

# **Tuberous Sclerosis Complex: Measuring Parental Stress, Depression and Quality of Life.**

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### Abstract

Tuberous Sclerosis Complex (TSC) is a hereditary disorder which causes benign non-invasive lesions on any organ within the human body. Roughly 85% of children who are affected with TSC develop behavioural, neuropsychological, intellectual and psychosocial problems. There is very little literature regarding the difficulties of parents who have children diagnosed with TSC. Currently, the Red Cross War Memorial Children's Hospital contains the only TSC clinic in sub-Saharan Africa and hence is the main source of information regarding the disorder. Due to the unpredictable nature of the disease and the difficulty associated with providing parents with a clear clinical picture it is likely that parents of children with TSC experience higher stress levels, more symptoms of depression and a lower quality of life. This study compared parental stress scores, depression scores and quality of life scores of parents of children with TSC ( $n = 15$ ), parents of children with autism spectrum disorder (ASD) ( $n = 15$ ) and parents of typically developing (TD) children ( $n = 12$ ). In line with hypotheses, results showed that parents of children with TSC reported the highest parental stress scores across all three groups. Additionally, parents of children with TSC reported lower quality of life scores than parents of children with ASD. However, this study found no group differences in depression scores. A limitation of the study is that these findings may not be accurate and applicable to the entire population due to the small sample size.

*Keywords: autism spectrum disorder, depression, parental stress, quality of life, tuberous sclerosis complex*

## **Literature Review**

### **Parenting a child with Tuberous Sclerosis**

The handbook of child neurology (Curatolo & Maria, 2013) describes Tuberous Sclerosis Complex (TSC) as a hereditary disorder which causes benign non-invasive lesions on any organ (Curatolo & Maria, 2013; De Vries, Hunt, & Bolton, 2007). Approximately 85% of children and adolescents affected with TSC go on to develop behavioural, neuropsychological, intellectual and psychosocial problems (Curatolo & Maria, 2013; De Vries et al, 2007; Kingswood, 2015). Furthermore, sleep disorders and anxiety and mood related disorders are also commonly associated with TSC. In addition, physical manifestations which typically present in the cutaneous, renal, cardiac, ophthalmic and pulmonary regions could result in further medical difficulties for the child. Due to the unpredictable nature of the disease and the consequential difficulty in providing caregivers with a clear clinical picture, it seems likely that parents of children with TSC will experience a greater level of concerns, anxieties and stress than parents of typically developing (TD) children. Additionally, research in this field has shown that parents of children with developmental disorders such as Autism Spectrum Disorder (ASD) face greater stress and anxiety levels than parents of typically developing children (Gupta, 2007). It is not clear whether parents of children with TSC would be as affected, or more affected than these parents, given the unpredictability of the TSC disease process. Therefore, this study seeks to investigate the parental stress, depression scores and quality of life of parents raising a child with TSC in comparison with those raising TD children or children that have been diagnosed with ASD.

### **Parenting a child with Special Needs**

Literature on parenting difficulties in TSC specifically is sparse, but there is evidence of increased demands and parenting stress when looking more generally at raising children with special needs. For example, a study by Leiter, Krauss, Anderson and Wells (2004) focusing on maternal caregiving, suggested that being the main caregiver of an adolescent with special needs includes a range of unique caregiving requirements and parenting techniques which differ from those necessary for typically developing (TD) children. Such requirements may involve specific knowledge of the disorder, cooperation and partnership with a wide range of health professionals. These requirements would result in parents acquiring disease specific practices in order to care for their children. One such example is given in the paper. The study found that mothers of children with sickle cell disease

frequently developed amateur caregiving tactics such as those that would generally be provided by nurses or other healthcare professionals in order to fill gaps in medical assistance. Another example of this is parents of children whose illness or disorder requires the use of complex equipment - parents are obliged to learn how to operate this equipment. These techniques or requirements are specifically tailored to the child based on the intensity and complexity of the disease. This results in an increase in time and energy needed for caregiving, consequently creating greater parental stress (Leiter, Krauss, Anderson, & Wells, 2004; Gupta, 2007). Such was evidenced by Gupta (2007), who revealed that caregivers of children and adolescents with disruptive behaviours (e.g. Attention Deficit-Hyperactivity Disorder) and developmental disabilities (e.g. ASD) reported greater levels of stress than parents of children and adolescents with life-long medical illnesses like HIV and asthma.

Furthermore, a review of the literature on the occurrence of maternal despair in families with at least one child with a disability found that in fourteen studies (of seventeen), mothers crossed the threshold for depressive indicators on a self-report scale (Roberts, 1980). The review further indicated that sixty-five percent of seventeen studies revealed that higher rates of depressive symptoms were reported by mothers of children with disabilities compared to controls. Fourteen studies in the review also included paternal caregivers and compared depression status/symptoms with their counterparts. Nine of the studies found that maternal caregivers had greater depressive indicators than fathers, while the remaining five found little to no observable differences in depressive symptoms between maternal and paternal caregivers of children with special needs.

A more qualitative perspective is provided by a meta-synthesis study which was done with 11 parents of children with disabilities. It found seven recurring thematic metaphors which outline and assisted in explaining their lived experiences. Some of the themes reported were: 'living worried,' 'carrying the burden,' 'survival as a family' and 'staying in the struggle' (Coffey, 2006). The parents in the study explained that parenting a child with a disability meant that they spent their lives in a constant state of worry about their child's condition, even on good days; or they were worried that that their child's condition may worsen. They were concerned about their family as a whole and sometimes felt inadequate as parents because their attention would be so heavily concentrated on one child that they feared neglecting their other children (Coffey, 2006). Such results emphasise the necessity for services and support structures to assist caregivers in their parental abilities as well as in addressing their additional anxieties, concerns and stressors.



Loyd and Rosman (2005) revealed that reasons for increased stress include problems in obtaining suitable childcare, as well as in sustaining a consistent employment schedule. This stress is more evident in families who are low on resources such as time, money and adequate support systems, as it was found that in lower socio-economic contexts, mothers of children with special needs showed greater levels of stress than mothers of TD children (Loyd & Rosman, 2005).

A study done by Whitemere & Lewis (2010) found three effects on parents and caregivers of children with TSC in particular. The first is that parents of children with TSC are concerned about the effects that caring for child with a long term illness would have on their marriage. Another effect is that they may become so engrossed in caring for their child, that they neglect their relationships with others, including other children and spouses. And finally, the study noted that caregivers of children with TSC were at a higher risk for marital distress.

As these studies were conducted in international populations it is important to note that not much emphasis is placed on the effects that SES has on parental stress and quality of life. Many of the studies reviewed in this paper were conducted in American populations (Coffey, 2006; Gupta, 2007; Loyd & Rosman, 2005 & Whitemere & Lewis, 2010) and as South Africa is a low middle-income country and the United States is a high-income country (World Bank), we can assume that low-income parents in South Africa might face more difficulties raising disabled children than low-income parents in the United States, for example.

### **Parenting a Child with Autism Spectrum Disorder**

Factors that would impact parental stress in parents of children with ASD have been well documented, especially in developed contexts. Autism Spectrum Disorder is described as a pervasive developmental disorder (PDD) which is categorized by the early onset of substantial deficiencies in social contact and communication in addition to stereotyped behaviours (Allik, Larsson, & Smedje, 2006; Schieve, Blumberg, Rice, Visser, & Boyle, 2017). Previously, it was assumed that ASD only occurred in western countries, yet knowledge pertaining to ASD and its increasing prevalence has been recorded in various regions globally over the last ten years (Unit, Hospital, Haven, & State, 2011). Bakere & Munir (2011) noted that much of the available literature pertaining to ASD has been produced in western contexts. Thus, there is a large gap in the body of knowledge in relation to ASD in an African context.

Children who are diagnosed with ASD often face behavioral and developmental challenges which require various therapies in diverse disciplines (e.g speech therapy, occupational therapy etc.) (Schieve et al., 2017). Whilst a certain amount of stress is deemed natural when raising a child, research has shown that parents of children with ASD experience higher levels of stress due to the wide-ranging behavioural difficulties that their children display (Mcstay et al 2014; Pottie, Cohen, & Ingram, 2017). Difficulties associated with raising a child with ASD include, but are not limited to, factors such as difficulties communicating with their children, challenging behaviours, challenges associated with self-care, social seclusion and the absence of community understanding (Schieve et al., 2017). A study conducted by McStay et al (2014) hypothesized that parents of children with ASD would experience significantly higher stress levels than parents of TD children. This hypothesis was supported by their findings. In addition, this study found no significant difference in parental stress levels across various age cohorts. Moreover, the aforementioned study found that verbal ability and the severity of symptoms had significant bearing on parental stress levels. Furthermore, a study done on parents of children with Aspergers Syndrome (AS) and High Functioning Autism (HFA; as defined in DSM-IV-TR) found that these parents reported weakened physical health when compared to parents of children with disabilities such as cerebral palsy (Allik et al., 2006).

According to Pottie, Cohen, & Ingram (2017) care-givers of children with ASD experience greater levels of parental stress than parents of TD children. It was argued that this was largely due to a lack of social support. In this context, social support can be described as a multidimensional expression which encompasses an individual's social milieu. These social settings, or support systems often serve to enhance the psychological and physical well-being of the individual. This study found that parents who had strong emotional support reported greater levels of positive mood daily.

Parents of children with ASD or TSC would face significant parenting difficulties due to the unpredictable and varying nature of both of these disorders. Thus, it would be worthwhile to investigate whether parents of children with TSC face similar or worse strains than parents of children with ASD. This becomes particularly essential in a South African context where low SES is common amongst a large portion of the population.

### **Socio-economic Status and Parental Stress/Quality of Life**

In a study done in the informal settlement of the Orange Farm, South Africa, Saloojee, Phohole, Saloojee, & Ijsselmuiden, (2007) found insufficient resources and services available to assist children with disabilities. Additionally, the study found that the flexibility

necessary to prioritize the needs of a child that has a disability is lower when parents live in a low socio-economic area. This stress is further aggravated in areas with low employment rates and little to no external support from the child's father or other family members (Saloojee et al., 2007).

