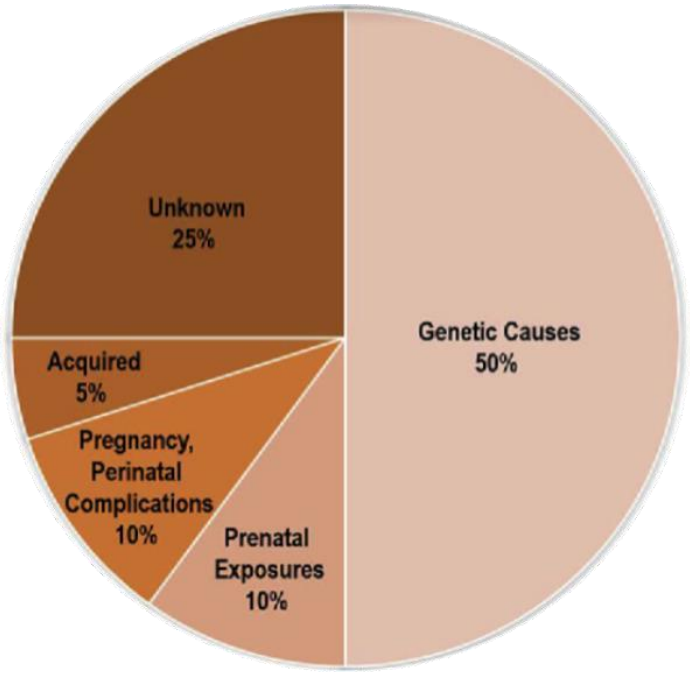


Figure 1: Causes of intellectual disability and their respective percentages (Vissers *et al*., 2017).

1.3. Quality of life definition and assessment

Quality of life is a universal concept that has different political, philosophical and health related definitions. It refers to an individual's sense of overall well-being, including physical, psychological, emotional, social, and spiritual dimensions (Cohen & Biesecker, 2010). Quality of life can be defined in many ways, which makes its measurement and incorporation into scientific research difficult (Donald, 2009). Quality of life has been noted and discussed in academic literature since the 1960's and has become more important in healthcare. Today, treatment is able to extend length of life, sometimes at the expense of QOL, not only of those affected, but also those caring for them (Karimi & Brazier, 2016).

Different approaches to define QOL exist and are mostly classified as subjective or objective. Some of the subjective definitions are: "a conscious cognitive judgement of satisfaction with one's life" and "an individual's perception of their position in life in the context of the culture and value system in which they live and in relation to their goals, expectations, standards and concerns" (Donald, 2009). An objective definition of QOL is: "an overall, general well-being that comprises objective descriptors and subjective evaluations of physical, material, social and emotional well-being together with the extent of personal development and purposeful activity, all weighted by a personal set of values" (Donald, 2009).

Quality of life is usually assessed with a questionnaire, incorporating physical, functional, psychological, emotional, social, and occupational components, whereby the individual is asked to score various questions on a number of factors that can influence QOL (Donald, 2009).

1.4. Factors that influence quality of life

Quality of life is not a single measurement, but rather a composition of a number of smaller aspects influencing the overall satisfaction with life. Expectations regarding health and the ability to cope with limitations (physical, personal, financial etc.) and disability can greatly affect a person's perception of their own health and satisfaction with life. Multiple individuals with the same health status may have very different qualities of life (Testa & Simonson, 1996). This points to the presence of other factors that influence QOL, which should also be taken into consideration when measuring QOL. Since QOL can be conceptualised as a person's wellbeing, and not just their health, there are many determinants of that well-being. Factors such as health, welfare, human rights, availability of shelter, health care, education facilities, and income must all be considered when determining QOL (Kehler, 2013).

A child with a developmental disorder presents multiple parenting challenges. For example, parents have reported heightened levels of stress, especially in domains related to taking care of children withed developmental disorders. The stress might not be because of the child's developmental disorder, but due to the increased demands resulting from the child's support needs as well as the influence of behaviour problems that might have been underestimated (Baker et.al., 2003).

