

https://dspace.org/

Assessment of Maternal-Infant-Bonding between mothers and their babies with a cleft lip

Nienaber, Elzette

2017

https://hdl.handle.net/10539/25607

Downloaded from a DSpace repository. https://dspace.org/

ASSESSMENT OF MATERNAL-INFANT BONDING BETWEEN MOTHERS AND THEIR BABIES WITH A CLEFT LIP

Elzette Nienaber

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

Johannesburg, 2017

Declaration

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



(signature of candidate)

16th day of November in 2017 in Johannesburg

Contribution of the candidate to the paper

Declaration: Student's contribution to article(s) and agreement of co-author(s)

I, Elzette Nienaber, student number 762941, declare that this Research Report is my own work and that I contributed significantly towards research findings presented in the paper intended for publication below.

Signature of Student 

Date: 16 November 2017

Name of Primary Supervisor: Merlyn Glass

Signature of Primary Supervisor 

Date: 16 November 2017

Agreement by co-authors: By signing this declaration, the co-authors listed below agree to the use of the article(s) by the student as part of her Research Report. In cases where the student is not the 1st author of a published article, the primary supervisor must explain (under comments) why the student is entitled to use the paper for his/her degree purposes.

Article Title: Assessment of Maternal-Infant Bonding between Mothers and their Babies with a Cleft Lip

Authors	Name	Signature	Date
1st author	Elzette Nienaber		16 November 2017
2nd author	Lindsay Lambie		16 November 2017
3rd author	Merlyn Glass		16 November 2017

Presentations arising from this research project

1. SASHG Biennial Congress 2017, 13th -16th August, Durban, KwaZulu-Natal (poster presentation)
2. University of the Witwatersrand Cross Faculty Symposium, 25th of October (poster presentation)
3. Division of Human Genetics Departmental Seminar, 8th November, National Health Laboratory Service (oral presentation)

Abstract

Currently there is no literature exploring how having a child with a cleft lip with or without a cleft palate affects the maternal-infant bonding in a South African context. This study aimed to describe bonding between mothers and their affected infants and to determine whether demographic factors are associated with bonding impairment. Thirty two mothers with affected children were recruited from two clinics at Johannesburg State Hospitals. The Postpartum Bonding Questionnaire was used to establish bonding impairment in this group. The results indicated that 4 (12.5%) of the mothers had some impaired bonding while the majority of mothers 28 (87.5%) had normal bonding. Some mothers displayed a degree of anxiety concerning planned reconstructive surgery. In conclusion, we found that having a child with a cleft lip does not significantly influence maternal-infant bonding, however investigations with larger cohort is needed for further clarification.

Acknowledgements

I would like to thank my supervisors, Ms Merlyn Glass and Dr Lindsay Lambie for their guidance and the support they have offered during this year. It has been a privilege working with you and learning from your experiences.

Thank you to every mother who agreed to participate in this study. The road has not always been an easy one but you choose to walk with boldness and courage each day. Your bravery is admirable.

I also wish to acknowledge the Health Science Research Office Biostatistics Department of The University of the Witwatersrand for assisting me with the statistical analysis of this project.

Ms Shelley Macaulay for her assistance with statistical analysis and reviewing my work.

To my parents, Thinus and Monica Nienaber, thank you for the continuous love and support through these last two years.

Bianca and Melissa my dearest friends, thank you for making this journey worth every step. Without your laughter, encouragement and friendship this would not have been possible. I have learned so much from each of you.

To my Heavenly Father for strengthening me, for giving me wisdom and clarity and for always being my true North.

Therefore, my beloved, be steadfast, immovable, always excelling in the work of the Lord, because you know that in the Lord your labour is not in vain.

1 Corinthians 15:58

Table of Contents

Declaration i

Contribution of the candidate to the paper ii

Presentations arising from this research project iii

Abstract iv

Acknowledgements v

Research report in the format of a "submissible" paper

Title page

Abstract 1

Keywords 1

Introduction 2

Materials and Methods 3

Results 5

Discussion 9

Conclusion 11

References 13

Appendices

Approved research protocol

Ethics clearance certificate

Turn-it-in report

Archives of Women's Mental Health Author Guidelines

List of Tables

Table 1: Association between demographic factors and bonding in mothers with normal bonding and mothers with impaired bonding. 6

Table 2: Types of clefts observed in study population. 7

Table 3: Mean PBQ scores for mothers with and without impaired bonding. 8

Assessment of Maternal-Infant Bonding between Mothers and their Babies with a Cleft Lip

Elzette Nienaber[1], Lindsay Lambie[1,2], Merlyn Glass[1,2]

[1]Division of Human Genetics, School of Pathology, Faculty of Health Sciences, The University of the Witwatersrand, Johannesburg, South Africa; [2]Division of Human Genetics, National Health Laboratory Service, Johannesburg, South Africa

elzettenienaber@gmail.com

Tel: +27 83 604 4036

Abstract

Purpose: A mother's attitude and emotions towards her infant is of crucial importance for the infant's development and well-being. It has been suggested that facial deformities, such as a cleft lip with or without a cleft palate, can elicit feelings of shock and desperation, which may have implications for the bonding process between mother and infant. Currently there is no literature exploring how having a child with a cleft lip with or without a cleft palate affects the maternal-infant bonding in a South African context. The aim of this study was to describe bonding between mothers and their infants with a cleft lip with or without a cleft palate and to determine whether any demographic factors were associated with bonding impairment.

Methods: This was a descriptive study. Thirty two mothers with a child affected by a cleft lip with or without a cleft palate were recruited from specialised clinics at two Johannesburg State Hospitals. The Postpartum Bonding Questionnaire was administered in an interview format to establish if there was any bonding impairment in the study group.

Results: A total of 32 mothers were included in the study. The results indicated that four (12.5%) of the mothers had a measure of impaired bonding while the majority of mothers 28 (87.5%) had normal bonding with their child. Some mothers did, however, display a degree of anxiety concerning planned reconstructive surgery.

Conclusion: We found that having a child with a cleft lip with or without a cleft palate does not significantly influence maternal-infant bonding, however, further investigations using a larger sample size would be needed for further clarification.

Keywords

Breastfeeding; cleft lip with or without a cleft palate; maternal-infant bonding; reconstruction surgery anxiety

Introduction

Orofacial clefts are amongst the most common birth defects worldwide affecting 1 in 700 newborn babies (Mossey and Modell, 2012). An orofacial cleft refers to a defect in the normal fusion of the lip and palate that results from arrested embryonic development in the first trimester. Clefts are referred to as syndromic when one or more additional defects are associated, but the majority are non-syndromic where the cleft occurs in the absence of other birth defects. Non-syndromic orofacial clefts include cleft lip (CL) only, cleft lip with a cleft palate (CLP) and cleft palate (CP) only (Stanier and Moore, 2004). Clefts can be categorized as unilateral or bilateral (involving one or both sides of the lip and/or palate). Males are more commonly affected by a CLP whilst females are predominantly affected with an isolated CP (Hopper et al., 2007).

The aetiology of the development of a non-syndromic CLP remains unclear and most are believed to result from a combination of genetic factors and prenatal teratogenic exposure including tobacco, retinoic acid and anticonvulsant medication (Mossey et al., 2009). Babies born with a CLP require extensive and coordinated care from a multidisciplinary team to optimise management outcome.

Newborn babies benefit from the physical closeness of breastfeeding and it has been established that mother-infant attachment and successful breastfeeding are closely linked (Phuma-Ngaiyaye and Kalembo, 2016). Feeding children with a CLP in the neonatal period is known to be difficult (Britton et al., 2006), and this may pose a challenge in the bonding process between mothers and their infants.

Having a child with CL or CLP is stressful in many ways; parents need to cope with the anxiety and shock of having a baby with a facial anomaly which may lead to feelings of anger, guilt and depression (Black et al., 2009). Sischo et al. (2015) indicated that parents of children with chronic conditions such as CLP often experience five stages of grief: shock denial, anger, adaptation and reorganization. The burden of caring for a child with CLP is substantial due to the ongoing evaluation, treatment and hospitalization which begins during infancy and extends over a period of time, often into young adulthood (Sischo et al., 2015).

Maternal-infant bonding is defined as the development of the reciprocal relationship between mother and child (Spinner, 1978). Bonding is a physical and psychological experience for both

mothers and babies that provides comfort and protection. The mother is considered the "safe haven" for the establishment of the first emotional attachments of the child (Perrelli et al., 2014). Multiple factors have been associated with maternal-infant bonding such as level of education, employment, social support, parity, type of residence, pregnancy planning status as well as gender of the baby and feeding pattern (Rizk, 2016). Craniofacial anomalies may interfere with the initiation of parental nurturing which could impair bonding; structural facial abnormalities that affect the mouth may impede the recognition of smile responses or distort the vocalization necessary for the attachment process (Pillemer and Cook, 1989). It has also been hypothesized that affected children may be at risk for psychological problems and the bonding deficiency that they may encounter early in life may not be easily overcome, even with reconstructive surgery and improved facial appearance later in life (Perillo et al., 2017).

In general, there is a paucity of recent literature on how CLP affects maternal-infant bonding. In addition, this subject has never been researched in South Africa. Therefore the aim of this study was to describe bonding between mothers and their infants with a cleft lip with or without a cleft palate (CL/P) and to determine whether any demographic factors were associated with bonding impairment.

Materials and Methods

Study design and setting

This was a descriptive study and non-probability purposive sampling was used based on the availability and willingness of subjects to participate. The study population comprised of mothers who had an infant with a CL/P in Johannesburg, Gauteng, South Africa. Participation was voluntary and mothers were recruited from the Cleft Lip and Palate Clinic and Speech Therapy Departments at Charlotte Maxeke Johannesburg Academic Hospital (CMJAH) and Chris Hani Baragwanath Academic Hospital (CHBAH). A total of 32 participants were recruited into the study over a three month period spanning from May to July 2017.