In the case of TSC, the current treatment and management is symptomatic, essentially occurring on a 'bit-by-bit' basis, treating the symptoms as they manifest, rather than providing a permanent cure (Curatolo & Maria, 2013; Samia, Donald, Schlegel & Wilmshurst., 2015). Currently the Red Cross War Memorial Children's Hospital contains the only TSC clinic in sub-Saharan Africa and is consequently the main source of information regarding the disorder. This further highlights the lack of information resources available to parents in South Africa, including the Western Cape (Samia, Donald, Schlegel, & Wilmshurst, 2015). Not much is understood about the everyday needs of caregivers of children that have disabilities in low and middle class socio-economic areas (Curatolo & Maria 2013). Whilst there is an abundance of global literature stating that parents of special needs children encounter more significant proportions of stress and depression than parents of TD children, not much focuses on the specific needs and complexities of TSC. Given that TSC is chronic, children affected by it will require long term care which may include numerous visits to the doctor, various therapies and long term medications to treat conditions such as epilepsy. Additionally, with the unpredictable nature of TSC the lesions could form anywhere within the body. Thus it is difficult to provide parents with a clear picture of what to expect throughout their child's lifetime, ultimately handicapping their ability to cope (Samia, Donald, Schlegel & Wilmshurst., 2015). When going through the literature available it became clear that not many studies have been done on the impact that caring for a child with TSC would have on parents. Following that, it is also important to take into account how heightened parental stress and depression would affect the overall health of the child. Therefore, this study will address the gap in the literature by focusing specifically on the stress and quality of life of parents of children with TSC. Furthermore, given that the majority of the South African population live below the poverty line and that lower socio-economic status (SES) has shown to increase stress in parents of children with special needs (Lloyd and Rosman., 2005), it is important to investigate stress experienced by parents of children with TSC and ASD in South Africa. Both groups of children may manifest with severe behavioural and psychosocial difficulties, but TSC children have additional and unpredictable medical problems. It is thus of interest to compare parent stressors across these groups.

## **Rationale and Aims**

A good support structure consisting of a healthy and physically able caregiver is important in the treatment process of any illness (Pottie, Cohen, & Ingram, 2018) as long term care has the potential to become strenuous on the caregiver. Owing to the fact that there is currently no known cure for TSC, the existing treatment focuses on the management of symptoms as they appear (Curatolo & Maria, 2013). Thus, children of parents with TSC have the added stress of unexpected medical emergencies and expenses. As discussed in the literature review, few studies have focused on the needs and the strain that caregivers of children with TSC undergo. Moreover, there is a lack of information pertaining to South African families in particular. Thus, not much can be gained by looking at the global literature for a South African context as South Africa is still a developing country and majority of the research has been done in first world countries. Moreover, access to healthcare and information regarding TSC is a major concern as Red Cross Children's hospital is currently the only TSC clinic in South Africa.

Based on the above rationale, this study aims to address the current gaps in the literature by assessing the parental stress, depression scores and quality of life of parents of children with TSC and ASD, whilst comparing them to a TD parent control group. It is of interest to examine similarities and differences between experiences of parents raising children with these two very challenging disorders. The abovementioned phenomena will be examined through the use of self-report questionnaires, thus, adding to the body of literature on TSC in the hopes that it would lead to the betterment of the support structures that are currently available to the families. This study forms part of a larger study investigating the cognitive, behavioural and psychological profile of children with TSC. Results from the larger investigation and current research study will aid in creating individualised interventions in the future, as well as support groups for both the caregivers and children.

## **Hypotheses**

1. Parents of children with TSC will report high parental stress scores. I hypothesize that parental stress will be greatest in TSC, then in ASD and lowest in parents of TD children.
2. Parents of children with TSC will score higher on depression scales. I hypothesize that depression scores will be highest in the TSC parenting group, followed by the ASD parenting group and lowest in the TD parenting group.

3. Parents of children with TSC will report lower quality of life scores. I hypothesize that parents of TSC children will report the lowest quality of life scores, followed by the ASD parenting group, with the TD parenting group reporting the highest quality of life scores.

## Methods

### Research design

This study formed part of a larger on-going investigation into the cognitive and behavioural profiles of children with TSC, and the difficulties faced by caregivers. Thus the study had already been granted ethical approval and data collection had been on going. This sub-study placed specific focus on the effects that raising a child with TSC has on parents. The study is cross-sectional, quantitative and quasi-experimental, comparing parents of children with TSC with those raising children with ASD, along with a control group of parents raising TD children. This study made use of three questionnaires to determine and compare parental stress levels, depression scores and quality of life scores across these groups. A purposive sampling technique was used to recruit TSC cases. The ASD data was collected from an already existing dataset that was made available for the purpose of this study. Typically developing controls were recruited from one of three primary schools that consented to participate in the study. Three key socio-demographic factors (SES, child age and home language) were used to case match TSC participants with ASD and TD parent groups. Other factors such as, number of siblings and child gender were also taken into consideration and matched as closely as possible.

### Participants

In order to establish the sample size needed to find an effect, *a priori* power analysis was conducted using G\*Power 3.10. Owing to the fact that no previous studies exist where a similar method is employed, standard values were used to determine a sample size. A large effect size was estimated for the ANOVA as a review of the literature indicated that parents of children with TSC would report higher stress levels. Thus, taking  $f = .60$ ,  $\alpha = .05$ , and  $\beta = .95$ , a total sample size of  $n = 48$  was suggested.

The primary caregivers of children with TSC have been recruited for the larger investigation and formed part of this sub-study's sample. The participants were from varying socio-economic backgrounds within the Western Cape. Thirty families currently attend the TSC clinic at Red Cross Children's hospital. These families were approached to participate in the study, however, not all of the families consented to take part in the study. Due to the fact that data collection occurs on the day of the participant's clinic appointment and families only attend the clinic every three months, the study reported here managed to recruit and complete only fifteen TSC participants.

Parents of children with ASD and TD children were recruited and case matched to a TSC participant on three variables (SES, child age and home language). Thus, fifteen ASD participants were recruited, however, due to difficulty case matching variables only thirteen TD participants were recruited. Thus, the total sample size was 43, slightly lower than the 48 indicated by the power analysis'.

The eligibility criterion for the parent participants in all three groups was that they must reside with and look after the child. Eligibility criterion for the ASD parenting group was that the child had to have been diagnosed with ASD. TD participants were excluded from the study if the child had previously suffered a head injury or any neurological or psychiatric illness.

## **Measures**

**General measures.** After completing the informed consent form (see Appendix A, B and C), participants were asked to complete a socio-demographic questionnaire (see Appendix D). The socio-demographic questionnaire obtained information specific to the participant (e.g. name, age, sex, SES), as well as imperative exclusion criteria pertaining to the child (e.g. head injury, psychiatric illness or neurological disorder). As this sample consisted of first language isiXhosa and Afrikaans as well as English speakers both of the afore-mentioned forms were translated and back translated into the respective languages of the participants.

**Parent Questionnaires.** This study employed three questionnaires to assess parental stress, depression, and quality of life across the groups. A translator was made available at all times to assist the participants to answer the questionnaires in the language of their choice (isiXhosa or Afrikaans) as these questionnaires had not yet been translated into the respective languages.

***Parenting Stress Index Short Form (PSI-SF).*** The Parenting Stress Index Short Form (PSI-SF) is a Likert-type self-report survey which consists of 54 parent-focused questions and 47 child focused questions (Reitman, Currier, & Stickle, 2002). The parent section has seven subsections which measure depression, attachment, role restriction, sense of competence, social isolation, relationship with spouse, and parental health. The section which is focused on the child consists of six sections which measure levels of adaptability, acceptability, demandingness, mood, distractibility/hyperactivity, and reinforces parent (Reitman, Currier & Stickle, 2002). A study done by Shochet, Dadds, Ham and Montague (2010) sought to assess the validity of the PSI-SF. The investigation found that the scores of the PSI-SF remained constant over the span of a year (Shochet, Dadds, Ham, & Montague,

2010). Moreover, the PSI-SF has been used successfully in a South African context (Wallace, Franzsen & Potterton, 2016).

***Centre for Epidemiologic Studies Depression (CES-D).*** The Centre for Epidemiologic Studies Depression (CES-D) scale is a self-report scale comprised of twenty questions (Roberts, 1980). Research has shown that the CES-D scale has excellent internal consistency and very good test-retest stability (Roberts, 1980). Again, no studies focusing on TSC which used this measure could be found, however many studies relating to depression in the cancer population were found. One such study found that the CES-D exhibited outstanding internal consistency (Hann, Winter, & Jacobsen, 1999). Moreover, the CES-D has been used successfully in numerous studies in South Africa, particularly in HIV studies, (Myer, Smit & Roux et al, 2008; Joska, Westgarth-Taylor & Myer et al, 2011; Futterman, Shea & Besser et al, 2010).

***World Health Organization Quality of Life B.R.E.F (WHOQOL-BREF).*** The WHOQOL-BREF consists of 26 questions that represent four major domains of quality of life, namely, physical, social, psychological and environmental (Skevington, Lotfy, & O'connell, 2004). The WHOQOL-BREF was developed as an abridged version to the WHOQOL assessment, which is a multicultural evaluation comprised of 100 questions representing six major domains of quality of life. The World Health Organization (WHO) describes quality of life as the individual's insight into their status in life within the context of their culture and denomination. Whilst no studies relating to the validity of the WHOQOL-BREF could be found with regards to TSC, one study was found that investigated the validity of the assessment in patients with traumatic spinal cord injuries. This study found neither a floor nor ceiling effect in the WHOQOL design (Jang, Hsieh, Wang, & Wu, 2004). A broad look at the literature found that the WHOQOL-BREF has been utilized in many studies relating to quality of life in HIV patients in South Africa, showing that the test can be administered within a South African context and population (Peltzer, Friend-du Preez, Ramlagan, & Anderson 2010).

## **Procedures**

As this study is part of a larger investigation, ethical approval had already been granted and data collection had been on-going. Before the data collection begun, participants were given information and consent forms (Appendix A, B and C) which outlined the purpose and procedure of the study. Participants were then asked to fill out the consent forms and socio-demographic questionnaires, followed by the various questionnaires.



The data was collected at RXH on the day of the respective TSC participant's appointment at the TSC clinic. ASD data was collected from an already existing data set which was made available for the purpose of this investigation. The control group completed the questionnaires at the participating school with the aid of the researcher. In the event that the questionnaires were not completed in one session the researcher continued the interview telephonically. If any of the participants became fatigued at any point, they were encouraged to take a short break.