1.5. Influence of having a child with a developmental disorder on quality of life

It has been suggested that parents of children with ID have higher levels of child-related stress, anxiety, and depression than parents with children who develop normally (McConnell & Savage, 2015). Caring for a child with a disability places parents at a higher risk for depression, which adversely affects their QOL. Chronic psychological distress could place parents at an increased risk of marital disruption, family dysfunction, and various mental and physical conditions (McConnell & Savage, 2015). Parents' perception of having a child with a developmental disorder, the characteristics of the parents, the internal and external resources the parents have, and the characteristics of the child are all factors that can influence the amount of pressure parents face when caring for a child with a developmental disorder. Some parents may view the situation as uncontrollably threatening, while others view the added pressure as a challenge and become stronger in the process. However the parents choose to cope will have an effect on their QOL. Parents might have to face immediate decisions about their child's medical care and treatment, along with the added financial responsibility of caring for the child (Pritzlaff, 2001).

Lower socio-economic status in low-to-middle-income countries (LMIC), such as South Africa, is related to higher parental depression and lower QOL in parents of children with developmental disorders. Children with developmental disorders are also at risk of developing other mental and physical conditions later in life. Individuals with developmental disorders experience worse health and poorer access to health care than the general population. They are vulnerable to high rates of health risks including physical inactivity, obesity, smoking and inadequate emotional support. They also experience high rates of chronic health conditions such as diabetes, high blood pressure, arthritis, chronic pain, and heart disease (Havercamp & Scott, 2015). An accurate diagnosis of mental disorders can be difficult to make in individuals with ID, because of the difficulty in communication and physical health difficulties. They may have a higher risk for psychiatric disorders that the general population and having one psychiatric disorder puts individuals with ID at risk of developing more. Schizophrenia-spectrum psychoses appear to be more debilitating among individuals with ID (Axmon *et al*, 2018). The above examples illustrate why a parent's QOL can be affected by caring for a child with a developmental disorder.

1.6. Interventions for improving quality of life

Knowing the QOL of mothers with children with developmental disorders may have many benefits. One of these benefits is improving personal care and decision making. Specific information on whether having a child with a developmental disorder impacts a woman's life can lead to better decisions surrounding her own care and what she might need to improve her QOL. Interventions surrounding resource allocation and healthcare policy can be made. All healthcare systems have to confront the reality of a finite budget and infinite demands (Donald, 2009). Interventions aimed at modifying psychosocial and contextual factors that influence QOL should also be implemented. Within the stress and coping theoretical framework, coping is the mediator between the stressor (having a child with a developmental disorder) and QOL. These coping strategies are a significant indicator of QOL. Interventions can therefore be made to mediate effective coping strategies, affecting QOL in a positive way (Cohen & Biesecker, 2010).

1.7. Quality of life in the South African setting

South Africa, as a multicultural LMIC, has a unique set of factors that must be considered when assessing the QOL of its citizens. Factors such as poverty and inequality, crime, HIV/AIDS and poor health, migration and housing, race, class and gender-based discrimination must all be considered. Challenges such as urbanisation and the housing backlog, poverty, social exclusion and welfare challenges also arise in South Africa. All these aspects have an effect on QOL (Møller, 2006). South Africa is struggling to overcome the burden of race, class and genderbased inequality inherited during the periods of colonialism and Apartheid. Previous research has shown that poor black women’s access to resources, opportunities and education, as well as access to growth and wealth of the country is severely limited. Black rural women are the ones faced with an even greater lack of access to resources and prosperity and therefore live under immense poverty (Kehler, 2013). This lack of support makes it important to understand the QOL of women who have children with developmental disorders, who will often face additional challenges. There are prevailing cultural norms that classify women as less important members of society and they mainly have a domestic role of bearing and taking care of children (Kehler, 2013). A study found that mothers of children with autism, generally report greater distress than fathers. This could be because it is traditionally the role of the woman to look after the children and thus, she would be spending more time with them (Herring et.al., 2006). It is therefore important to establish the QOL of women with children with developmental disorders, who according to cultural norms are the primary caregivers.

2. Motivation for research

To the best of our knowledge, no research has been done to establish the QOL of mothers of children with developmental disorders in South Africa. This study will provide valuable insight into the QOL of mothers caring for children with developmental disorders. By assessing the QOL of these mothers, targeted interventions can be recommended to assist them in getting the appropriate care and support to help them take care of their children. This study will help researchers and medical professionals better understand the impact of challenges that mothers of children with developmental disorders face.

3. Aims and objectives

The aim of the study is to establish whether having a child with a developmental disorder significantly influences the QOL of their mothers.