Study participants

Mothers of any age, sociodemographic status, ethnicity or religion were eligible and they had to be proficient in English. Mothers were included if they have a biological child with a CL/P, and if

the child was three years of age or younger, irrespective of whether reconstructive surgery had been performed or not. Mothers whose infants had other birth defects or were diagnosed with a known syndrome were excluded from the study, as were mothers whose infant had an isolated CP, as this does not present with a visible facial deformity.

Research tool and data collection process

The research tool comprised of two structured questionnaires; a *Demographic Questionnaire* consisting of 19 items was administered which assessed socioeconomic status and maternal characteristics and the *Postpartum Bonding Questionnaire* (PBQ), designed by Brockington et al. (2001). The PBQ is one of the most frequently utilised tools used to evaluate a mother's emotional response to her baby in the postpartum period (Perrelli et al., 2014). The questionnaire consists of 25 items that are assessed on a 6-point Likert scale. The aim of this tool is to identify problems in the relationship between mother and infant based on four components: 1) impaired bonding, 2) rejection and anger, 3) anxiety about caring for the baby, and 4) risk of abuse (Brockington et al., 2001). The Risk of Abuse scale has been found to be unhelpful in identifying mothers considering abuse (Wittkowski et al., 2007) and was eliminated from the assessment, making the questionnaire 23 items in length. The questionnaires were administered in English in an interview format by the principle investigator to ensure that accurate information was received by the participant and to answer any questions that may have arisen during the completion of the questionnaires.

Exposure variables

Household socioeconomic status (SES) indicators in the form of data on household assets were collected from the mother in the demographic questionnaire. This scoring system has been successfully used before in South African studies (Jones et al., 2008; Feeley et al., 2013). The information collected included access to electricity and ownership of electronic items (i.e. a fridge, microwave, cell phone, television, washing machine, car). The household assets were categorized as binary variables; if a participant had a particular household durable item they scored 1, and if they did not have the household asset, they scored 0.

Statistical analysis

Data were analysed using Microsoft Excel 2016 and STATA statistical software package version 15 (StataCorp LP). Prior to any statistical analysis normality of continuous variables (mothers' ages, SES scores, number of children per mother and infant ages) was assessed using the Shapiro-Wilk test. Sample characteristics were summarized using descriptive statistics for each variable. Normally distributed data were reported as means ± standard deviation (SD), and data that was not normally distributed were reported as medians and interquartile ranges (IQR). For categorical variables, frequencies and percentages are presented. Associations between categorical variables and bonding were assessed using the Fisher Exact Test. Associations between bonding and normally distributed continuous variables were assessed using the T-test and the Wilcoxon-Mann-Whitney Test was used for variables that were not normally distributed. A p-value of 0.05 or less was considered significant.

Ethics

Ethical clearance was obtained from The Human Research Ethics Committee (Medical) at the University of the Witwatersrand (Clearance Certificate Number M170258). Study participants gave written informed consent and were provided with an information document explaining the study with a contact number should they have any questions. Confidentiality was maintained during all stages of the research by ensuring that no names were disclosed and participants were allocated an identification number which was used on all questionnaires.

Results

Participant demographics

A total of 32 mothers participated in the study. The mothers' ages ranged between 19 years and 41 years with a mean age of 28.72 years ± 6.50 years. The majority of mothers (24/32; 75%) were black African women who had completed high school (15/32; 47%). A large proportion of the mothers were unemployed (28/32; 88%) at the time the study was conducted and half (16/32; 50%) received child support grants from the government. Eighteen (56%) of the mothers were single and 14 (44%) were either married or living with their partner. The mean number of

children per mother was 2.22 ±1.24; primiparous mothers represented the largest group (12/32; 37%). A comprehensive summary of the demographic findings can be seen in Table 1.

Half (16/32; 50%) of the infants had undergone CL repair. Twenty-two (69%) of the mothers reported that they were not able to breastfeed. The reasons they gave for not breastfeeding were mostly due to the CL or CP. Some mothers reported that they did not produce milk and were thus unable to breastfeed. Only two (6%) of the mothers received the diagnosis of a CL/P prenatally (Table 1).

Table 1. *Association between demographic factors and bonding in mothers with normal bonding and mothers with impaired bonding.*

Characteristics	**Total**	**Mothers with normal bonding**	**Mothers with impaired bonding**	**P-value**
	N = 32	**N = 28**	**N = 4**	
Mother's age (years) mean ± SD	28.72 ± 6.5	28.93 ± 6.27	27.25 ± 7.79	**0.63[a]**
Marital status				**0.79[b]**
Single	18 (56%)	15 (54%)	3 (75%)	
Cohabiting/married	14 (44%)	13 (46%)	1 (25%)	
Population group				**0.66[b]**
Black African	24 (75%)	20 (71%)	4 (100%)	
White	6 (19%)	1 (4%)	0 (0%)	
Indian	1 (3%)	1 (4%)	0 (0%)	
Coloured	1 (3%)	6 (21%)	0 (0%)	
Level of education				**0.32[b]**
Primary school	1 (3%)	1 (4%)	0 (0%)	
Some secondary schooling	10 (31%)	7 (25%)	3 (75%)	
Completed secondary school	15 (47%)	14 (50%)	1 (25%)	
Tertiary	6 (19%)	6 (21%)	0 (0%)	
Occupation				**1.00[b]**
Working	4 (12%)	4 (14%)	0 (0%)	
Not working	28 (88%)	24 (86%)	4 (100%)	
Socioeconomic status mean ± SD	8.25 ± 3.26	8.50 ± 2.87	7.53 ± 4.24	**0.67[a]**
Social Grant				**0.60[b]**
Child support grant	16 (50%)	13 (46%)	3 (75%)	
No grant	16 (50%)	15 (54%)	1 (25%)	
Type of residence				**0.68[b]**
Flat	4 (12%)	4 (14%)	0 (0%)	
House	16 (50%)	13 (46%)	3 (75%)	
Rent	7 (22%)	7 (25%)	0 (0%)	
Informal settlement	5 (16%)	4 (14%)	1 (25%)	
Number of children mean ± SD	2.22 ±1.24	2.25 ±1.14	2 ±2.00	**0.058[b]**
One	12 (37%)	4 (14%)	3 (75%)	

Two	8 (25%)	7 (25%)	0 (0%)	
Three	7 (22%)	9 (33%)	0 (0%)	
Four or more	5 (16%)	8 (27%)	1 (25%)	
Age of child with cleft (months) median (IQR)	7.00 (1.17-15.25)	7.00 (1.00-15.00)	17.50 (8.75-27.00)	**0.09[c]**
Mode of delivery				**0.55[b]**
Normal vaginal delivery	24 (75%)	20 (71%)	4 (100%)	
Caesarean section	8 (25%)	8 (29%)	0 (0%)	
Gender of child with cleft				**1.00[b]**
Male	22 (69%)	19 (68%)	3 (75%)	
Female	10 (31%)	9 (32%)	1 (25%)	
Cleft lip with/without palate				**1.00[b]**
Cleft lip	9 (28%)	8 (29%)	1 (25%)	
Cleft lip and palate	23 (72%)	20 (71%)	3 (75%)	
Type of cleft				**0.30[b]**
Unilateral	23 (72%)	19 (68%)	4 (100%)	
Bilateral	9 (28%)	9 (32%)	0 (0%)	
Cleft repaired				**0.61[b]**
Yes	16 (50%)	13 (46%)	3 (75%)	
No	16 (50%)	15 (54%)	1 (25%)	
Breastfeed				**0.11[b]**
Yes	10 (31%)	8 (29%)	3 (75%)	
No	22 (69%)	20 (71%)	1 (25%)	
Diagnosed				**1.00[b]**
Prenatally	2 (6%)	2 (7%)	0 (0%)	
Postnatally	30 (94%)	26 (93%)	4 (100%)	

[a]T-test; [b]Fisher Exact Test; [c]Wilcoxon-Mann-Whitney Test

Regarding infants with CL/P

Twenty four (75%) of the mothers reported that their child with the CL/P was born by normal vaginal delivery while the remainder had a Caesarean section. There were more than double the number of affected male infants than females (69% vs 31%). The majority of infants had a unilateral CLP, followed by infants with a bilateral CLP and unilateral CL (Table 2).

Table 2. *Types of clefts observed in study population.*

	Bilateral N (%)	**Unilateral N (%)**	**Total N (%)**
Cleft lip	1 (3%)	8(25%)	9 (28%)
Cleft lip and palate	8 (25%)	15 (47%)	23(72%)
Total	9 (28%)	23 (72%)	**32**

Scores on the Postpartum Bonding Questionnaire

Of the 32 mothers participating in the study, 28 (88%) did not show an indication of impaired bonding on the PBQ. Four (13%) mothers did show a measure of impaired bonding in one or more of the three scales. Of the four mothers who demonstrated impaired bonding, two (6%) had scores higher than 11 (scores of 12 and 13) for scale 1 (general impaired bonding) only, and two (6%) mothers had scores above the cut-off values for scale 1 and scale 3 (anxiety about care). The mean scores for mothers with normal bonding and mothers with impaired bonding are shown in Table 3.

Table 3. *Mean PBQ scores for mothers with and without impaired bonding.*

Groups	**Count**	**Scale**		
		1	2	3
Range of scores		0-59	0-34	0-20
Cut-off point		>11	>16	>9
Mothers without impaired bonding	28	4.57 ± 3.11	0.46 ± 0.73	2.39 ± 2.27
Range of scores		0-9	0-2	0-9
Mothers with impaired bonding	4	15.5 ± 3.04	7.25 ± 4.66	7 ± 5.24
Range of scores		12-19	2-14	10-14
Total scores (all mothers)	32	**5.94 ± 4.76**	**1.31 ± 2.87**	**2.97 ± 3.21**

[a]Results given as means ± SD or medians (IQR)

In the four mothers who indicated impaired bonding, all were of black African ancestry, unemployed and three of the four mothers were single parents. The infants of these mothers were delivered by normal vaginal delivery and diagnosed postnatally. Three (75%) of the mothers reported that the affected baby was their first child and three (75%) were affected by a unilateral CLP.