### **Ethical Considerations**

This study formed part of a larger study titled *Determining the Neurocognitive and Psychological Profile of an established group of Children with Tuberous Sclerosis Complex within the Western Cape Region of South Africa*. The larger study had received ethical approval from Red Cross Children's Hospital Research Committee (School of Child and Adolescent Health Division Research Committee; see Appendix E) and UCT Faculty of Health Sciences Human Research Ethics Committee (See Appendix F); note this approval includes both the TSC clinical group and matched controls. The sub-study sought additional approval from the UCT Department of Psychology Research Ethics Committee (See Appendix G). Moreover, ethical approval for the ASD parent group was granted by the UCT Faculty of Humanities (see Appendix H). Additionally, the procedures that have been delineated in this study were done in accord with the Declaration of Helsinki (2013) and the UCT guidelines for research on human subjects.

**Consent, voluntary participation and confidentiality.** Prior to the study taking place all participants were asked to give informed consent. Participants were free to ask any questions that they had related to the objectives of the study. The identity of the participants and their data has been kept strictly confidential and only the researchers working on the project have had access to the data. Once the data intake was completed and the data was being compiled electronically each participant was given a unique identification number. Participants were free to withdraw from the study at any point in time without fear of any repercussions. TSC participants who chose to withdraw from the study will still receive their standard care at RXH.

**Risk and benefits.** The study poses no risk to the participants and is completely voluntary. There were no costs incurred by the participants by partaking in this study. Participants were free to take breaks from questionnaires should they become fatigued. As this study is nestled within a larger study, TSC families will receive overall feedback from

the study as a whole which is inclusive of feedback from both my section of the study and the larger investigation. Families of children with TSC will receive more detailed feedback from Natalia Berghoff with regards to the intervention strategies that will be employed for the individual families and their child's cognitive and psychological strengths and weaknesses. In addition to the one-on-one feedback, families will also receive a written report which summarises the discussion in the feedback session. As parents they will have a better understanding of themselves and of their children as their participation in the larger investigation will allow them to learn more about their child's cognitive and psychological abilities.

ASD and TD families did not receive in-depth feedback as these participants were unique to this sub-study investigation and did not form part of the larger study. They will, however, receive general feedback about the study findings. Furthermore, where parental stress and depression scores were high, ASD and TD families were encouraged to pursue supplementary professional assistance and advice. ASD and TD families will not benefit directly from their participation in this study but their participation will aid in adding to the body of knowledge surrounding TSC in South Africa.

**Costs and compensation.** There were no additional costs sustained to RXH, UCT, researchers or participants in the study. However, if data collection took place on a day that is not on the TSC participants' scheduled clinic date they received R100.00 travel compensation. Data collection for the ASD group had already been collected prior to this investigation. TD data was collected at the participating school and most of the families lived within walking distance of the school, thus no travel costs were incurred.

### **Data Analysis**

This study made use of IBM SPSS (v23) in order to process and analyse the data. Descriptive statistics were first examined and reported. Furthermore the data was examined to see if parametric assumptions had been upheld – several variables were not normally distributed (See Appendix I). Due to this non-normality, and the small sample size, bootstrapping methods which based parameter estimates on 1000 repeated samples (Field, 2013) were used for analysis.

A series of one-way ANOVAs were used to assess for group differences of the parental stress scores, depression scores and quality of life scores across the TSC, ASD and TD groups. Where indicated, posthoc pairwise contrasts were performed using the Games-Howell method. Significance levels were set at the conventional  $p < .05$ .

## Results:

### Socio-demographic Information

Participants were case matched on socio-economic status (SES), child age and home language. . However, given that there was a relatively small sample size, additional case matching on child gender and number of siblings was not possible for every case, although these were matched as closely as possible. A summary of the socio-demographic information can be found in Table 1.

Table 1

#### *Summary of Socio-demographic Information*

Variable	TSC ( <i>n</i> =15)	ASD ( <i>n</i> =15)	TD ( <i>n</i> =12)	<i>F</i> / $\chi^2$	<i>p</i>
SES	34 736.25 (54410.60)	130 833.33 (185 635.94)	41 107.22 (39 623.38)	2.40	.107 <sup>a</sup>
Child Age	9.64 (3.73)	9.27 (3.71)	9.18 (3.52)	.06	.943
Number of Siblings	1.93 (1.14)	.87 (.74)	1.73 (1.90)	2.78	.075
Home Language					
Eng/Afr:Afr:Eng:isiXhosa	1:3:8:2	1:1:11:2	1:3:7:1	b	
Gender:					
Male:Female	6:8	12:3	4:8	6.84	.033*

*Note:* Means with standard deviations in brackets reported

a: Note extreme variability around mean for ASD group; for ns posthoc pairwise contrasts see Appendix J

b: Could not perform  $\chi^2$  because 75% cells have an expected count < 5.

\* Significant at  $p < .05$ ; more boys than expected in ASD group

The groups were successfully matched on the key variables, i.e. SES, child age and home language. A significant difference was not found between the groups on the number of siblings within the family. The group differences in home language could not be investigated due to very small cell sizes, however, majority of the parents were English speaking. A significant difference on gender was found across the parent groups. There were a higher number of boys in the ASD group: this can be explained by the fact that there is a higher prevalence of ASD amongst boys (Nicholas, Charles & Carpenter et al, 2008).

### **Parental Stress, depression and quality of life**

A series of one-way ANOVAs were run to examine the impact of group (TSC, ASD, TD) on the outcome variables, parental stress, depression and quality of life (See Table 2). Given the small group sizes within our sample which were a result of difficulties recruiting TSC participants and matching across groups, the data on the variables of interest was not normally distributed. (Figure 1, Appendix I). In order to obtain more reliable statistical results bootstrapping was applied. A simple method was employed, basing parameter estimates on 1000 repeated samples from the data (Field, 2013).

Table 2.

#### *Parenting stress, depression and quality of life*

Variable	TSC ( <i>n</i> = 15)	ASD ( <i>n</i> = 15)	TD ( <i>n</i> = 12)	<i>F</i>	<i>p</i>
Parental Stress	129.86 (23.44)	104.67 (16.06)	109.92 (39.78)	3.37	.045*
Depression Scores	15.36 (14.32)	21.33 (11.51)	21.00 (18.29)	.727	.490
Quality of Life	54.67 (9.62)	500.80 (1669.15)	54.08 (9.50)	117.94	<.0001**

*Note:* Means with standard deviations in brackets reported

\**p* < .05

\*\**p* < .0001

When examining the between group differences of the parental stress scores, the ANOVA was statistically significant,  $F(2,38) = 3.37, p = .045$ . A summary of these findings can be found in Table 3. The post-hoc Games-Howell tests found a significant difference between the TSC and ASD ( $M = 104.67; SD = 16.06$ ) parenting groups, with TSC parents reporting significantly higher stress scores ( $M = 129.86; SD = 23.44$ ). There was no significant difference in parental stress scores between parents of TSC children and parents of TD children, or between parents of ASD and TD children. A summary of the posthoc contrasts can be found in Table 4.

Table 3.

Summary of ANOVA results: Parental Stress Scores

	SS	df	MS	F	p
Between Groups	5000.48	2	2500.24	3.37	.045
Within Groups	28157.96	38	741.00		
Total	33158.44	40			

Table 4.

Games-Howell posthoc pairwise contrasts

Parental Stress Scores

		Mean Difference		
(I) Label	(J) Label	(I-J)	Std. Error	p.
TSC	ASD	25.19*	7.51	.008
	Control	19.94	13.08	.304
ASD	TSC	-25.19*	7.51	.008
	Control	-5.25	12.21	.904

Note: \*. The mean difference is significant at the 0.05 level.

A one-way ANOVA was conducted to investigate the between group differences between the TSC, ASD and TD parent groups' depression scores. The results show that the ANOVA was not significant  $F(2,38) = .727; p = .490$ . Thus, no significant difference was

found in depression scores across the three groups. A summary of the Depression scores ANOVA is given in Table 5.

Table 5.

*Summary of ANOVA results: Depression Scores*

	<i>SS</i>	<i>df</i>	<i>MS</i>	<i>F</i>	<i>p</i>
Between Groups	313.89	2	156.95	.727	.490
Within Groups	8200.55	38	215.80		
Total	8514.44	40			

A one-way ANOVA was run to examine the between group differences between the quality of life scores across all three groups. The results of the ANOVA were significant,  $F(2,39) = 117.94$ ,  $p < .0001$ . A summary of these findings is given in Table 6. Post-hoc tests found that the TSC ( $M = 54.67$ ;  $SD = 9.62$ ) parent group differed significantly from parents of children with ASD ( $M = 500.80$ ;  $SD = 1669.15$ ), with ASD parents reporting a better QOL. The post-hoc test also found that parents of children with ASD ( $M = 500.80$ ;  $SD = 1669.15$ ) reported significantly better quality of life than parents of TD children ( $M = 54.08$ ;  $SD = 9.50$ ). A summary of these findings can be found in Table 7.

Table 6.

*Summary of ANOVA results: Quality of Life Scores*

	<i>SS</i>	<i>df</i>	<i>MS</i>	<i>F</i>	<i>p</i>
Between Groups	235929856.30	2	117964928.20	117.94	<.0001
Within Groups	39007128.65	39	1000182.79		
Total	274936985.00	41			

Table 7.

*Games-Howell posthoc pairwise contrasts*

## Quality of Life Scores

(I) Label	(J) Label	Mean Difference (I-J)	Std. Error	p
TSC	ASD	-4946.13*	430.98	<.0001**
	Control	.58	3.70	.986
ASD	TSC	4946.13*	430.98	<.0001**
	Control	4946.72*	430.98	<.0001**

*Note:* \*. The mean difference is significant at the 0.05 level.

\*\* $p < .0001$

### **Examination of associations between key demographic variables and parental stress and quality of life**

Whilst there was no significant difference on SES between the three groups, the descriptive statistics look slightly uneven (see Table 1.), with the mean SES in the ASD group seemingly much higher than the other groups. This lack of difference across SES is likely due to the large degree of variability in SES in the ASD group. The literature indicates that low SES can have an impact on parental stress levels (Saloojee et al., 2007).

Moreover, no significant difference was found on the number of siblings in the family across all three groups (see Table 1), yet descriptively this data seems uneven as the TSC parenting group reports a higher number of siblings.