The objectives are to:

1. Compare the QOL scores of mothers of children with developmental disorders to those of mothers of children without developmental disorders
2. Determine factors associated with a good or poor QOL
3. Identify areas where interventions can be made to improve QOL

4. Participants and methods

This will be a descriptive case-control study using quantitative research methods. It will be nested within two larger studies, the first is Deciphering Developmental Delay (DDD) in Africa, that the Division of Human Genetics, the National Health Laboratory Service (NHLS) and the University of the Witwatersrand (Wits) is undertaking. The Principal investigator (PI), Professor Zané Lombard has given permission for us to recruit participants from the DDD Africa study. The second study, Long-term Effects of Gestational Diabetes, is being undertaken by The Developmental Pathways of Health Research Unit (DPHRU) within the Department of Paediatrics at Wits. Professor Shane Norris is the director of DPHRU and has given permission for us to recruit participants from his study.

4.1. Sampling

Convenience sampling will be used. Mothers of children with developmental disorders will be recruited from the DDD Africa study where data collection days will be held monthly at the NHLS in Braamfontein, Johannesburg. A development disorder gets diagnosed in a child through a developmental assessment by either a medical geneticist or neurodevelopmental doctor who are HPCSA registered and trained medical professionals. Mothers of children without a developmental disorder will be recruited from the DPHRU, in Soweto, where data collection days will be held three times a week. Cases and controls will be matched as best as possible based on the age of the mothers.

4.1.1. Sample size

The DDD Africa study is aiming to recruit 200 participants (200 mothers and their children) in 2019. This equates to ± 17 mothers per month, and therefore approximately 51 cases over a three-month data collection period. We will thus aim to recruit 30 – 50 participants in the case group. This number can be matched in the control group; the DPHRU is aiming to recruit 15 mothers per week into their study, starting in March 2019. This equates to 60 participants per month, allowing us to recruit 30 – 50 participants in the control group.

4.2. Data collection

The data collection will take place at two sites, the NHLS in Braamfontein and the DPHRU in Soweto.

4.2.1. Data collection tool

The World Health Organization Quality of Life Measure Abbreviated version (WHOQOL-BREF) will be used to measure the QOL of the mothers (Appendix B). This validated questionnaire consists of 26 questions wherein the participant chooses the answers that seems the most appropriate to them. The following broad domains will be tested in the WHOQOLBREF questionnaire: physical health, psychological health, social relationships, and the environment. The questionnaire will be anonymous, and the identity of the participants will be protected. It will be a researcher-administered questionnaire. The same questionnaire will be given to both the mothers of children with developmental disorders and the control group of mothers of appropriately developing children. Demographic and socio-economic questions have been added to the questionnaire, in order to match the mothers from the control group and the case group. A question has been added to ask whether the participants have any suggestions to improve the QOL of mothers in South Africa.

4.2.2. Controls

As mentioned previously, the control group will be recruited from DPHRU's study that will be assessing the long-term effects of gestational diabetes. The participants in the control group were originally enrolled in a research study 3 – 5 years ago through DPHRU. The study assessed pregnant women with and without gestational diabetes (Macaulay *et al*., 2018). The children of these women are now between the ages of 3 – 5 years. Together with their children, these women have been asked to return, for the new study. The children will receive a cognitive examination as part of the DPHRU study, we can thus rule out a developmental disorder in them. In addition, type 1 diabetes, congenital heart defects and epilepsy will also exclude children from the study. Regarding recruitment for the QOL study, the researcher will recruit the control participants by introducing herself to the mothers at DPHRU individually. The researcher will explain the study to each of them and ask if they are willing to participate. If they agree, they will be given an information sheet. Study participants will be interviewed after signing an informed consent form.

4.2.3. Cases

At the DDD Africa study, the researcher will approach each potential participant individually and invite them to participate. The mothers in this study all have a child with a moderate to severe developmental disorder. The participants will then receive an information sheet and an informed consent form before the questionnaire is administered.

4.3. Inclusion and exclusion criteria

There are inclusion and exclusion criteria for both the control group and the case group, as indicated in Table 1.