Association between demographic factors and bonding impairment

A comparison was made between the demographic factors of mothers with normal bonding and mothers with impaired bonding. No significant association was observed between any of the variables and maternal-infant bonding. Table 1 summarises the relationship between the demographic variables and the bonding scale.

Discussion

The feelings mothers experience after a child with a CLP is born can often be characterized as discouraging and confusing. Parents are unprepared for the challenging situation and the complications that are associated with having a baby with a CLP (Kuttenberger et al., 2010).

We anticipated that there would be higher levels of impaired bonding in mothers with children with CL/P due to the psychological impact and the associated complex challenges. It was also thought that some of the sociodemographic factors would be strongly associated with impaired bonding. However, in our study population, we observed that the majority of mothers had normal bonding with their infants.

The reasons for the four mothers showing a level of impaired bonding might be due to other causes, including the mother's relationship with her partner, poor socioeconomic status or unemployment. These may be contributing factors for impaired bonding or may have exacerbated the bonding impairment. A study conducted in Pennsylvania, USA, using the PBQ in a cohort of first-time mothers, revealed a socioeconomic bias in that older, more educated women who were not living in poverty and were married, reported better maternal-infant bonding (lower scores) (Kinsey et al., 2014). Support from the father of the baby, as well as social support, also resulted in better bonding. None of the sociodemographic factors showed a significant association with impaired bonding in our study sample, although the sample size was small.

Drotar et al. (1975) explained that due to the increased anxiety mothers experience when having a child with a congenital malformation, they typically described their relationship with their child as "closer than normal". This in turn can lead to an overly close relationship which develops between a mother and a child with an abnormality (Drotar et al., 1975). This may be an explanation for the normal bonding observed in the mothers in our study.

There was no statistical significance in the scores of mothers with babies who had undergone cleft repair and those who had not. However, from the PBQ, the question that had the highest score overall was question 19, "My baby makes me anxious". This may be attributed to the concern about potential surgical procedures as well as anxiety around feeding difficulties. Comments made during the interviews suggested that the mothers were anxious about the upcoming surgery for the cleft repair. A study by Kuttenberger et al. (2010) indicated that

information about surgery and feeding are most frequently mentioned as the most important information that the parents require. According to Kuttenberger et al. (2010), the majority of parents prefer to see photographs of other children who have had surgery and also appreciate information about cleft patient organizations and support groups. In order to reduce anxiety and uncertainty in parents, clear and consistent information needs to be provided about the anomaly, treatment, surgery and the prognosis (Kuttenberger et al., 2010).

Currently in South Africa, there appears to be a lack of comprehensive preparation for parents of children undergoing surgery to alleviate feelings of anxiety and concern about the actual procedure. These preparation processes mentioned above could be adopted for parents of children undergoing any type of surgery. It is suggested from the current study and other studies that there is a need to counsel the patient and family to discuss pre and post-surgery concerns and discuss the relevant information (De Sousa et al., 2009). Empowering the parents, and teaching them the essentials of cleft care in the early stages after the baby is born will help alleviate the initial fear and feelings of hopelessness (Rey-Bellet and Hohlfeld, 2004).

Most mothers who did not breastfeed their babies reported that it was due to the CP which made feeding often difficult or not possible. This might have contributed to the increase in anxiety scores of the mothers with infants with CLP. In many cases, mother and child are temporarily separated after birth because of assumed feeding problems (Kuttenberger et al., 2010). Literature shows that an inability to breastfeed is related to an increase in anxiety and depression in postpartum mothers (Ystrom, 2012). Initial counselling after the birth of a baby with a cleft is of significant importance for parents who need extra encouragement and support during this vulnerable period (Kuttenberger et al., 2010). These are important matters for doctors, nurses, midwifes and genetic counsellors to consider in the initial consultation with a mother with a child with a CL/P. Focussing on pre- and post-surgical preparation can also be of great value to parents.

There was no association between bonding and mothers who knew the diagnosis prenatally compared to those diagnosed postnatally. These findings may have been affected by the small sample size. It has been shown that there are some advantages in receiving a diagnosis prenatally as parents can prepare physically and emotionally for the birth of a baby with an anomaly, which may decrease anxiety levels. Rey-Bellet and Hohlfeld (2004) showed that in a group of couples who received a diagnosis of CL/P prenatally, 96% found the early diagnosis to be beneficial. The

reason for the majority of mothers in our study not receiving a diagnosis of CL/P prenatally could be due to numerous factors. In Johannesburg, most local antenatal clinics in the public healthcare sector (at which most women receive their antenatal care) do not have the functional equipment or trained personnel to offer detailed ultrasound which may be able to detect a CL/P.

Although the PBQ is a well-established and validated questionnaire, a mother's subjective account based on a self-rating instrument will not be able to reliably assess the mother-infant relationship entirely. The gold standard will still remain direct clinical observation (Brockington et al., 2001). Further research could explore a wider range of sociodemographic factors such as support from the father of the baby, trauma during birth and maternal stress and depression.

Limitations

The small sample size obtained was due to time constraints and language barriers as English is not the first language in the majority of the South African population. Mothers attending regular follow-up appointments at the clinics affected the recruitment of new participants and limited the sample size, as well as the fact that infants were brought to the clinic by caregivers and family members and not the biological mothers. This may have caused a bias towards recruitment of mothers who are unemployed (87% unemployed vs 13% employed). Limitations related to generalizability of this study include, the overrepresentation of mothers who are committed to the management of their infant with a cleft lip by attending specialized clinics. Recruitment of mothers who are proficient in English may have introduced a bias, and mothers who might have benefitted the study may have been excluded due to their inability to answer the questionnaire effectively. The small sample size affected the power of this study and in further research a larger sample size will increase the statistical significance. The inclusion of a comparison group could strengthen the study.

Conclusion

Of all the mothers that had an infant with a CL/P, only four (12.5%) showed a measure of impaired bonding. The majority of mothers had good bonding with their infants with CL/P. The authors felt that this demonstrated the significance of prenatal maternal-infant bonding and the unconditional acceptance of a mother for her child. In this study, a need was identified for improved preparation and counselling for parents with regards to the reconstructive surgical

process. Further research in a larger cohort is required to make more definitive conclusions and to substantiate the findings in this study with regards to the effect of a baby's CL on maternal-infant bonding. However, this research study has provided an indication of the current status of maternal-infant bonding in a group of mothers from Johannesburg. In summary, it was established from this study that having a child with a CL/P does not have a major influence on the bonding relationship between mother and infant.

References

Black JD, Girotto JA, Chapman KE & Oppenheimer AJ (2009) When my child was born: cross-cultural reactions to the birth of a child with cleft lip and/or palate. *Cleft Palate Craniofacial Journal,* 46**,** 545-8.

Britton JR, Britton HL & Gronwaldt V (2006) Breastfeeding, sensitivity, and attachment. *Pediatrics,* 118**,** e1436-e1443.

Brockington IF, Oates J, George S, Turner D, Vostanis P, Sullivan M, Loh C & Murdoch C (2001) A Screening Questionnaire for mother-infant bonding disorders. *Archives of Women's Mental Health,* 3**,** 133-140.

De Sousa A, Devare S & Ghanshani J (2009) Psychological issues in cleft lip and cleft palate. *Journal of Indian Association of Pediatric Surgeons,* 14**,** 55.

Drotar D, Baskiewicz A, Irvin N, Kennell J & Klaus M (1975) The adaptation of parents to the birth of an infant with a congenital malformation: A hypothetical model. *Pediatrics,* 56**,** 710-717.

Feeley AB, Musenge E, Pettifor JM & Norris SA (2013) Investigation into longitudinal dietary behaviours and household socio-economic indicators and their association with BMI Z-score and fat mass in South African adolescents: the Birth to Twenty (Bt20) cohort. *Public health nutrition,* 16**,** 693-703.

Figueiredo B, Costa R, Pacheco A & Pais Á (2009) Mother-to-infant emotional involvement at birth. *Maternal and Child Health Journal,* 13**,** 539-549.

Hopper RA, Cutting C & Grayson B (2007) Cleft lip and palate. *Grabb and Smith's Plastic Surgery. 6th Edition. Philladelphia: Lippincott Williams and Wilkins,* 201.

Jones LL, Griffiths PL, Adair LS, Norris SA, Richter LM & Cameron N (2008) A comparison of the socio-economic determinants of growth retardation in South African and Filipino infants. *Public health nutrition,* 11**,** 1220-1228.

Kinsey CB, Baptiste-Roberts K, Zhu J & Kjerulff KH (2014) Birth-related, psychosocial, and emotional correlates of positive maternal–infant bonding in a cohort of first-time mothers. *Midwifery,* 30**,** e188-e194.

Kuttenberger J, Ohmer J & Polska E (2010) Initial counselling for cleft lip and palate: Parents' evaluation, needs and expectations. *International journal of oral and maxillofacial surgery,* 39**,** 214-220.

Moehler E, Brunner R, Wiebel A, Reck C & Resch F (2006) Maternal depressive symptoms in the postnatal period are associated with long-term impairment of mother–child bonding. *Archives of women's mental health,* 9**,** 273-278.

Mossey PA, Little J, Munger RG, Dixon MJ & Shaw WC (2009) Cleft lip and palate. *The Lancet,* 374**,** 1773-1785.

Mossey PA & Modell B (2012) Epidemiology of oral clefts 2012: an international perspective. *Cleft Lip and Palate.* Karger Publishers.

Perillo L, D'apuzzo F, Eslami S & Jamilian A (2017) Cleft Lip and Palate Patients: Diagnosis and Treatment. *Designing Strategies for Cleft Lip and Palate Care.* InTech.