Thus, bootstrapped pearson correlations were conducted to determine the relationship that socio-economic status and number of siblings might have on the outcome variables that differed significantly across groups in the ANOVA analyses above: parental stress and quality of life.

SES did not correlate significantly with parental stress ( $r = -.13$ ,  $p = .225$ ). SES was also found to have no significant correlation with the quality of life scores ( $r = -.17$ ,  $p = .165$ ). In this small sample, it thus seems that SES is not associated with these outcome variables.

The number of siblings was found to have no significant correlation with the parental stress scores across all three groups ( $r = .18, p = .133$ ). However, the number of siblings was found to have a significant inverse correlation with quality of life scores across all three parenting groups ( $r = -.42, p = .004$ ) – an increased number of children is thus associated with lower quality of life.



## Discussion

### Summary of Main Findings

The aim of this investigation was to determine whether parents of children with TSC would report higher levels of stress, depression and a lower quality of life than parents of children with ASD and parents of TD children. The study found that there was a significant difference with regards to parental stress scores, with parents of children with TSC reporting the highest parental stress scores. There was no significant difference found between the TSC, ASD and TD parent groups' depression scores. This study found a significant difference between the quality of life scores amongst the three parenting groups. TSC and ASD parenting groups differed significantly, with TSC parents reporting lower quality of life scores than parents of children with ASD. Moreover, SES was found to have no correlation to parental stress or quality of life scores. The number of siblings in a family was found to have no correlation with parental stress scores. However, there was a significant inverse correlation between number of siblings and quality of life scores, in that more siblings in a family was associated with reduced quality of life across all three groups.

### Parental Stress

In concurrence with the first hypothesis TSC parents reported the highest parental stress scores. Particularly, the posthoc tests found a significant difference between the TSC and ASD parent groups, with parents of children with TSC reporting higher parental stress scores. The disparities in parental stress could perhaps be attributed to the lack of support experienced by parents of children with TSC. In the Western Cape ASD is fairly well supported, for example, there are weekly Autism Western Cape sessions at Red Cross Children's Hospital. Moreover, there are many NGOs who provide additional support and information to ASD parents (e.g. Autism Western Cape, Association for Autism, Ernie Els Centre for Autism etc.). In addition to these support systems, parents of children with ASD have the option of various special education facilities which cater specifically for children with autism (Vera School for Learners with Autism, Alpha School) whereas no such option exists for parents of children with TSC. Another factor which may influence the parental stress scores of parents of children with TSC is the unpredictable nature of the disease. In addition to behavioural problems, TSC patients are also at risk for irregular hospital visits and trips to the doctor as TSC tubers can appear on any organ in the body, at any time. This unpredictability undoubtedly adds to the general stress that parents of children with TSC undergo. Moreover, there is currently no known cure for TSC, therefore, doctors are forced to

treat TSC symptoms as they appear (Curatolo & Maria, 2013). Red Cross Children's Hospital is currently the only TSC clinic in Sub-Saharan Africa, however, this clinic only occurs once every three months. Thus, TSC parents do not have a consistent support service when compared to the services run for ASD at Red Cross. Furthermore, parents of children with TSC who do not live in Cape Town would have to incur the additional costs of travel and accommodation.

### **Depression**

In contrast with the original hypothesis, this study found no significant difference in the depression scores across the parent groups. One reason for this may be that over time parents of children with TSC and ASD have developed certain levels of resilience. Studies on resilience of parents of children with special needs maintain that parents of children with special needs may initially react with negative emotions when first receiving the diagnosis, yet through support from family members, adequate literature and information on the child's disorder or disability these parents regain their belief in their child and in their child's future (Heiman, 2002). Another major factor which studies have found has a large impact on resilience is the ability to identify specific resilience processes. Examples of resilience processes include closer family ties, acquiring spiritual or religious strength and finding an optimistic and affirmative meaning with the child's disability (Bayat, 2007).

Another possible interpretation may be that all three parent groups are relatively equal on the depression scores (TSC:  $M = 15.36$ ;  $SD = 14.32$ ; ASD:  $M = 21.33$ ;  $SD = 11.51$ ; TD:  $M = 21.00$ ;  $SD = 18.29$ ), thus no significant difference would be found. Scores on the CES-D measure could range from 0 – 60, wherein a score of 16 or higher has generally been used as a cut-off point for elevated depressive indicators (Radloff, 1977). Using this cut-off score, all three groups are reporting some level of depression (TSC mean is just at cut off).

### **Quality of Life**

This investigation found a significant difference in the quality of life scores across the three parent groups. In line with the original hypothesis, the posthoc analysis found a significant difference in the ASD and TSC ( $M = 54.67$ ;  $SD = 9.62$ ) parenting groups, with parents of children with ASD ( $M = 500.80$ ;  $SD = 1669.15$ ) reporting the higher quality of life scores. Currently there is no literature that reflects the finding that TSC parents have a lower quality of life than parents of children with ASD, a PDD which is known to cause significant distress to parents.

In contrast to the original hypothesis, this study found that parents of children with ASD ( $M = 500.80$ ;  $SD = 1669.15$ ) reported a significantly higher quality of life than parents

of TD ( $M = 54.06$ ;  $SD = 9.50$ ) children. One could possibly argue that this finding is due to the fact that the TD parenting group in particular resides in an under resourced community. Although we found no correlation between SES and the significant outcomes (parental stress and quality of life), this may be due to the large variability around the mean SES for the ASD parent group as the mean SES in this parent cohort is particularly high. Whereas, parents of TD children do not have as much variability in SES as they are collectively in a lower SES bracket. Therefore, it could be argued that TD parents face substantial challenges which are related to their economic disadvantage and could negatively impact their quality of life.

### **Limitations and Future Directions**

Owing to the fact that this study reported a small sample size, the findings may not be completely reliable or applicable nationally. Although bootstrapping methods were employed in order to generate findings that were as reliable as possible it is hard to be confident that the estimated parameters accurately represent the population, even in the Western Cape. Therefore, in order to report truly meaningful and applicable data a larger sample size would be essential.

Moreover, due to the small sample size, it was particularly difficult to case-match participants on socio-demographic variables as we had initially hoped to do. The TD parent participants were mainly recruited from an underprivileged community. Unfortunately, due to difficulties such as time constraints and ethical approval only being granted in August this investigation was unable to recruit TD participants from varying socio-economic backgrounds. Better accuracy in case matching would lead to results that are more accurate and are able to be applied to a wider context - as this study could only endeavour to match participants within their income bands, SES may still be uneven across the groups. In addition, it's important to note that overall the TD parent scores did not fall into the expected rank on any of the measures used in this study. The hypotheses outlined in this investigation always placed this group in the least affected position yet they seem to score in close proximity to the TSC group in all the measures. Their negative parenting experiences are thus likely to be related to economic disadvantages and all the challenges that this brings. Likewise, because the TSC and TD groups had a similar SES range one could argue that some stress experienced by the TSC group may be related to economic disadvantage rather than just factors related to TSC. In future, studies should be sure to recruit adequate numbers of parents which are accurately stratified across the SES range.

Importantly, as this study made use of self-report measures participants were vulnerable to confounding factors such as social desirability bias and willingness to disclose.

In particular, I found one of the questions in the WHOQOL-BREF (“How satisfied are you with your sex life”) often made the participants uncomfortable as they may not have been expecting such a personal question and perhaps felt it was inappropriate. Future studies may consider using structured psychiatric interviews, focus groups or interviews in order to get detailed, comprehensive information from the participants.

### **Conclusion**

In conclusion, literature in the field of TSC is sparse, particularly in the South African context. This investigation found that parents of children with TSC are significantly more stressed than parents of children with ASD. Moreover, this study found that parents of children with TSC have a lower quality of life than parents of children with ASD. At present, there is no literature which reflects this finding in a South African context – this study thus makes an important preliminary contribution to this field of investigation.

As Pottie et al (2017) stated, long term care of an individual with a terminal illness or disorder has the possibility of becoming strenuous to the caregiver’s physical and mental health, which is evidenced by the conclusions drawn from this study. Psychosocial support structures are imperative in aiding parents of children with TSC manage their stress and aid in improving their quality of life.

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## Appendices

### Appendix A:

HREC REF 273/2016

### Consent Form: For parents of children with Tuberous Sclerosis Complex

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#### 1. Invitation and Purpose

I am a researcher from the Department of Psychology at the University of Cape Town. You are invited to take part in a research study as you have a child who has been diagnosed with the disorder Tuberous sclerosis complex (TSC). This study will look at the cognitive (i.e., thinking, learning, remembering and reasoning) and psychological (emotional thinking) profile of children with TSC as well as provide families with support to help them understand TSC and the impact it has on a family's daily life. This study aims to obtain a greater understanding of the cognitive and psychological profile of individuals with TSC in order to provide support for the children for these findings and for the families. Therefore, by understanding the difficulties families face on a daily basis, we aim to use this key information to develop appropriate

support to help families which may in turn aid in improving the quality of life of individuals with TSC and their families. Approximately 33 children with TSC and their parents will be invited to take part in this study.

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## 2. Procedures

If you decide to allow your child to take part in this study, we will ask them to complete a series of pencil-and-paper cognitive (learning) and psychological (emotional thinking) tests. The tests will assess your child's general intellectual functioning, their memory, attention, planning, problem solving, inhibition, processing speed, and language abilities, as well as if they are depressed and or anxious. There will be two sessions and each session will take about 60-90 minutes. You or another caregiver may be present at the testing sessions. Your child will be allowed to take breaks whenever requested during the sessions. They may also withdraw from the session at any time. These sessions can be arranged at a time that is convenient to you and your child.

If you decide to take part in this study, we will ask you to be involved in various sessions. The first session will be to complete a series of questionnaires relating to your child's emotional and behavioural profile. The second session will involve a detailed interview to further explore the behavioural difficulties noted on the questionnaires. In the third session you may be asked to partake in a behavioural intervention that will focus on modifying the behavioural difficulties reported in the interview and questionnaires. Each session will last between 60-90minutes. You will be allowed to take breaks whenever requested during the sessions. You may also withdraw from any session at any time. Additional support group sessions will be run throughout the year every 2-3 months. These sessions are voluntary and will last approximately 90 minutes.