Table 1: The inclusion criteria and exclusion criteria

Inclusion	Exclusion
English-speaking ≥ 18 years of age English literate South African Must have a child between 1 – 5 years of age	Mothers whose children are critically ill Mothers with more than one child with DD and/or ID Pregnant woman Other caregivers who attend the clinic with the child Controls with children with DD and/or ID

4.4. Data analysis

Once the data has been collected, it will be organised into separate categories so that the QOL of the participants will be thoroughly investigated. Categories will include:

- Severity of the developmental disorder (highly severe to moderately severe as classified by the DDD Africa project)
- Socio-demographic categories (ethnicity, race, age)

The data will be captured in a RedCap database created for this study, and descriptive statistical analysis using Stata version 15 will be performed. For the continuous data, normally distributed data will be presented as means and standard deviations and non-normally distributed data will be presented as medians and ranges. Categorical data will be presented as frequencies and percentages. Tests such as the chi-square test will be used to assess the difference between categorical variables, between cases and controls, the student t-test and Mann-Whitney test will be used to assess the difference between continuous variables between cases and controls.

Linear regression analysis will be used to determine predictors for good or poor QOL scores. Predictors will be variables that are shown to be significantly different between cases and controls. The outcome variable will be the QOL score (which will be kept continuous).

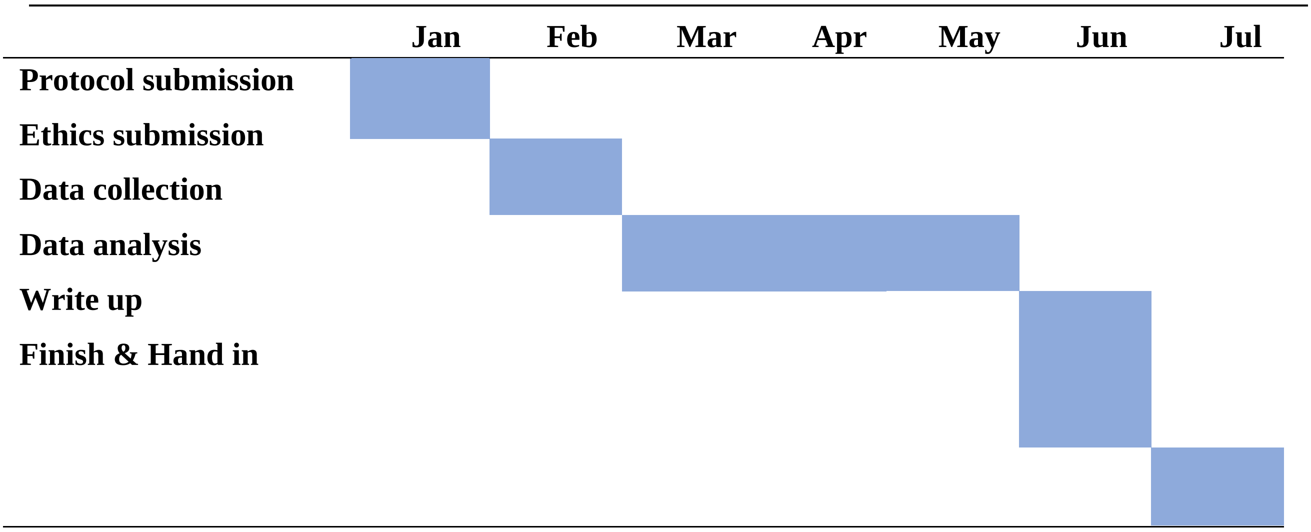
6. Ethical considerations

Ethical clearance has been granted for the DDD Africa and Long-term Effects of Gestational Diabetes study by the Human Research Ethics Committee (HREC) (Medical) of the University of the Witwatersrand; M180678 and M180316 respectively.
The researcher will apply for ethics clearance for the QOL study through the HREC at the University of the Witwatersrand.

The anonymity of the mothers will be protected. Before the questionnaire is administered, participants will be asked to sign an informed consent form, which will be discussed with them by the researcher. Should they wish to drop out of the study, they are free to do so without fear of their decision affecting the care of them or their child in future. Participants will be asked whether they would like to be contacted should their QOL score be poor. They will be given the option of leaving their name and contact details after completing the questionnaire. They will then be contacted and seen for a counselling session by one of my supervisors at a genetic clinic and referred as appropriate. Should a poor QOL score or any signs indicate that the mother is not coping, or is distressed, my supervisors, who are trained HPCSA registered genetic counsellors will be available to intervene and refer for further management and help as appropriate, at no cost to the participant. A network of referrals to psychologists, psychiatrists and social workers is already in place and being used by the Genetic Counsellors of the Clinical and Counselling Section at the NHLS/Wits.

7. Timing

2019



8. Funding

I will apply for the Faculty Research Committee grant, otherwise the funding will be provided by the Division of Human Genetics. Travel costs by the DDD participants will be covered by the DDD Africa project. The controls will be funded by the DPHRU.