Perrelli JGA, Zambaldi CF, Cantilino A & Sougey EB (2014) Mother-child bonding assessment tools. *Revista Paulista de Pediatria,* 32**,** 257-265.

Phuma-Ngaiyaye E & Kalembo FW (2016) Supporting mothers to bond with their newborn babies: Strategies used in a neonatal intensive care unit at a tertiary hospital in Malawi. *International Journal of Nursing Sciences,* 3**,** 362-366.

Pillemer FG & Cook K (1989) The psychosocial adjustment of pediatric craniofacial patients after surgery. *Cleft Palate J,* 26**,** 201-7.

Rey-Bellet C & Hohlfeld J (2004) Prenatal diagnosis of facial clefts: evaluation of a specialised counselling. *Swiss medical weekly,* 134**,** 640-644.

Rizk SA (2016) Factors Associated with Maternal-infant Attachment One Month Postnatally. *Journal of High Institute of Public Health,* 42**,** 103-118.

Sischo L, Broder HL & Phillips C (2015) Coping with cleft: A conceptual framework of caregiver responses to nasoalveolar molding. *The Cleft Palate-Craniofacial Journal,* 52**,** 640-650.

Spinner MR (1978) Maternal-infant bonding. *Canadian Family Physician,* 24**,** 1151.

Stanier P & Moore GE (2004) Genetics of cleft lip and palate: syndromic genes contribute to the incidence of non-syndromic clefts. *Human molecular genetics,* 13**,** R73-R81.

Wittkowski A, Wieck A & Mann S (2007) An evaluation of two bonding questionnaires: a comparison of the Mother-to-Infant Bonding Scale with the Postpartum Bonding Questionnaire in a sample of primiparous mothers. *Archives of Women's Mental Health,* 10**,** 171-5.

Ystrom E (2012) Breastfeeding cessation and symptoms of anxiety and depression: a longitudinal cohort study. *BMC pregnancy and childbirth,* 12**,** 36.

CANDIDATE'S SURNAME: Nienaber	FIRST NAME/S: Elzette	STUDENT NUMBER: 762941
CURRENT QUALIFICATIONS: MSc (Med) Molecular and Cellular Biology		
TEL: 011 489 9223	CELL: 083 604 4036	**E-MAIL**: elzettenienaber@gmail.com
DEGREE FOR WHICH PROTOCOL IS BEING SUBMITTED: MSc (Med) Genetic Counselling		
PART-TIME OR FULL-TIME: Full-time		
FIRST REGISTERED FOR THIS DEGREE:	TERM : 1	YEAR: 2016
DEPARTMENT: Human Genetics		
TITLE OF PROPOSED RESEARCH: Assessment of maternal-infant bonding between mothers and their babies with a cleft lip.		
CANDIDATE'S SIGNATURE: Nienaber		DATE: 27/01/2017
SUPERVISOR #1'S NAME: Merlyn Glass		
% OF SUPERVISION TO BE PROVIDED BY SUPERVISOR #1: 50%		
SUPERVISOR #1'S QUALIFICATIONS: RN, RM, Dip Paeds, MSc (Nursing Education)		
SUPERVISOR #1'S DEPARTMENT: Human Genetics		
SUPERVISOR #1'S ADDRESS / TEL / **E-MAIL**: Room 4, First Floor, Jack Metz Building, Cnr Hospital & De Korte Str, Braamfontein/ Tel: 011 489 9335/Email:merlyn.glass@nhls.ac.za		
SUPERVISOR #2'S NAME: Lindsay Lambie		
% OF SUPERVISION TO BE PROVIDED BY SUPERVISOR #2: 50%		
SUPERVISOR #2'S QUALIFICATIONS: MBBCh, DCH(SA), FCPaed(SA), Cert(Medical Genetics)(SA), MSc (Med)(Genetic Counselling).		
SUPERVISOR #2'S DEPARTMENT: Human Genetics		
SUPERVISOR #2'S ADDRESS / TEL / **E-MAIL**: Room 3, First Floor, Jack Metz Building, Cnr Hospital & De Korte Str, Braamfontein/ Tel: 0114899336/Email:Lindsay.lambie@nhls.ac.za		

SYNOPSIS OF RESEARCH:

Orofacial clefts are among the most common birth defects worldwide. Some studies suggest that having a child with a cleft lip with/without a cleft palate may have detrimental effects on parent-child bonding. Yet other research found that "children with clefts and their mothers are doing as well as families without cleft with regards to emotional development and mother-child relationships". Little is known, however, about the effect of cleft lip and palate on mother-infant bonding in the South African context.The aim of this study is to describe maternal-infant bonding between mothers and their infants with a cleft lip, as well as to explore risk factors which may affect bonding. The study population will comprise of mothers who have an infant with a cleft lip with/without a cleft palate in Johannesburg, Gauteng, South Africa. The research tool will comprise two structured questionnaires, a Demographic Questionnaire and the Postpartum Bonding Questionnaire. The data will be analysed quantitatively. This research could facilitate the understanding of how a birth defect, such as a cleft lip may affect mothers' initial bonding experiences with their infants in South Africa. It could assist genetic counsellors to address potential concerns and make parents aware of the potential difficulties, thus assisting them to establish a better relationship with their babies.

ETHICS PENDING: Ψ ETHICS APPROVED: Ψ (circle appropriate symbol)	IF Y SUPPLY ETHICS CLEARANCE No:
SIGNATURE OF SUPERVISOR/S: MGlass	Lambie

Assessment of maternal-infant bonding between mothers and their babies with a cleft lip

Elzette Nienaber

762941

MSc. (Med) Genetic Counselling

Supervisor 1

Merlyn Glass

(RN, RM, Dip Paeds, MSc) (Nursing Education)

Supervisor 2

Dr Lindsay Lambie

MBBCh, DCH(SA),FCPaed(SA),Cert(Medical Genetics)(SA), MScMed(Genetic Counselling)

1. Background

1.1 Introduction to cleft lip and palate

Orofacial clefts are among the most common birth defects worldwide and one in 700 new born babies have a cleft lip or palate or both (Tolarová and Cervenka, 1998). They represent a heterogeneous group of disorders affecting the lips and oral cavity (Mossey et al., 2009). Orofacial clefts refer to a defect or opening in the normal fusion of the lip and palate that results from arrested embryonic development in the first trimester. Clefts are referred to as syndromic where one or more additional defects are associated. The majority are non-syndromic where the cleft lip and/or palate occur in the absence of other birth defects. Non-syndromic orofacial clefts include, cleft lip only (CL), cleft lip with a cleft palate (CLP) and cleft palate only (CP) (Stanier and Moore, 2004).

1.2 Epidemiology of cleft lip and palate

Clefts of the lip and palate are divided into two general groups, cleft lip with or without palate and isolated cleft palate. Occurrence estimates range between one in 1 500 for cleft palate only and between one in 300 and one in 2 500 for cleft lip with or without palate (Wyszynski et al., 1996). The most common is CLP (46%), followed by isolated CP (33%) and lastly isolated CL (21%). Clefts can be categorized as unilateral or bilateral (involving one or both sides of the lip and/or palate), and as 'incomplete' or 'complete' (involving only the soft palate versus both the soft and hard palates, and/or involving only the lip versus the lip and gumline) (Hopper et al., 2007).

The occurrence of a unilateral cleft is nine times more common than a bilateral cleft and occurs mostly on the left side. Males are more commonly affected by a cleft lip and palate whilst females are predominantly affected with isolated cleft palates (Hopper et al., 2007).

1.3 Aetiology of cleft lip and palate

The aetiology of CLP is complex and is thought to involve genetic factors with variable influences from the environment. The majority of clefts are non-syndromic and thus not associated with any other birth defects. The aetiology for the development of a non-syndromic cleft lip or palate remains unclear and most are believed to result from a combination of genetic factors and prenatal teratogenic exposure including tobacco, retinoic acid and anticonvulsant medication (Mossey et al., 2009). The multigenic inheritance combined with non-genetic factors makes it challenging to identifying key genes involved in CLP. Through linkage studies, various loci have been reported to have a causal role in the formation of CLP (Prescott et al., 2000). A meta-analysis of 13 genome scans by Marazita et al. (2004) revealed multiple CLP genes on 16 chromosomal regions. Although there is a definite genetic component influencing the development of non-syndromic cleft lip and/or palate, it still seems to be attributed to multifactorial factors (Hopper et al., 2007).

1.4 Management

Babies born with a cleft lip or palate require extensive and coordinated care from a multidisciplinary team to optimise outcome. Clefts may be detected by ultrasound in utero, but many cases are still being missed and the diagnosis is often only made after birth. Cleft lip is normally repaired surgically by five months of age, followed by the cleft palate repair when children are between 12 and 18 months old. Surgical revision and follow up procedures are scheduled according to the patients' needs.

New-born babies benefit from the physical closeness of breastfeeding and it has been established that there is a link between mother-infant attachment and breastfeeding, however, feeding children with a CLP in the neonatal period is known to be difficult (Britton et al., 2006). This may also pose a challenge in the bonding process of mothers and their infants. The effects of CLP on appearance, speech, hearing and the psychological impact can lead to long-lasting adverse effects for health and

social integration (Mossey et al., 2009). Rehabilitation is possible with good and effective health care; orofacial clefts can pose a great burden on the individual, family and society.

1.5 Mother-infant bonding

Maternal-infant bonding is defined as the development of the reciprocal relationship between mother and child (Spinner, 1978). Bonding is a physical and psychological experience for both mothers and babies which provides comfort and protection. The mother is considered the "safe haven" for the establishment of the first emotional attachments of the child (Biaggio et al. 2001). Having a child with CLP is stressful in many ways; parents need to cope with the anxiety and shock of having a baby with a facial anomaly which may lead to feelings of anger, guilt and depression. Research has indicated that parents with children with chronic conditions such as cleft lip and/or palate often experience five stages of grief: shock denial, anger, adaptation and reorganization (Sischo et al., 2015). As well as coping with the reality of having a child who is "different", parents must also endure the added care associated with CLP which reportedly increases parental stress. The burden of caring for a child with CLP is substantial due to the ongoing evaluation, treatment and hospitalisation which begins during infancy and extends over a period of time, often involving young adulthood (Sischo et al., 2015).