## 3. Risks, Discomforts & Inconveniences

If you decide to take part in this study you will be exposed to minimal physical, social, and psychological risk. You will not be asked to perform any potentially harmful tasks. The only possible risk is that you or your child may become fatigued during the interviews or interventions. If you or your child does become fatigued, it will be possible to take a break. You will be able to withdraw from the study at any time, with no penalty and with no effect upon clinical or other services being delivered (or potentially being delivered).

#### 4. **Benefits**

This study provides families with a better understanding of both the cognitive and psychological profile of children with TSC from varying social demographic backgrounds.

HREC REF 273/2016 ; will be provided with essential information about TSC to help them better understand the condition and in turn be better equipped to deal with the difficulties their children face on a daily basis. Thirdly, families will be provided with support and advice in a group setting which provides a setting for the families to open up, share, and learn from other families that may be experiencing the same difficulties. And finally, this study also provides interventions or appropriate support through referrals to suitable doctors or therapists, which allows families to obtain the assistance they need which may in turn help them improve their quality of life.

#### 5. **Privacy and Confidentiality**

Information collected during each session will be stored in locked filing cabinets or in computers with security passwords. Only the researchers on the study have the right to review these research records.

#### 6. **Money Matters**

Participants will not incur any costs when participating in this study.

#### 7. **Questions**

If you have questions or concerns about the study please contact the principal investigator, Natalia M. Ing (cell) 071-689-5843 (email) [Natalia.Ing@uct.ac.za](mailto:Natalia.Ing@uct.ac.za), or Susan Malcolm-Smith (office) 021-650-4605 (email) [susanmalcolmsmith@gmail.com](mailto:susanmalcolmsmith@gmail.com), or [Jo Wilmshurst \(office\)](#) +27 21 658 5111 ex 5370, 5434 (email) [jo.wilmshurst@uct.ac.za](mailto:jo.wilmshurst@uct.ac.za).

## Consent Form

The study has been explained to me, and my questions have been answered.

I understand that participation in this study is voluntary, and that I may withdraw at any point.

I understand that I will not be identified except by an initial, and that this anonymity will be maintained throughout the study and when the research is published.

I consent to allow my child to participate in this study.

**Child's name** \_\_\_\_\_

**Signature of parent/guardian** \_\_\_\_\_

**Date** \_\_\_\_\_

I consent to participate in the questionnaire component of this study.

**Parent's name** \_\_\_\_\_

**Signature of parent** \_\_\_\_\_

**Date** \_\_\_\_\_

I consent to participate in the interview and intervention component of this study.

**Parent's name** \_\_\_\_\_

**Signature of parent** \_\_\_\_\_

**Date** \_\_\_\_\_

I have explained the study to the participant, and in my opinion s/he understands that participation is voluntary and is able to give informed consent.

**Researcher** \_\_\_\_\_

**Signature** \_\_\_\_\_

**Date** \_\_\_\_\_

## Use of Samples/Data for Future Research

With your permission, we would like to store the unused parts of your tests for use in future research. This test data will also form part of a large TSC research cohort database that can be accessed by the researchers on this project. This database is essential for the development of future research in South Africa. This is your choice entirely and you are free to say no and you will still be able to take part in the study. Please check the boxes that apply to your choice:

I do not want my information to be used for any future research. \_\_\_\_

You may use my information for any future research. \_\_\_\_

***Please indicate below if you would like to be notified of future research projects conducted by our research group:***

\_\_\_\_\_ (initial) Yes, I would like to be added to your research participation pool and be notified of research projects in which I might participate in the future.

**Method of contact:**

Phone number: \_\_\_\_\_

Cell phone number: \_\_\_\_\_

E-mail address: \_\_\_\_\_

Mailing address: \_\_\_\_\_

\_\_\_\_\_

\_\_\_\_\_

### Assent Form

#### Research on Tuberous Sclerosis Complex in South Africa

Hello! My name is Natalia. I'm here for a study on behalf of the University of Cape Town.

We're working with children with tuberous sclerosis complex within the Western Cape to gather information about how children with tuberous sclerosis think and feel. Therefore we are interested in how children remember things, how well they can concentrate, how well they can plan and solve problems, and also how they feel inside.

We would like to play some games with you and ask you some questions that will take place over two sessions with each session lasting about 90 minutes. You may take a break at any time during the games. I would like you to do them by yourself, but if you would like, you can ask for a parent, guardian or teacher to be with you at any time. I would like you to do your best in all of the games.

If you feel tired or uncomfortable during any of the games you may stop at any time. You do not have to continue playing if you don't want to.

If you decide to participate in this study, you will have the chance to help us better understand how children with tuberous sclerosis think and feel so that we can find ways to help with any problems they might have.

You do not have to take part in this study if you do not want to. If you do decide to take part in this study but change your mind later, you may stop participating and this will not change how you are looked after at the hospital.

If you agree to take part in this study, everything will be kept confidential. That means that all your information will be kept private between you and me and only people involved in this study will see this information.

Do you have any questions about what was just mentioned? If you think of any questions in the future, you can reach me at 071 689 5843.

Would you like to participate in the research?

**Date:** \_\_\_\_\_

**Child's Name/Agreement:** \_\_\_\_\_

**Parent's/Guardian's Agreement:** \_\_\_\_\_

**Researcher's Signature:** \_\_\_\_\_

## Appendix B:

### Consent Form: ASD Parents

#### Principal Researchers:

Dr. Kevin G. F Thomas  
Associate Professor  
Department of Psychology  
University of Cape Town  
021-650-4608

Natalia Ing  
Doctoral candidate  
Department of Psychology  
University of Cape Town  
071 689 5843

Dr Susan Malcolm-Smith  
Senior Lecturer  
Department of Psychology  
University of Cape Town  
021-650-4605

#### 7. Invitation and Purpose

You are invited to take part in a research study about the services provided to individuals on the autism spectrum and their families as well as the impact service delivery may have on the quality of life of the parents of autism spectrum disorder (ASD) children. I am a researcher from the Department of Psychology at the University of Cape Town. This study aims to obtain a greater understanding of the services provided (and those not provided) to individuals with autism and their families, as well as the impact aspects of service delivery have on the quality of life of individuals with autism and their families. This study will help provide key information regarding the shortfalls and limitations of the services provided and will help improve future services for individuals with autism which may in turn aid in improving the quality of life of individuals with autism and their families. Approximately 150 parents of children with autism will participate in this study.

#### 8. Procedures

If you decide to take part in this study, we will ask you complete a series of questionnaires and surveys relating to service delivery and quality of life. This will last between 60-90minutes. You may withdraw from the session at any time if you feel uncomfortable. You can come in to complete the survey or you could complete an online survey that would be sent to you via email. You have the option of participating in the second session involving focus groups with 4 other parents to discuss various topics concerning service delivery and quality of life in parents of children with autism. You do not have to participate in the second session, this participation is optional even if you are participating in the first session. If you are asked to partake in the focus group session, this session will last 90 minutes. You will be allowed to take breaks whenever requested during the sessions.

#### 9. Risks, Discomforts & Inconveniences

If you decide to take part in this study you will be exposed to no physical, social, or psychological risk. You will not be asked to perform any potentially harmful tasks. You will be able to withdraw from the study at any time, with no penalty and with no effect upon clinical or other services being delivered (or potentially being delivered).

#### 10. **Benefits**

Although there are no direct benefits by participating in this study, the information from this study may help improve our understanding of what services are (and are not) provided to individuals with autism and their families. This information may also provide us with a better understanding of the quality of life of parents with children with autism as well as the possible impact service delivery may have on the quality of life of parents of children with autism within South Africa. This study also aims to provide information to help lobby for better service provision for the autism community.

#### 11. **Privacy and Confidentiality**

Information collected during each session will be stored in locked filing cabinets or in computers with security passwords. Only certain people have the right to review these research records. These people include the researchers for this study and certain University of Cape Town officials.

#### 12. **Money Matters**

Participants will not incur any costs when participating in this study. A small compensation of R100 will be provided to all participants that come in to complete the survey.

#### 7. **Questions**

If you have questions or concerns about the study please contact the principal investigator Susan Malcolm-Smith, PhD: (office) 021-650-4605 (email) [susanmalcolmsmith@gmail.com](mailto:susanmalcolmsmith@gmail.com) , Kevin G. F. Thomas, PhD: (office) 021-650-4608 (email) [Kevin.Thomas@uct.ac.za](mailto:Kevin.Thomas@uct.ac.za), or Natalia M. Ing (cell) 071-689-5843 (email) [Natalia.Ing@uct.ac.za](mailto:Natalia.Ing@uct.ac.za).



## Consent Form

The study has been explained to me, and my questions have been answered.

I understand that participation in this study is voluntary, and that I may withdraw at any point.

I understand that I will not be identified except by an initial, and that this anonymity will be maintained throughout the study and when the research is published.

I consent to participate in the survey component of this study.

**Parent's name** \_\_\_\_\_

**Signature of parent** \_\_\_\_\_

**Date** \_\_\_\_\_

Please select one of the options below:

\_\_\_\_ I would like to come in and complete the survey in person with the researcher

\_\_\_\_ I would like to be sent a link via email to complete the survey online

### **Use of Samples/Data for Future Research**

With your permission, we would like to store the unused parts of your tests for use in future research. This is your choice entirely and you are free to say no and you will still be able to take part in the study. Please check the boxes that apply to your choice:

I do not want my samples to be used for any future research. \_\_\_\_

You may use my samples for any future research. \_\_\_\_

*Please indicate below if you would like to be notified of future research projects conducted by our research group:*

\_\_\_\_\_ (initial) Yes, I would like to be added to your research participation pool and be notified of research projects in which I might participate in the future.

Would you like to partake in a focus group session with 4 other parents?

YES

NO

I consent to participate in the focus group component of this study.

**Parent's name** \_\_\_\_\_

**Signature of parent** \_\_\_\_\_

**Date** \_\_\_\_\_

Method of contact:

Phone number: \_\_\_\_\_

Cell phone number: \_\_\_\_\_

E-mail address: \_\_\_\_\_

Mailing address: \_\_\_\_\_

\_\_\_\_\_  
\_\_\_\_\_

**Researcher section:**

I have explained the study to the participant, and in my opinion s/he understands that participation is voluntary and is able to give informed consent.