Table 2: Estimated budget

Description	Cost
Print out of questionnaires forms	39c x 700 = R 273.00
Travel to clinics	36 km x 2 x 12 x R 1.05 = R 907.20
Total	**R 1180.20**

9. Potential limitations

As with all research studies there are some limitations to this study. These include:

-The questionnaire will be administered in English and will not be translated. -There are language barriers that exist between the researcher and the participants, as the research will by necessity be conducted in English, which will be a second language for most participants.

-There are some time restraints inherent in completing research for degree purposes. -We are only looking at mothers and the research will not be representative of all parents.

-There is no information on whether the mother has been diagnosed with a mental illness, such as a psychological/psychiatric disorder or undiagnosed mental illness.

-Maternal stress on a cofounder is not being evaluated.

10. References

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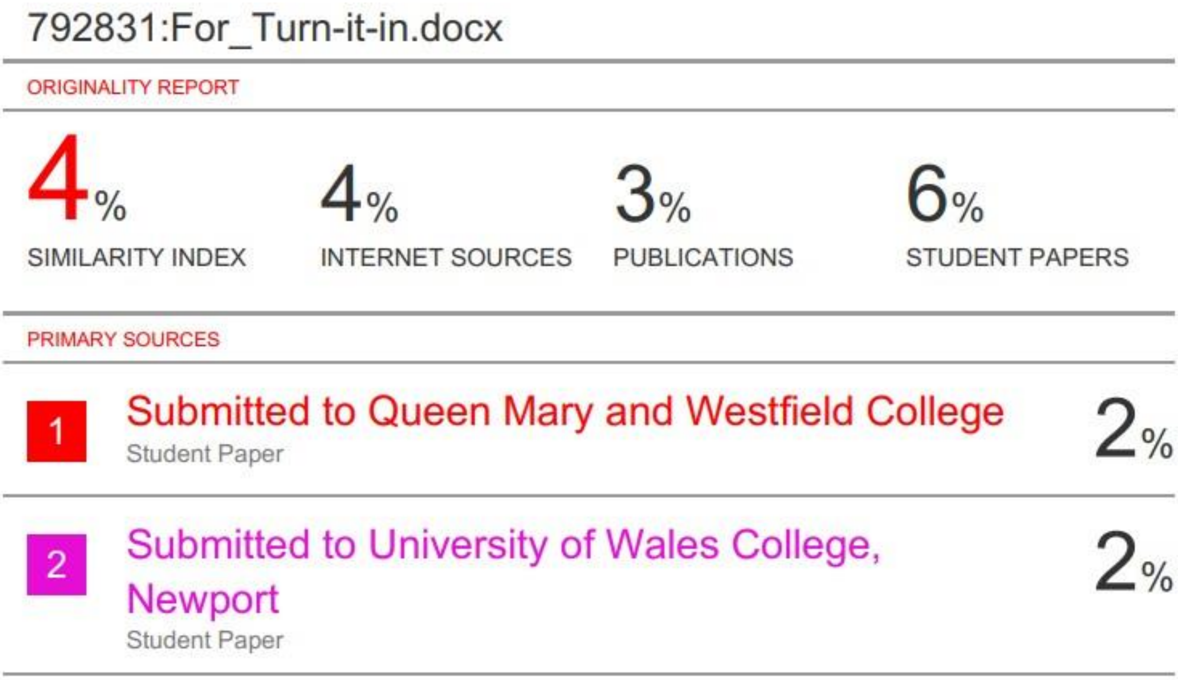
Appendix E: Turn-it-in report