The birth of a baby with a chronic condition such as a CLP has an immense impact, not only on the parents, but on the entire family and can profoundly disrupt the family system (Sischo et al., 2015). St. John et al., (2003), found that divorce rates are higher in families with a child with a craniofacial anomaly compared to a control group. Another study, however, concluded that families with children with chronic conditions fare the same if not better than the healthy control group (Rodrigues and Patterson, 2007). The effect/impact that a child with CLP has on the family structure and function may be influenced by the type of cleft, the characteristics of the family (such as age of the child, number of

children and income), background factors (such as family values), social support and cultural issues (Sischo et al., 2015).

In addition to parental stress and the impact on family system, the effect of a CLP on parent-infant bonding and interaction has been explored (Sischo et al., 2015). Some studies suggest that having a child with a CLP may have detrimental effects on parent-child bonding. Murray et al. (2008) found that mothers of children with a cleft have less maternal sensitivity towards their children compared with controls. Late surgical repair of a child's cleft also resulted in mothers who were less positively involved and looked at their children less often than the controls did (Murray et al., 2008). Yet other research found that "children with clefts and their mothers are doing as well as families without cleft with regards to emotional development and mother-child relationships" (Habersaat et al., 2013). Little is known, however, about the effect of CLP on maternal-infant bonding in the South African context.

2. Study objectives

2.1 Aim

The aim of this study is to describe maternal-infant bonding between mothers and their infants with a cleft lip attending specialised clinics at Johannesburg State Hospitals, as well as to explore risk factors which may affect bonding.

2.2 Objectives

- To determine whether there is bonding impairment in the maternal-infant relationship in mothers with infants with a cleft lip attending clinics at Johannesburg State Hospitals.
- To explore whether sociodemographic factors influence bonding in mothers with infants with a cleft lip.
- To establish if there is better maternal-infant bonding when infants have had their cleft repaired.

- To establish whether breastfeeding was possible and if not, was the cleft lip the reason for this.
- To determine whether there is a difference in maternal-infant bonding in mothers who knew the diagnosis prenatally, compared to mothers who did not know the diagnosis prenatally.

3. Methodology

3.1 Study participants and sampling

The study population will comprise of mothers who have an infant with a cleft lip with/without a cleft palate in Johannesburg, Gauteng, South Africa. Participation will be voluntary and mothers will be recruited from the cleft lip and palate clinic and speech therapy departments at Charlotte Maxeke Johannesburg Academic Hospital (CMJAH) and Chris Hani Baragwanath Academic Hospital (CHBAH). Between 30 and 60 participants will be recruited from these clinics.

Non-probability, purposive sampling will be used based on the availability and willingness of subjects to participate in the study. A verbal explanation will be given by the principle investigator (PI) describing the aim of the study and what will be required of them after which, any queries will be clarified. Participants will be provided with an information document with all the details of the study (Appendix A). The individuals who are willing to participate in the study, and who meet the inclusion criteria, will be required to sign informed consent (Appendix B). The questionnaires will be administered in an interview format by the PI to ensure accurate information is obtained by the participant and to answer any questions that may arise during the completion of the questionnaire. A separate room will be arranged in the respective departments where the questionnaires will be completed to ensure confidentiality. The questionnaires that will be used in this study will require approximately 20 minutes to complete. Participants will not be disadvantaged in any way with regards to their waiting time or treatment if they choose to participate in the study. During the recruitment and

questionnaire process, if participants are identified that require further counselling or support, referrals will be made to the appropriate professionals. A Genetic Counselling pamphlet with information about the service and contact information will be given to participants should they wish to seek additional service.

Inclusion criteria

- Women of any age, sociodemographic group, ethnicity or religion.
- Mothers with
    - A biological child with a cleft lip with/without cleft palate.
    - An infant or child younger than 3 years of age, irrespective of whether reconstructive surgery has been performed.
- Women who are able to communicate proficiently in English.

Exclusion criteria

- Mothers with infants/children who have other birth defects or have been diagnosed with a known syndrome.
- Infants with an isolated cleft palate.

3.2 Study Design

This will be a non-experimental study as there is no manipulation of the independent variable, nor is the setting controlled (Brink et al., 2006). The study will be a quantitative study focussing on a combination of descriptive and correlation research designs. The descriptive design will describe the variables in the data. It will be used to obtain the first objectives set out in this study. A descriptive correlational design will be used to determine and describe what relationships exist between independent and dependent variables and address the remaining objectives. The independent variable

will be the mothers with an infant with a cleft lip. The dependent variables will be the level of impaired bonding and the sociodemographic factors.

3.3 Research tool and data collection

The research tool will comprise two structured questionnaires. The questionnaires will be administered by the PI at State Hospitals (CMJAH and CHBAH) in Johannesburg together with an information document and informed consent document.

Demographic Questionnaire: Mothers will complete a questionnaire designed to obtain basic demographic information consisting of 20-items which will help identify commonalities and differences within the group (Appendix C).

Postpartum Bonding Questionnaire (PBQ): The PBQ, designed by Brockington et al. (2001) is one of the most frequently used tools to evaluate the mother's emotional response in relation to the baby in the postpartum period (Perrelli et al., 2014). The questionnaire consists of 25-items that are assessed on a 6-point Likert scale. This tool aims to identify problems in the mother-baby relationship based on four components: 1) impaired bonding, 2) rejection and anger, 3) anxiety about caring for the baby, and 4) risk of abuse (Brockington et al., 2001). The Risk of Abuse Scale was eliminated from the assessment, making the questionnaire 23 items in length. The Risk of Abuse scale has been found unhelpful in identifying mothers considering abuse. In addition, mothers may feel more comfortable completing the assessment if these items were omitted. It is a validated and reliable tool used to identify dysfunction in the relationships between a mother and her child and provides a consistent and valid indication of the early emotional bond (Brockington et al., 2006; van Bussel et al., 2010) (Appendix D).

4. Data analysis

The data will be analysed quantitatively. Data from the Demographic Questionnaire will be summarised using descriptive statistics to become familiar with the data and to characterize our sample. The descriptive statistics such as frequency distributions, measures of central tendency, dispersion and variability will be used to describe and summarize variables obtained in the data.

Spearman's rank correlation coefficient will be used to assess the relationship between continuous variables such as bonding score and sociodemographic factors. These analyses will help determine whether there is a relationship between these variables and the bonding level. Statistical analysis will be done using software that is freely available. The data obtained from the DQ and PBQ will be captured in an Excel spreadsheet and data will be analysed using Excel 2016.

5. Ethics

An information document explaining the research will be distributed to participants. The participants will be informed that the results of this study may be published. Participation is voluntary and participants are free to withdraw from the study at any time with no impact on their treatment or management. Written informed consent will be obtained from all participants,

Confidentiality will be maintained during all stages of the research by ensuring that no names are disclosed. Participants will be assigned a specific participant number in the research report. This number will be on an additional form where participants give their information and contact details for the PI to get in contact with them if indicated. The participant will be made aware that they will be personally contacted by the PI should the results of the PBQ indicate potential impaired bonding between mother and child. They will be offered a referral to a non-profit centre in Johannesburg specialising in mother-infant bonding relationship and support (Ububele Baby Mat Project) (Frost,

2012) where a contact person has been identified. This may be viewed as a breach of confidentiality, making the participants hesitant to participate in the study or affect the outcome of the questionnaire.

The Heads of the Speech Therapy Departments as well as the Cleft Lip and Palate Clinic at CMJAH and CHBAH have been contacted and are in the process of giving written permission (Appendix E). Thereafter, permission will be obtained from the CEO's at CMJAH and CHBAH (Appendix F). The Human Research Ethics Committee at the University of the Witwatersrand will be approached for ethics approval on the 7th of February 2017.

6. Timing

This study will commence in as soon as ethical approval is obtained and the final research report will be submitted at the end of August 2017.

Chart 1: Gant chart indicating time expected to be taken on the various components.

	Dec	Jan	Feb	Mar	Apr	May	Jun	Jul	Aug	Sep	Oct	Nov
Protocol planning and design												
Internal Protocol Assessment												
Protocol amendments												
Application to Ethics												
Protocol Assessment												
Collecting Data												
Data Analysis												
Write up - research report												
Write up - paper												

7. Funding

Predicted budget

Expenses	Estimated Cost
Travelling cost from NHLS to recruiting clinics twice a week for a period of 12 weeks.	NHLS to CMJAH = 2.7 km NHLS to CHBAH= 16.3 km 19 km x 24 = 456 km @ 82.4c/km **Total = R 375.74**
Printing cost : Information document, Consent form, DQ, PBQ (6 pages) for a maximum of 60 participants.	6 x 60 = 360 2 x 80 = 160 Total of 520 pages @ 39c/page

Research report (max of 80 pages, 2 copies)	**Total = R 202.80**
Total	**R 578.54**

No funding is required for conducting this study as participation will be voluntary during attendance at the respective clinics. The research tool is freely available and statistical analysis will be done using available software. Travelling and printing costs will be covered by the PI.

8. Potential limitations.

A potential limitation of this study may be reaching the anticipated number of participants. This will depend on willingness to participate in the study, and finding participants who meet the inclusion criteria. The small sample size will also potentially limit the statistical power of this study.

The characteristics of mothers who agree to participate may confound the results. Mothers who feel they have the capacity to engage in the research report at this difficult time in their lives may represent those with higher coping mechanisms. This may affect the data by reflecting a highly motivated and more capable group than exists in the general population. The external validity may be affected by participant bias.

External validity may be affected by experimenter effects when the PI administers the questionnaires in an interview manner (when researcher characteristics or behaviour influence subject behaviour). Internal validity will be affected by the purposive selection bias.