**Researcher** \_\_\_\_\_

**Signature** \_\_\_\_\_

**Date** \_\_\_\_\_

**Appendix C:****Consent Form: For Parents of Typically Developing Children****Principal Researchers:**

Prof Jo Wilmshurst

Professor

Department of Paediatrics and Child Health

Red Cross Children's War Memorial Hospital

+27 21 658 5111 ex 5370, 5434

Dr Susan Malcolm-Smith

Senior Lecturer

Department of Psychology

University of Cape Town

021 650 4605

Natalia Berghoff/Ing

Doctoral candidate

Department of Psychology

University of Cape Town

071 689 5843

**1. Invitation and Purpose**

I am a researcher from the Department of Psychology at the University of Cape Town. You are invited to take part in this study which will look at the cognitive (i.e., thinking, learning, remembering and reasoning) and psychological (emotional thinking) profile of children with Tuberous Sclerosis Complex (TSC) compared to typically developing children. This study aims to obtain a greater understanding of the cognitive and psychological profile of individuals with TSC in order to provide support for the children and their families. By understanding the difficulties families face on a daily basis, we aim to use this key information to develop appropriate support to help families which may in turn aid in improving the quality of life of

individuals with TSC. Approximately 33 children with TSC and 33 children without TSC and their parents will be invited to take part in this study.

## **2. Procedures**

If you decide to allow your child to take part in this study, we will ask them to complete a series of pencil-and-paper cognitive (learning) and psychological (emotional thinking) tests. The tests will assess your child's general intellectual functioning, their memory, attention, planning, problem solving, inhibition, processing speed, and language abilities, as well as if they are depressed and or anxious. There will be two to three sessions and each session will take about 60-90 minutes. You or another caregiver may be present at the testing sessions. Your child will be allowed to take breaks whenever requested during the sessions. They may also withdraw from the session at any time. These sessions can be arranged either at the University of Cape Town, Red Cross War Memorial Children's Hospital or at your child's school at a time that is convenient to you and your child.

If you decide to take part in this study, we will ask you to complete a series of questionnaires relating to your child's emotional and behavioural profile. This session will last between 60-90minutes. You will be allowed to take breaks whenever requested during the session. You may also withdraw from the session at any time.

## **3. Risks, Discomforts & Inconveniences**

If you decide to take part in this study you and your child will be exposed to minimal physical, social, and psychological risk. You will not be asked to perform any potentially harmful tasks. The only possible risk is that you or your child may become tired during the assessments. If you or your child does become tired, it will be possible to take a break. You will be able to withdraw from the study at any time, with no penalty and with no effect upon clinical or other services being delivered (or potentially being delivered). If you or your child feel uncomfortable or encounter psychological distress during interviews, appropriate support will be provided.

#### **4. Benefits**

Following the completion of the assessments, you will be provided with feedback regarding your child's performance on the cognitive and psychological tasks. Furthermore, your willingness to assist us will help us in providing families from varying sociodemographic backgrounds with essential information about TSC to help them better understand the condition and in turn be better equipped to deal with the difficulties their children face on a daily basis. Thus, through your participation you can take pride in knowing that you assisted in furthering the scientific knowledge of TSC.

#### **5. Privacy and Confidentiality**

Information collected during each session will be stored in locked filing cabinets or in computers with security passwords. Only the researchers on the study have the right to review these research records.

#### **6. Questions**

If you have questions or concerns about the study please contact the principal investigator, Natalia M. Berghoff/ Ing (cell) 071-689-5843 (email) [Natalia.Ing@uct.ac.za](mailto:Natalia.Ing@uct.ac.za), or Susan Malcolm-Smith (office) 021-650-4605 (email) [susanmalcolmsmith@gmail.com](mailto:susanmalcolmsmith@gmail.com), or Jo Wilmshurst (office) +27 21 658 5111 ex 5370, 5434 (email) [jo.wilmshurst@uct.ac.za](mailto:jo.wilmshurst@uct.ac.za) .

## Consent Form

The study has been explained to me, and my questions have been answered.  
I understand that participation in this study is voluntary, and that I may withdraw at any point without repercussions.  
I understand that I will not be identified except by an initial, and that this anonymity will be maintained throughout the study and when the research is published.  
I consent to allow my child to participate in this study.

**Child's name** \_\_\_\_\_

**Signature of parent/guardian** \_\_\_\_\_

**Date** \_\_\_\_\_

I consent to participate in the questionnaire component of this study.

**Parent's name** \_\_\_\_\_

**Signature of parent** \_\_\_\_\_

**Date** \_\_\_\_\_

I consent to allow researchers to contact me following the commencement of this study, inviting me to participate in the larger investigation which aims to develop interventions

**Parent's name** \_\_\_\_\_

**Signature of parent** \_\_\_\_\_

**Date** \_\_\_\_\_

I have explained the study to the participant, and in my opinion s/he understands that participation is voluntary and is able to give informed consent.

**Researcher** \_\_\_\_\_

**Signature** \_\_\_\_\_

**Date** \_\_\_\_\_

## Use of Samples/Data for Future Research

With your permission, we would like to store the unused parts of your tests for use in future research. This test data will also form part of a large research cohort database that can be accessed by the researchers on this project. This database is essential for the development of future research in South Africa. This is your choice entirely and you are free to say no and you will still be able to take part in the study. Please check the boxes that apply to your choice:

I do not want my information to be used for any future research. \_\_\_\_

You may use my information for any future research. \_\_\_\_

***Please indicate below if you would like to be notified of future research projects conducted by our research group:***

\_\_\_\_\_ (initial) Yes, I would like to be added to your research participation pool and be notified of research projects in which I might participate in the future.

**Method of contact:**

Phone number: \_\_\_\_\_

Cell phone number: \_\_\_\_\_

E-mail address: \_\_\_\_\_

Mailing address: \_\_\_\_\_  
 \_\_\_\_\_  
 \_\_\_\_\_

**Assent Form****Research on Tuberous Sclerosis Complex in South Africa**

Hello! My name is \_\_\_\_\_ (researcher's name). I'm here for a study on behalf of the University of Cape Town.

We're working with children with tuberous sclerosis complex (TSC) within the Western Cape to gather information about how children with TSC think and feel. Therefore we are interested in how children remember things, how well they can concentrate, how well they can plan and solve problems, and also how they feel inside.

We would like to play some games with you and ask you some questions that will take place over two or three sessions with each session lasting about 90 minutes. You may take a break at any time during the games. I would like you to do them by yourself, but if you would like, you can ask for a parent, guardian or teacher to be with you at any time. I would like you to do your best in all of the games.

If you feel tired or uncomfortable during any of the games you may stop at any time. You do not have to continue playing if you don't want to.

If you decide to participate in this study, you will have the chance to help us better understand how children with TSC think and feel so that we can find ways to help with any problems they might have.

You do not have to take part in this study if you do not want to. If you do decide to take part in this study but change your mind later, you may stop participating without repercussions.

If you agree to take part in this study, everything will be kept confidential. That means that all your information will be kept private between you and me and only people involved in this study will see this information.

Do you have any questions about what was just mentioned? If you think of any questions in the future, you can reach me at \_\_\_\_\_ (researcher's contact number).

Would you like to participate in the research?

**Date:** \_\_\_\_\_

**Child's Name/Agreement:** \_\_\_\_\_

**Parent's/Guardian's Agreement:** \_\_\_\_\_

**Researcher's Signature:** \_\_\_\_\_





12. Has your child ever experienced learning difficulties such as dyslexia or attention-deficit / hyperactivity disorder (ADD/ ADHD)?

YES NO

If yes, please specify:

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---

13. Has your child ever experienced a head injury? (e.g., being hit on the head with an object and losing consciousness as a result)

YES NO

If yes, please give details:

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---



---

14. Has your child ever experienced any of the following medical conditions:

- a. Neurological problems (e.g., epilepsy, meningitis, cerebral palsy, encephalitis, Tourette's syndrome, brain tumour, other)

YES NO

If yes, please specify:

---



---



---

- b. Depression

YES NO

If yes, please specify:

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---



---

- c. Memory problems

YES NO

If yes, please specify:

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- d. Problems with their vision:

YES NO

If yes, please specify:

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---



---

- e. Problems with their hearing (e.g. difficulty hearing, needing hearing aids, needing grommets):

YES NO

If yes, please specify (please include details on how this affected their language development):

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f. Is he/she currently taking any prescription medication? YES NO

If yes, what medication(s)?

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---



---

**B. Parent Information:**

Please note that information on the primary caregiver is required. If the child is adopted, please indicate this. If the primary caregiver is not the biological or adoptive mother or father, please place their information under "Guardian".

Name: \_\_\_\_\_ Date of birth (dd/mm/jj): \_\_\_\_\_  
 Age: \_\_\_\_\_ Sex (circle one): Male Female

1. Ethnicity: White Black Indian Coloured Asian Other

If other please specify: \_\_\_\_\_

2. Home Language: \_\_\_\_\_

3. Relationship to the child (circle one):

Biological Parent Caregiver Adoptive parent Step parent Gaurdian  
 Foster Parent

**1. What is the total monthly income of the household in which you live? (Tick the appropriate block):**

[NOTE: This should be total household income, not personal income.]

0 – R2999	R3000 – R6299	R6300 – R 10 499	R10 500 – R 14599
R14 600 – R18 799	R18 800 – R22 999	R23 000 – R26 999	R27 000 – R31 299
R31 300 – R35 499	R35 500 - R39 499	R39 500 – R43 750	more than R43 750:

**What is the estimated value of your total monthly household income: R \_\_\_\_\_**

**2. Highest level of education completed for mother, father, and/or guardian (please circle appropriate number).**

	Mother	Father	Guardian
0) 0 years (Never went to school)	0	0	0
1) Grade 1	1	1	1
2) Grade 2	2	2	2
3) Grade 3 / Standard 1	3	3	3
4) Grade 4 / Standard 2	4	4	4
5) Grade 5 / Standard 3	5	5	5
6) Grade 6 / Standard 4	6	6	6
7) Grade 7 / Standard 5 [Completed primary school]	7	7	7
8) Grade 8 / Standard 6	8	8	8
9) Grade 9 / Standard 7	9	9	9
10) Grade 10 / Standard 8	10	10	10
11) Grade 11 / Standard 9	11	11	11
12) Grade 12 / Standard 10 [Matric; Completed high school]	12	12	12

13) Tertiary education: Higher education certificate	13	13	13
14) Tertiary education: Diploma received	14	14	14
15) Tertiary education: Bachelor's degree received	15	15	15
16) Tertiary education: Post graduate degree received	16	16	16
17) Don't know	17	17	17

### 3. Parental employment: (Please circle appropriate number)

	Biological mother	Biological father	Guardian
1. Higher executives, owners of large businesses, major professionals (e.g. doctors, lawyers)	1	1	1
2. Business managers of medium sized businesses, professions (e.g. nurses, opticians, pharmacists, social workers, teachers, accountants)	2	2	2
3. Administrative personnel, managers, owners / proprietors of small businesses (decorator, actor, reporter, travel agent)	3	3	3
4. Clerical and sales, technicians, small businesses (e.g. bank teller, bookkeeper, clerk, draftsman, timekeeper, secretary)	4	4	4
5. Skilled manual – usually having had training (e.g. baker, barber, chef, electrician, fireman, machinist, mechanic, painter, welder, police, plumber, electrician)	5	5	5
6. Semi-skilled (e.g. hospital aide, painter, bartender, bus driver, cook, garage guard, checker, waiter, machine operator)	6	6	6
7. Unskilled (e.g. attendant, janitor, construction helper, unspecified labour, porter)	7	7	7
8. Homemaker	8	8	8
9. Student, disabled, no occupation	9	9	9

### 4. Material and financial resources (please answer for each item).

Which of the following items, in working order, does your household have?