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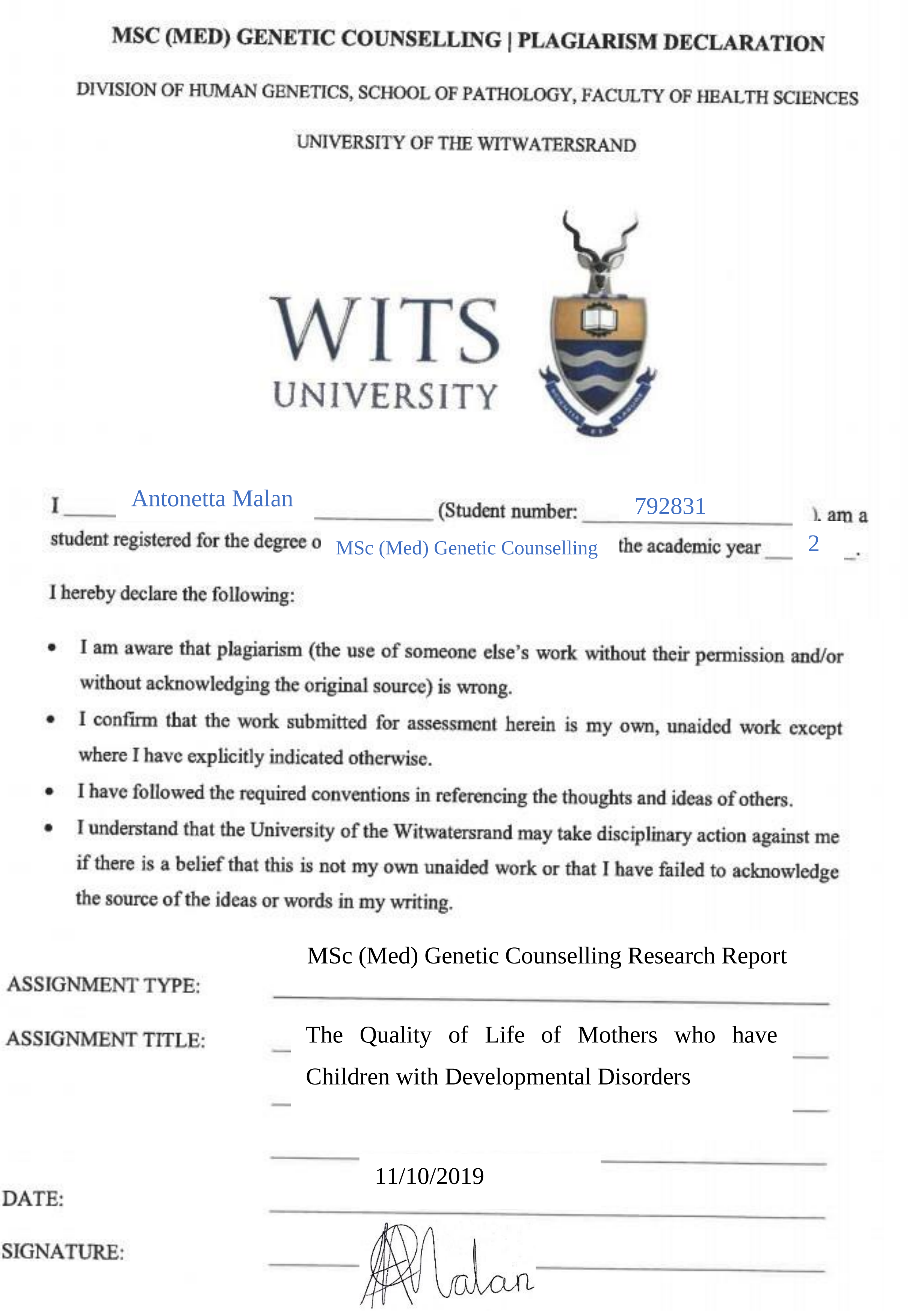
Appendix F: Signed plagiarism declaration



MSC (MED) GENETIC COUNSELLING | PLAGIARISM DECLARATION

DIVISION OF HUMAN GENETICS, SCHOOL OF PATHOLOGY, FACULTY OF HEALTH SCIENCES

UNIVERSITY OF THE WITWATERSRAND

WITS UNIVERSITY

I ____ Antonetta Malan ________ (Student number: 792831 __________). am a student registered for the degree o MSc (Med) Genetic Counselling the academic year __ 2 _.

I hereby declare the following:

- I am aware that plagiarism (the use of someone else's work without their permission and/or without acknowledging the original source) is wrong.
- I confirm that the work submitted for assessment herein is my own, unaided work except where I have explicitly indicated otherwise.
- I have followed the required conventions in referencing the thoughts and ideas of others.
- I understand that the University of the Witwatersrand may take disciplinary action against me if there is a belief that this is not my own unaided work or that I have failed to acknowledge the source of the ideas or words in my writing.

ASSIGNMENT TYPE: MSc (Med) Genetic Counselling Research Report

ASSIGNMENT TITLE: The Quality of Life of Mothers who have Children with Developmental Disorders

DATE: 11/10/2019

SIGNATURE: AMalan