Value of the study

This research could facilitate the understanding of how a birth defect, such as a cleft lip may affect mothers' initial bonding experiences with their infants in South Africa. It could assist genetic counsellors to address potential concerns and make parents aware of the potential difficulties, thus assisting them to establish a better relationship with their babies. The study will assist in the

implementation of coping strategies for parents to help deal with the initial shock, denial and anger, which often occurs when a baby is born with a birth defect. This study will make a valuable contribution in improving the understanding and effect that facial deformities may have on mother-infant bonding.

9. References

Brink, H., Van Der Walt, C. & Van Rensburg, G. 2006. *Fundamentals of research methodology for health care professionals*, Juta and Company Ltd.

Britton, J. R., Britton, H. L. & Gronwaldt, V. 2006. Breastfeeding, sensitivity, and attachment. *Pediatrics,* 118**,** e1436-e1443.

Brockington, I., Fraser, C. & Wilson, D. 2006. The postpartum bonding questionnaire: a validation. *Archives of women's mental health,* 9**,** 233-242.

Brockington, I. F., Oates, J., George, S., Turner, D., Vostanis, P., Sullivan, M., Loh, C. & Murdoch, C. 2001. A Screening Questionnaire for mother-infant bonding disorders. *Archives of Women's Mental Health,* 3**,** 133-140.

Frost, K. 2012. The Ububele Baby Mat Project: a community-based parent-infant intervention at primary health care clinics in Alexandra Township, Johannesburg. *South African Journal of Psychology,* 42**,** 608-616.

Habersaat, S., Monnier, M., Peter, C., Bolomey, L., Borghini, A., Despars, J., Pierrehumbert, B., Müller-Nix, C., Ansermet, F. & Hohlfeld, J. 2013. Early mother-child interaction and later quality of attachment in infants with an orofacial cleft compared to infants without cleft. *The Cleft Palate-Craniofacial Journal,* 50**,** 704-712.

Hopper, R. A., Cutting, C. & Grayson, B. 2007. Cleft lip and palate. *Grabb and Smith's Plastic Surgery. 6th Edition. Philladelphia: Lippincott Williams and Wilkins,* 201.

John, D. S., Pai, L., Belfer, M. L. & Mulliken, J. B. 2003. Effects of a child with a craniofacial anomaly on stability of the parental relationship. *Journal of Craniofacial Surgery,* 14**,** 704-708.

Marazita, M. L., Murray, J. C., Lidral, A. C., Arcos-Burgos, M., Cooper, M. E., Goldstein, T., Maher, B. S., Daack-Hirsch, S., Schultz, R. & Mansilla, M. A. 2004. Meta-analysis of 13 genome scans reveals multiple cleft lip/palate genes with novel loci on 9q21 and 2q32-35. *The American Journal of Human Genetics,* 75**,** 161-173.

Mossey, P. A., Little, J., Munger, R. G., Dixon, M. J. & Shaw, W. C. 2009. Cleft lip and palate. *The Lancet,* 374**,** 1773-1785.

Murray, L., Hentges, F., Hill, J., Karpf, J., Mistry, B., Kreutz, M., Woodall, P., Moss, T. & Goodacre, T. 2008. The effect of cleft lip and palate, and the timing of lip repair on mother–infant interactions and infant development. *Journal of Child Psychology and Psychiatry,* 49**,** 115-123.

Perrelli, J. G. A., Zambaldi, C. F., Cantilino, A. & Sougey, E. B. 2014. Mother-child bonding assessment tools. *Revista Paulista de Pediatria,* 32**,** 257-265.

Prescott, N. J., Lees, M. M., Winter, R. M. & Malcolm, S. 2000. Identification of susceptibility loci for nonsyndromic cleft lip with or without cleft palate in a two stage genome scan of affected sib-pairs. *Human genetics,* 106**,** 345-350.

Rodrigues, N. & Patterson, J. M. 2007. Impact of severity of a child's chronic condition on the functioning of two-parent families. *Journal of Pediatric Psychology,* 32**,** 417-426.

Sischo, L., Broder, H. L. & Phillips, C. 2015. Coping with cleft: A conceptual framework of caregiver responses to nasoalveolar molding. *The Cleft Palate-Craniofacial Journal,* 52**,** 640-650.

Spinner, M. R. 1978. Maternal-infant bonding. *Canadian Family Physician,* 24**,** 1151.

Stanier, P. & Moore, G. E. 2004. Genetics of cleft lip and palate: syndromic genes contribute to the incidence of non-syndromic clefts. *Human molecular genetics,* 13**,** R73-R81.

Tolarová, M. M. & Cervenka, J. 1998. Classification and birth prevalence of orofacial clefts. *American Journal of Medical Genetics Part A,* 75**,** 126-137.

Van Bussel, J. C., Spitz, B. & Demyttenaere, K. 2010. Three self-report questionnaires of the early mother-to-infant bond: reliability and validity of the Dutch version of the MPAS, PBQ and MIBS. *Arch Womens Ment Health,* 13**,** 373-84.

Wyszynski, D. F., Beaty, T. H. & Maestri, N. E. 1996. Genetics of nonsyndromic oral clefts revisited. *The Cleft palate-craniofacial journal,* 33**,** 406-417.

APPENDIX A
INFORMATION DOCUMENT

Good day,

Introduction: My name is Elzette Nienaber and I am currently doing a Master's Degree in Genetic Counselling at the University of the Witwatersrand. I am conducting a study for research purposes to describe maternal-infant bonding between mothers and their infants/children with a cleft lip, as well as to find out about risk factors which may affect bonding. Research is just the process to learn the answer to a question.

Invitation to participate: You are invited to take part in this research study. If you choose to participate in this study, you will need to answer two questionnaires, which will take approximately 20 minutes to complete.

- The first questionnaire will be basic questions about your age, marital status, residence as well as other details.
- The second questionnaire is called the Postpartum Bonding Questionnaire which will assess the bonding between you and your child.

Risks: There is no risk in participating in this study.
Benefits: By choosing to participate we will be able to determine if there is impaired bonding in mothers with babies with a cleft lip.

Participation is voluntary: You do not have to take part in the study. If you choose to take part, you do not have to answer all the questions. You will not be disadvantaged in any way with regards to your waiting time or treatment and we will ensure that you do not lose your place in the queue if you choose to participate in the study. You also have the right to withdraw from the study at any time and this will not affect your child's management and treatment in any way.

Confidentiality: If you choose to take part in the study, you will remain anonymous, as no names will appear on the questionnaire, which will be kept strictly confidential. On completion of the questionnaire you will be required to put it in the provided, unmarked envelope which you will seal yourself. Only I will have a list to link you to this number, and will not give this information to anyone without your permission.

If the results of the Postpartum Bonding Questionnaire indicate potential impaired bonding between you and your baby I, Elzette Nienaber, will refer to my list to contact you directly. We will refer you to a psychologist at Ububele, a non-profit centre in Johannesburg specialising in mother-infant bonding relationship and support. A genetic counsellor will also be readily available if needed.

The appropriate committees of the University of the Witwatersrand as well as the Head of the Speech Therapy Department and the CEO of the study sites have granted approval for this study.

Should you have any queries regarding this study or about your participation in the study, I or my supervisors can be contacted on the following number:

Principle Investigator	Elzette Nienaber:	011 489 9223
Supervisors	Merlyn Glass	011 489 9335
	Lindsay Lambie	011 489 9336

Contact details of REC administrator and chair

Ms. Zanele Ndlovu	011 717 1252/2700/1234/2656	zanele.ndlovu@wits.ac.za
Mr. Rhulani Mkansi	011 717 1252/2700/1234/2656	rhulani.mkansi@wits.ac.za

NATIONAL HEALTH LABORATORY SERVICE
School of Pathology, University of the Witwatersrand
Division of Human Genetics

APPENDIX B

GENETIC COUNSELLING CLINIC

Hospital Street, Johannesburg, 2001 | PO Box 1038, Johannesburg, 2000
[T]: +27 11 489 9223 [F]: +27 11 489 9226 [E]: human.genetics@nhls.ac.za

Prof A Christianson: +27 11 489 9239 Prof A Krause +27 11 489 9219

INFORMED CONSENT

NR:____

I have read and understood the attached information sheet and agree to take part in the research study entitled "Assessment of maternal-infant bonding between mothers and their babies with a cleft lip", conducted by Elzette Nienaber.

I hereby acknowledge that my participation in this study will be completely voluntary and I am allowed to withdraw at any time. I understand that if the results of the Postpartum Bonding Questionnaire indicate potential impaired bonding between me and my baby, that I will be contacted and referred to a non-profit centre in Johannesburg specialising in mother-infant bonding relationship and support.

I understand that taking part in this study will not provide any direct benefit to my child or change the care and treatment of my child in any way.

Name: _____

Surname: _____

Contact number 1: _____

Contact number 2:_____

Date: _____ Signature: _____

This report is intended solely to record the observations and/or opinion of the writer. It does not constitute a medico-legal report.

APPENDIX C

Nr: ____

DEMOGRAPHIC QUESTIONNAIRE

DATE: _____

1. Mother's age: _____

2. Marital Status: ☐ Single ☐ Married ☐ Separated ☐ Divorced ☐ Widowed ☐ Living together

3. Population group: ☐ Black ☐ Coloured ☐ White ☐ Indian ☐ Other

4. Highest grade completed/level of education: _____

5. Occupation: _____ ☐ Not working

6. SASSA Grant for your child: ☐ Child support grant ☐ No Grant ☐ Care Dependency Grant

7. Household socio economic status

Item			
a) Electricity		h) Television	
b) Motor vehicle		i) Radio	
c) Fridge		j) Video machine/DVD	
d) Microwave		k) MNet	
e) Washing machine		l) DSTV/Satellite	
f) Landline telephone		m) Computer	
g) Cell phone		n) Internet access	

8. Type of residence: ☐ Flat ☐ House ☐ Rent

☐ Own ☐ Shack/Informal settlement

9. Number of children: _____
10. Ages of children: _____
11. Age of child with cleft lip with/without palate: _____ (months)
12. Mode of delivery: ☐ Cesarean section ☐ Normal vaginal delivery
13. Gender of child with CLP: ☐ Male ☐ Female
14. Cleft lip with/without palate: ☐ Cleft lip only ☐ Cleft lip and palate
15. Type of cleft: ☐ Unilateral ☐ Bilateral cleft
16. Cleft repaired: ☐ Yes ☐ No
    a. If YES, baby's age when repaired? _____
17. Did you breastfeed? ☐ Yes ☐ No
18. If not, why? _____
19. Did you receive the diagnosis ☐ Prenatally ☐ Postnatally

APPENDIX D

POSTPARTUM BONDING QUESTIONNAIRE (Brockington et al., 2001).