Items	Yes	No
1. A refrigerator or freezer	Yes	No
2. A vacuum cleaner or polisher	Yes	No
3. A television	Yes	No
4. A hi-fi or music center (radio excluded)	Yes	No
5. A microwave oven	Yes	No
6. A washing machine	Yes	No
7. A video cassette recorder or dvd player	Yes	No

Which of the following do you have in your home?

Items	Yes	No
1. Running water	Yes	No
2. A domestic servant	Yes	No
3. At least one car	Yes	No
4. A flush toilet	Yes	No

5. A built-in kitchen sink	Yes	No
6. An electric stove or hotplate	Yes	No
7. A working telephone	Yes	No

Do you personally do any of the following?

Items	Yes	No
1. Shop at supermarkets	Yes	No
2. Use any financial services such as a bank account, ATM card or credit card	Yes	No
3. Have an account or credit card at a retail store	Yes	No

## Appendix E



**UNIVERSITY OF CAPE TOWN**  
**Faculty of Health Sciences**  
**Human Research Ethics Committee**



Room E53-46 Old Main Building  
 Groote Schuur Hospital  
 Observatory 7925  
 Telephone [021] 406 6492  
 Email: [sumayah.arietdien@uct.ac.za](mailto:sumayah.arietdien@uct.ac.za)  
 Website: [www.health.uct.ac.za/fhs/research/humanethics/forms](http://www.health.uct.ac.za/fhs/research/humanethics/forms)

27 July 2016

**HREC REF: 273/2016**

**Prof J Wilmshurst**  
 Division of Neurology  
 Department of Paediatrics  
 5<sup>th</sup> Floor, ICH Building  
 Red Cross War Memorial Childrne's Hospital  
 Rondebosch

Dear Prof Wilmshurst

**PROJECT TITLE: EXAMINING THE NEUROCOGNITIVE AND BEHAVIOURAL PROFILE OF WESTERN CAPE CHILDREN WITH TUBEROUS SCLEROSIS COMPLEX TO DEVELOP APPROPRIATE INDIVIDUALIZED INTERVENTIONS TO ADDRESS BEHAVIOURAL DIFFICULTIES**

Thank you for your response letter, addressing the issues raised by the Human Research Ethics Committee (HREC).

It is a pleasure to inform you that the HREC has **formally approved** the above-mentioned study.

**Approval is granted for one year until the 30 July 2017.**

Please add the UCT FHS HREC contact details to the informed consent document.  
 Please submit study 2 for review.

Please submit a progress form, using the standardised Annual Report Form if the study continues beyond the approval period. Please submit a Standard Closure form if the study is completed within the approval period.  
 (Forms can be found on our website: [www.health.uct.ac.za/fhs/research/humanethics/forms](http://www.health.uct.ac.za/fhs/research/humanethics/forms))

**Please quote the HREC REF in all your correspondence.**

Please note that the ongoing ethical conduct of the study remains the responsibility of the principal investigator.

Please note that for all studies approved by the HREC, the principal investigator **must** obtain appropriate institutional approval before the research may occur.

HREC 273/2016

Yours sincerely

pp T. Burgess  
**PROFESSOR M. BLOCKMAN**  
**CHAIRPERSON, FHS HUMAN RESEARCH ETHICS COMMITTEE**

Federal Wide Assurance Number: FWA00001637.

Institutional Review Board (IRB) number: IRB00001938

This serves to confirm that the University of Cape Town Human Research Ethics Committee complies to the Ethics Standards for Clinical Research with a new drug in patients, based on the Medical Research Council (MRC-SA), Food and Drug Administration (FDA-USA), International Convention on Harmonisation Good Clinical Practice (ICH GCP), South African Good Clinical Practice Guidelines (DoH 2006), based on the Association of the British Pharmaceutical Industry Guidelines (ABPI), and Declaration of Helsinki (2013) guidelines.

The Human Research Ethics Committee granting this approval is in compliance with the ICH Harmonised Tripartite Guidelines E6: Note for Guidance on Good Clinical Practice (CPMP/ICH/135/95) and FDA Code Federal Regulation Part 50, 56 and 312.

## Appendix F:



FACULTY OF HEALTH SCIENCES  
Human Research Ethics Committee



## 8. Declarations and Signatures

This application will not be processed unless all the required declarations and signatures are completed according to the Committee's Standard Operating Procedures (SOP)

## 8.1 Head of Department or Division

My signature confirms that

- i. The researcher(s)/student(s)/supervisor(s) have the skills, training, experience and time to undertake this research
- ii. There are adequate resources (e.g. equipment, space, support services) to perform this research

Signature of Head		Date	18/4/16
Print name			

Note: Where the PI is also Head of Department, confirmation must be obtained from an authorised designee. PIs may not approve their own research.

## 8.2 Chairperson of the Departmental Research Committee (DRC)

My signature confirms that:

- i. This research has undergone peer review by a person(s) experienced in the field of study
- ii. This research is well-designed and scientifically sound
- iii. Where relevant, all methodological issues have been resolved to the satisfaction of the peer reviewer(s)
- iv. If conducted according to the protocol, this research is expected to yield valid and useful findings.

Signature of Chairperson		Date	22/03/2016
Print name	Brenda Morrow	Department of Paediatrics and Child Health Research Committee	

Note: Where the PI is also the Chairperson of the DRC, confirmation must be obtained from an authorised designee. PIs may not approve their own research.

## 8.3 Principal Investigator

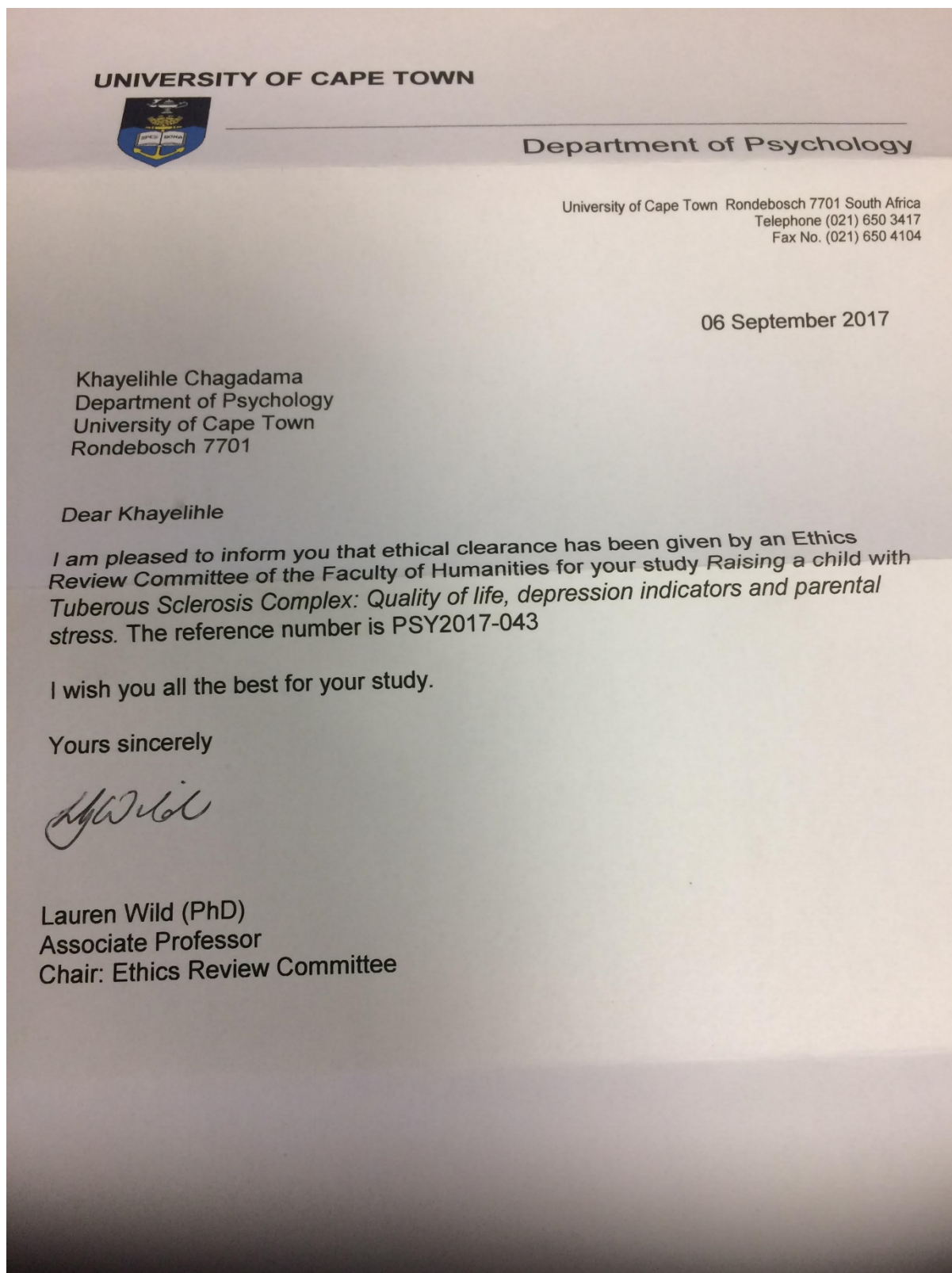
My signature confirms that:

- i. Information in this application is true and accurate
- ii. I will begin the research only after HREC approval is obtained
- iii. I accept full responsibility for the conduct of this research and the protection of participants' rights and welfare
- iv. I will conduct the research according to all ethical, regulatory and legal requirements stipulated in the HREC's Standard Operating Procedures.
- v. I will provide progress reports to the HREC as requested, including a final closing report at the end of the research
- vi. I will notify the HREC in writing if any change to the research is proposed and await approval before proceeding with the proposed change except when urgently necessary to protect participants' safety
- vii. I will notify the HREC in writing immediately if any adverse event or unanticipated problem occurs during the research
- viii. I will allow an audit of my research if requested by the HREC
- ix. I have the time, training, experience and resources to oversee this research

Signature of Principal Investigator		Date	30/01/2016
-------------------------------------	--	------	------------



## Appendix G:



## Appendix H:



## UNIVERSITY OF CAPE TOWN

DC: HUM /

## FACULTY OF HUMANITIES

## PROPOSAL APPROVAL FORM

<b>DOCTORATE</b> (A research proposal must accompany this form)	<b>RESEARCH MASTERS</b> (A research proposal must accompany this form)	<b>C/W MASTERS</b>
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**SECTION A: (To be completed by candidate)**

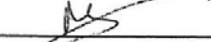
Please complete this form and return it to the Faculty Office once you have obtained the signatures of the supervisor(s) and Head of Department.