Please indicate how often the following are true for you.
There are no right or wrong answers:
Choose the answer which best seems right in your recent experience.

NUMBER:

DATE:

		Always	**Very Often**	**Quite Often**	**Some-times**	**Rarely**	**Never**
1.	I feel close to my baby						
2.	I wish the old days when I had no baby would come back						
3	I feel distant from my baby						
4.	I love to cuddle my baby						
5.	I regret having this baby						
6.	The baby does not seem to be mine						
7	The baby winds me up						
8.	My baby irritates me						
9.	I feel happy when my baby smiles or laughs						
10.	I love my baby to bits						
11.	I enjoy playing with my baby						
12.	My baby cries too much						
13.	I feel trapped as a mother						
14.	I feel angry with my baby						
15.	I resent my baby						
16.	My baby is the most beautiful baby in the world						
17.	I wish my baby would sometimes go away						
19.	My baby makes me anxious						
20.	I am afraid of my baby						
21.	My baby annoys me						
22.	I feel confident when changing my baby						
23.	I feel the only solution is for someone else to look after my baby						
25.	My baby is easily comforted						

GAUTENG PROVINCE
HEALTH
REPUBLIC OF SOUTH AFRICA

MEDICAL ADVISORY COMMITTEE
CHRIS HANI BARAGWANATH ACADEMIC HOSPITAL

PERMISSION TO CONDUCT RESEARCH

Date: 27 Feb 2017

TITLE OF PROJECT: Assessment of maternal-infant bonding between mothers and their babies with a cleft lip

UNIVERSITY: Witwatersrand

Principal Investigator: E Nienaber

Department: Human Genetics

Supervisor (If relevant): M Glass

Permission Head Department (where research conducted): Yes

Date of start of proposed study: Feb 2017
Date of completion of data collection: Dec 2019

The Medical Advisory Committee recommends that the said research be conducted at Chris Hani Baragwanath Hospital. The CEO /management of Chris Hani Baragwanath Hospital is accordingly informed and the study is subject to:-

- Permission having been granted by the Human Research Ethics Committee of the University of the Witwatersrand.
- the Hospital will not incur extra costs as a result of the research being conducted on its patients within the hospital
- the MAC will be informed of any serious adverse events as soon as they occur
- permission is granted for the duration of the Ethics Committee approval.

.....
Recommended
(On behalf of the MAC)
Date: 27 February 2017

.....
Approved/Not Approved
Hospital Management
Date: 28/12/17

GAUTENG PROVINCE
HEALTH
REPUBLIC OF SOUTH AFRICA

CHARLOTTE MAXEKE JOHANNESBURG ACADEMIC HOSPITAL

Enquiries:
Mr. Johannes Maepa
Office of the Clinical Director
Tell: (011) 488-3365
Email:johannes.maepa@gauteng.gov.za
04 April 2017

Dear Ms Elzette Neinaber

STUDY TITLE: "Assessment of maternal-infant bonding between mothers and their babies with a cleft lip".

Permission to conduct the above mentioned study is provisionally approved. Your study can only commence once Ethics approval is obtained. Please forward a copy of your ethics clearance as soon as the study is approved by the Ethics committee for the CEO's to give you the final approval to conduct the study.

Supported/~~not supported~~

Dr M.I. Mofokeng
Clinical Director
DATE: 5/4/2017

Approved/not approved

Bogoshi
Ms G. Bogoshi
Chief Executive Officer
Date: 06.04.2017

R14/49 Miss Elzette Nienaber

HUMAN RESEARCH ETHICS COMMITTEE (MEDICAL)

CLEARANCE CERTIFICATE NO. M170258

NAME: (Principal Investigator)	Miss Elzette Nienaber
DEPARTMENT:	Human Genetics National Health Laboratory Service
PROJECT TITLE:	Assessment of Maternal-Infant Bonding between Mothers and their Babies with a Cleft Lip
DATE CONSIDERED:	24/02/2017
DECISION:	Approved unconditionally
CONDITIONS:	
SUPERVISOR:	Ms Merlyn Glass and Dr Lindsay Lambie
APPROVED BY:	Professor P Cleaton-Jones, Chairperson, HREC (Medical)
DATE OF APPROVAL:	03/04/2017

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

DECLARATION OF INVESTIGATORS

To be completed in duplicate and **ONE COPY** returned to the Research Office Secretary in Room 301, Third Floor, Faculty of Health Sciences, Phillip Tobias Building, 29 Princess of Wales Terrace, Parktown, 2193, University of the Witwatersrand. I/we fully understand the conditions under which I am/we are authorized to carry out the above-mentioned research and I/we undertake to ensure compliance with these conditions. Should any departure be contemplated. from the research protocol as approved, I/we undertake to resubmit the application to the Committee. **I agree to submit a yearly progress report**. The date for annual re-certification will be one year after the date of convened meeting where the study was initially reviewed. In this case, the study was initially reviewed in February and will therefore be due in the month of February each year. Unreported changes to the application may invalidate the clearance given by the HREC (Medical).

Principal Investigator Signature

Date 4 April 2017

PLEASE QUOTE THE PROTOCOL NUMBER IN ALL ENQUIRIES

762941:TURNIT_IN_W_REF_AND_CP.docx

ORIGINALITY REPORT

9%	6%	8%	3%
SIMILARITY INDEX	INTERNET SOURCES	PUBLICATIONS	STUDENT PAPERS

PRIMARY SOURCES

1	europepmc.org Internet Source	1%
2	Feeley, Alison B, Eustasius Musenge, John M Pettifor, and Shane A Norris. "Investigation into longitudinal dietary behaviours and household socio-economic indicators and their association with BMI Z-score and fat mass in South African adolescents: the Birth to Twenty (Bt20) cohort", Public Health Nutrition, 2012. Publication	1%
3	journals.lww.com Internet Source	1%
4	Bonnie W.-M. Siu. "Impairment of Mother-Infant Relationship : Validation of the Chinese Version of Postpartum Bonding Questionnaire", The Journal of Nervous & Mental Disease, 03/2010 Publication	1%
5	repub.eur.nl Internet Source	1%

Sehgal, Virendra Srivastava, Govind Agga.

6	"Atopic dermatitis: A cross-sectional (descriptive) study of 100 cases.(E-IJD Original Article)(Clini", Indian Journal of Dermatology, Sept-Oct 2015 Issue Publication	1%
7	Nicholas Spoerke. "Effects of Ethanol Intoxication and Gender on Blood Coagulation :", The Journal of Trauma Injury Infection and Critical Care, 05/2010 Publication	1%
8	www.sajch.org.za Internet Source	<1%
9	thieme-connect.com Internet Source	<1%
10	Submitted to Grand Canyon University Student Paper	<1%
11	www.ajol.info Internet Source	<1%
12	Garcia-Esteve, Lluïsa, Anna Torres, Gracia Lasheras, Bruma Palacios-Hernández, Borja Farré-Sender, Susana Subirà, Manuel Valdés, and Ian Fraser Brockington. "Assessment of psychometric properties of the Postpartum Bonding Questionnaire (PBQ) in Spanish mothers", Archives of Women s Mental Health, 2015. Publication	<1%

13	www.jove.com Internet Source	<1%
14	Submitted to University of Bristol Student Paper	<1%
15	P. Stanier. "Genetics of cleft lip and palate: syndromic genes contribute to the incidence of non-syndromic clefts", Human Molecular Genetics, 01/13/2004 Publication	<1%
16	Natalia Millacura, Rosa Pardo, Lucia Cifuentes, José Suazo. "Effects of folic acid fortification on orofacial clefts prevalence: a meta-analysis", Public Health Nutrition, 2017 Publication	<1%
17	Karsten, Agneta, Margareta Larson, Ola Larson, and (for the Stockholm Cleft Palate Tea. "Length of the cleft in relation to the incidence of hypodontia of the second premolar and to inheritance of cleft lip and palate in children with isolated cleft palate", Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery, 2005. Publication	<1%
18	aje.oxfordjournals.org Internet Source	<1%

19	Beaty, T.. "A Case-Control Study of Nonsyndromic Oral Clefts in Maryland", Annals of Epidemiology, 200108 Publication	<1%

Exclude quotes	On	Exclude matches	< 3 words
Exclude bibliography	On		

Instructions for Authors

TYPES OF PAPERS

- Original Contributions / Research Articles should be arranged into sections conforming to standard scientific reporting style, i.e. under the following headings:
  Abstract:
  Should not exceed 150–250 words and be structured as follows: Purpose, Methods, Results, Conclusions
  Keywords: Not more than five, separated by semicolons
  - Introduction:
  A brief outline of the background literature leading to the objective(s) of the study
  - Materials and Methods:
  Describe the basic study design. State the setting (e.g., primary care, referral center). Explain selection of study subjects and state the system of diagnostic criteria used. Describe any interventions and include their duration and method of administration. Indicate the main outcome measure(s). Specify the dates in which data were collected (month/year to month/year).
  - Results:
  Include the key findings. Give specific data and their statistical significance, if possible. Subset Ns should accompany percentages if the total N is ‹100.
  - Discussion and Conclusions:
  Discuss your findings critically in comparison to existing literature and considering your methodological and other limitations.
  Conclusions should highlight the potential meaning for the field given the limitations.
  The main text (i.e. without abstract, references, figures, tables, or supplementary material) should not exceed 3000 words.
- Reviews should be comprehensive, fully referenced expositions of subjects of general interest, including background information and detailed critical analyses of current work in the field and its significance. They should be designed to serve as source materials.
  Meta-analyses or systematic reviews according to the PRISMA style are preferred.
  Reviews are not meant to be encyclopedic and should not exceed 4000 words. Reviews may contain figures and tables.
- Short Communications should be prepared as described above except for the following:
  The average length of Short Communications should not exceed 1500 words and may include a maximum of two figures or tables and up to 12 references. The summary should not exceed 80 words.
- Letters to the Editor
  Letters to the Editor should be a maximum of 750 words and may include one table or figure and up to five references.