Surname	INGE				First Name(s)	NATALIA
Title	Mr.	Ms.	Mrs.	Miss	Student No	INGNAT001
Address	25 Mitchells Way Edgemead					
Telephone(Home)					Work/Cell	071 689 5843


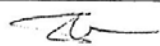
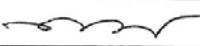

Note: Your UCT Email address is the default email address for all official communication – make sure that you access it regularly.

Department	Psychology
Title of Dissertation:	Service Delivery and its Implications for Quality of Life in ASD

Qualifications held			
Degree/Diploma	Major(s) & Subjects	Month/Year awarded	University
Masters in Psychological Research	Neuropsychology	2011	UCT
Honors in Psych		2008	UCT
BA in humanities	Psych + Sociology	2007	Stellenbosch

Signature of candidate:  Date: 05/08/13

**SECTION B:**

	Name	Signature	Date
Supervisor	Ivan Thomas		8/8/13
Co-supervisor (if applicable)	Susan Marinda Smit		8/8/13
HOD	M. Solms		12/8/13
Deputy-Dean: Research			
Ethics approval obtained where applicable	on behalf of Departmental Ethics Committee		8/8/13

August 8, 2013

**DEPARTMENT OF PSYCHOLOGY**  
**REPORT OF THESIS COMMITTEE**

**Student Name:** NATALIA ING

**Student #:** ING NAT001

**Degree:** PHD

**Title (as proposed)** SERVICE DELIVERY AND ITS IMPLICATION  
FOR QUALITY OF LIFE IN ASD

**Supervisor:** KEVIN THOMAS

**Co-supervisor:** SHARON SMITH

**Committee members:** CATHY WARD  
PROGRESS NIMBORO  
NORLITULA STABALA

WE:

1. Approve the proposal, and recommend that the student continue with the research.
2. Approve the proposal, and recommend that the student may continue with the research. However, we recommend that change(s), as noted below, be incorporated in the research, to the satisfaction of the supervisor.
3. Approve the proposal in terms of its ethical implications. If necessary, explanatory notes appear below.
4. Find the proposal unsatisfactory, for the reason(s) listed below. The student is hereby requested to re-present the proposal to a departmental thesis committee by \_\_\_\_\_.

**NOTES:** (1) Also guidelines to emerge in focus groups.

- ① Screen adults for comprehension - study needs to be replicable.
- ② Consider a deeper developmental theoretical approach instead of the breadth.
- ③ Avoid duplication of questions across instruments; post-work employment.
- ④ Consent forms: add recording and signature line for focus group; simplify consent for adult ASD participants and have a consent form for their guardians.
- ⑤ Include discussion of disabled status.

DEPARTMENT OF PSYCHOLOGY  
TERMS OF REFERENCE OF DEPARTMENTAL  
THESIS COMMITTEES

Students doing the research degrees in psychology (M.A. and M. Soc. Sc.) are required to present a research proposal to a departmental thesis committee before the end of May of the first year of registration for the degree.

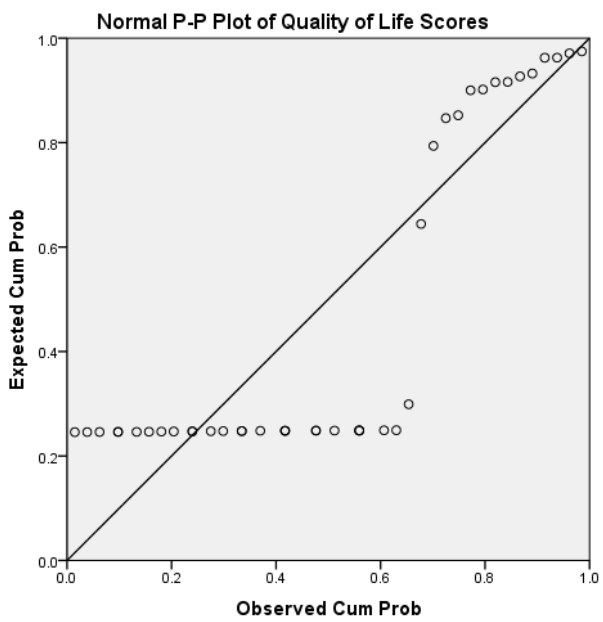
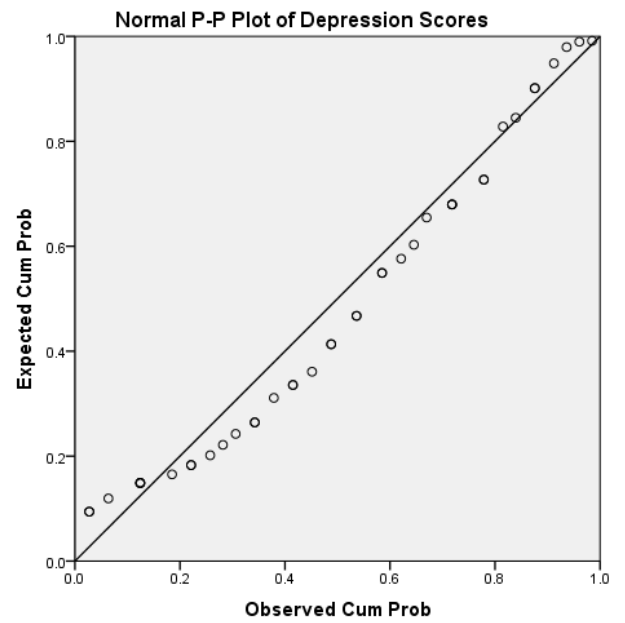
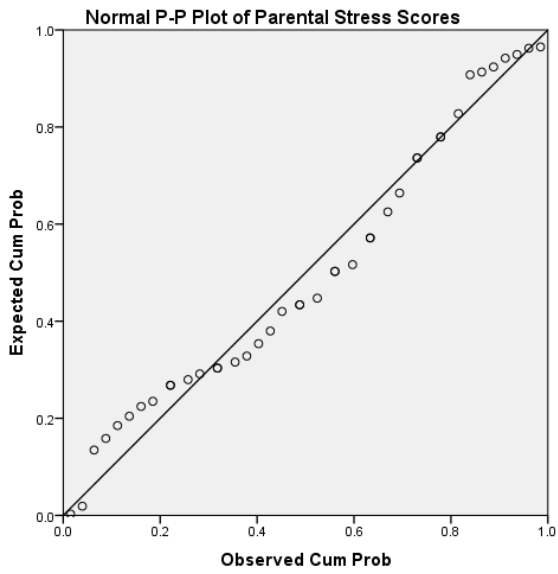
The committee will be constituted by the supervisor in consultation with the course convenor, and will consist of at least the Head of Department (ex officio), supervisor and three Department of Psychology staff members.

Students submit their proposals in written format to individual members at least one week before the scheduled presentation. The supervisor will act as chairperson at the presentation of the research proposal.

The committee is not an examination body, but it acts to approve a student's research proposal, and hence continued registration as a Master's student. It may make recommendations at the following levels:

- Approve the proposal, and that the student may continue with the research. The committee also acts in an advisory capacity, and may recommend changes to the research. Under normal circumstances it will be left to the discretion of the supervisor and student whether to incorporate such changes or not, unless the committee specifies that a particular recommendation has to be incorporated in the research. In such cases the committee is requested to submit a brief statement, containing the relevant details, to the course convenor. (The committee may elect a reporting member to do this).
- Refer the proposal back to the student, and request re-presentation within a specified period. Again the course convenor has to be informed of this in writing. If, at the second presentation, the committee still regards the proposal as unsatisfactory, it may recommend that the student de-registers due to lack of progress. Should this happen, the committee's decision will be conveyed via the course convenor to the Head of the Department, to whom both the student and the supervisor have the right to appeal. Students have a further right of appeal to the Dean.

The committee also considers the ethical implications of the study, and act as the departmental body that approves it in this regard.

**Appendix I:**

## Appendix J:

### Bootstrap for Multiple Comparisons

Dependent Variable: Socio-Economic Status

Games-Howell

(I) Label	(J) Label	Mean Difference (I- J)	Bias	Bootstrap <sup>a</sup>		
				Std. Error	95% Confidence Interval Lower	Upper
TSC	ASD	-96097.08333	-377.87657	50100.10643	-199426.394	-4441.99393
	Control	-6370.97222	668.56188	19576.45875	-40486.14967	36933.56536
ASD	TSC	96097.08333	377.87657	50100.10643	4441.99393	199426.3940
	Control	89726.11111	1046.43845	49368.78000	3305.04839	191963.0182
Control	TSC	6370.97222	-668.56188	19576.45875	-36933.56536	40486.14967
	ASD	-89726.11111	-1046.43845	49368.78000	-191963.018	-3305.04839

a. Unless otherwise noted, bootstrap results are based on 1000 bootstrap samples

*Note:* Significant mean differences are highlighted with an asterix.