MANUSCRIPT SUBMISSION

Manuscript Submission

Submission of a manuscript implies: that the work described has not been published before; that it is not under consideration for publication anywhere else; that its publication has been approved by all co-authors, if any, as well as by the responsible authorities – tacitly or explicitly – at the institute where the work has been carried out. The publisher will not be held legally responsible should there be any claims for compensation.

Permissions

Authors wishing to include figures, tables, or text passages that have already been published elsewhere are required to obtain permission from the copyright owner(s) for both the print and online format and to include evidence that such permission has

been granted when submitting their papers. Any material received without such evidence will be assumed to originate from the authors.

Online Submission

Please follow the hyperlink “Submit online” on the right and upload all of your manuscript files following the instructions given on the screen.

Reviewer Suggestion

Authors will be kindly asked to suggest up to 5 potential reviewers for their papers. These recommendations will be of help to ensure the journal’s high scientific level and will support a quick review process and thus shorten the time from manuscript submission to publication.
Please note that only reviewer suggestions from institutions of international reputation other than the institution of the corresponding author will be taken into consideration and you should not have published common articles with the suggested reviewers previously.

Please note that the journal does not offer pre-evaluation. Therefore please directly submit your manuscript to EditorialManager at the below link. The Editorial Office will then contact you.

- EditorialManager

TITLE PAGE

Title Page

The title page should include:

- The name(s) of the author(s)
- A concise and informative title
- The affiliation(s) and address(es) of the author(s)
- The e-mail address, and telephone number(s) of the corresponding author
- If available, the 16-digit ORCID of the author(s)

Abstract

Please provide a structured abstract of 150 to 250 words which should be divided into the following sections:

- Purpose (stating the main purposes and research question)
- Methods
- Results
- Conclusions

Keywords

Please provide 4 to 6 keywords which can be used for indexing purposes.

Important note:

Please ensure your authorship is correct, check spelling of authors' names, line up, etc.
No changes can be made once copyright has been transferred to us.

TEXT

Text Formatting

Manuscripts should be submitted in Word.

- Use a normal, plain font (e.g., 10-point Times Roman) for text.
- Use italics for emphasis.

- Use the automatic page numbering function to number the pages.
- Do not use field functions.
- Use tab stops or other commands for indents, not the space bar.
- Use the table function, not spreadsheets, to make tables.
- Use the equation editor or MathType for equations.
- Save your file in docx format (Word 2007 or higher) or doc format (older Word versions).

Manuscripts with mathematical content can also be submitted in LaTeX.

- LaTeX macro package (zip, 182 kB)

Headings

Please use no more than three levels of displayed headings.

Abbreviations

Abbreviations should be defined at first mention and used consistently thereafter.

Footnotes

Footnotes can be used to give additional information, which may include the citation of a reference included in the reference list. They should not consist solely of a reference citation, and they should never include the bibliographic details of a reference. They should also not contain any figures or tables.
Footnotes to the text are numbered consecutively; those to tables should be indicated by superscript lower-case letters (or asterisks for significance values and other statistical data). Footnotes to the title or the authors of the article are not given reference symbols.
Always use footnotes instead of endnotes.

Acknowledgments

Acknowledgments of people, grants, funds, etc. should be placed in a separate section on the title page. The names of funding organizations should be written in full.

REFERENCES

Citation

Cite references in the text by name and year in parentheses. Some examples:

- Negotiation research spans many disciplines (Thompson 1990).
- This result was later contradicted by Becker and Seligman (1996).
- This effect has been widely studied (Abbott 1991; Barakat et al. 1995a, b; Kelso and Smith 1998; Medvec et al. 1999, 2000).

Reference list

The list of references should only include works that are cited in the text and that have been published or accepted for publication. Personal communications and unpublished works should only be mentioned in the text. Do not use footnotes or endnotes as a substitute for a reference list.
Reference list entries should be alphabetized by the last names of the first author of each work. Order multi-author publications of the same first author alphabetically with respect to second, third, etc. author. Publications of exactly the same author(s) must be ordered chronologically.

- Journal article

Gamelin FX, Baquet G, Berthoin S, Thevenet D, Nourry C, Nottin S, Bosquet L (2009) Effect of high intensity intermittent training on heart rate variability in prepubescent children. Eur J Appl Physiol 105:731-738. doi: 10.1007/s00421-008-0955-8

Ideally, the names of all authors should be provided, but the usage of "et al" in long author lists will also be accepted:

Smith J, Jones M Jr, Houghton L et al (1999) Future of health insurance. N Engl J Med 965:325–329

- Article by DOI

  Slifka MK, Whitton JL (2000) Clinical implications of dysregulated cytokine production. J Mol Med. doi:10.1007/s001090000086

- Book

  South J, Blass B (2001) The future of modern genomics. Blackwell, London

- Book chapter

  Brown B, Aaron M (2001) The politics of nature. In: Smith J (ed) The rise of modern genomics, 3rd edn. Wiley, New York, pp 230-257

- Online document

  Cartwright J (2007) Big stars have weather too. IOP Publishing PhysicsWeb. http://physicsweb.org/articles/news/11/6/16/1. Accessed 26 June 2007

- Dissertation

  Trent JW (1975) Experimental acute renal failure. Dissertation, University of California

Always use the standard abbreviation of a journal's name according to the ISSN List of Title Word Abbreviations, see

- ISSN LTWA

If you are unsure, please use the full journal title.
For authors using EndNote, Springer provides an output style that supports the formatting of in-text citations and reference list.

- EndNote style (zip, 2 kB)

TABLES

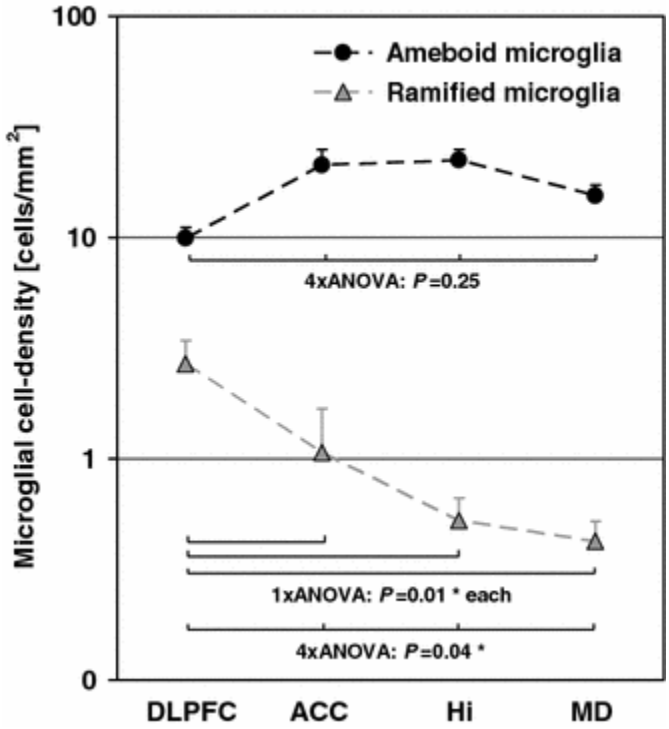
- All tables are to be numbered using Arabic numerals.
- Tables should always be cited in text in consecutive numerical order.
- For each table, please supply a table caption (title) explaining the components of the table.
- Identify any previously published material by giving the original source in the form of a reference at the end of the table caption.
- Footnotes to tables should be indicated by superscript lower-case letters (or asterisks for significance values and other statistical data) and included beneath the table body.

ARTWORK AND ILLUSTRATIONS GUIDELINES

Electronic Figure Submission

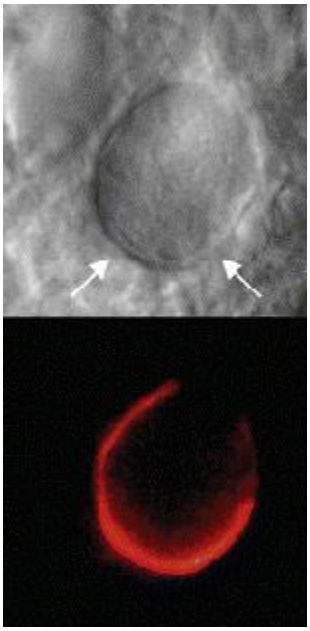
- Supply all figures electronically.
- Indicate what graphics program was used to create the artwork.
- For vector graphics, the preferred format is EPS; for halftones, please use TIFF format. MSOffice files are also acceptable.
- Vector graphics containing fonts must have the fonts embedded in the files.
- Name your figure files with "Fig" and the figure number, e.g., Fig1.eps.

Line Art



- Definition: Black and white graphic with no shading.
- Do not use faint lines and/or lettering and check that all lines and lettering within the figures are legible at final size.
- All lines should be at least 0.1 mm (0.3 pt) wide.
- Scanned line drawings and line drawings in bitmap format should have a minimum resolution of 1200 dpi.
- Vector graphics containing fonts must have the fonts embedded in the files.

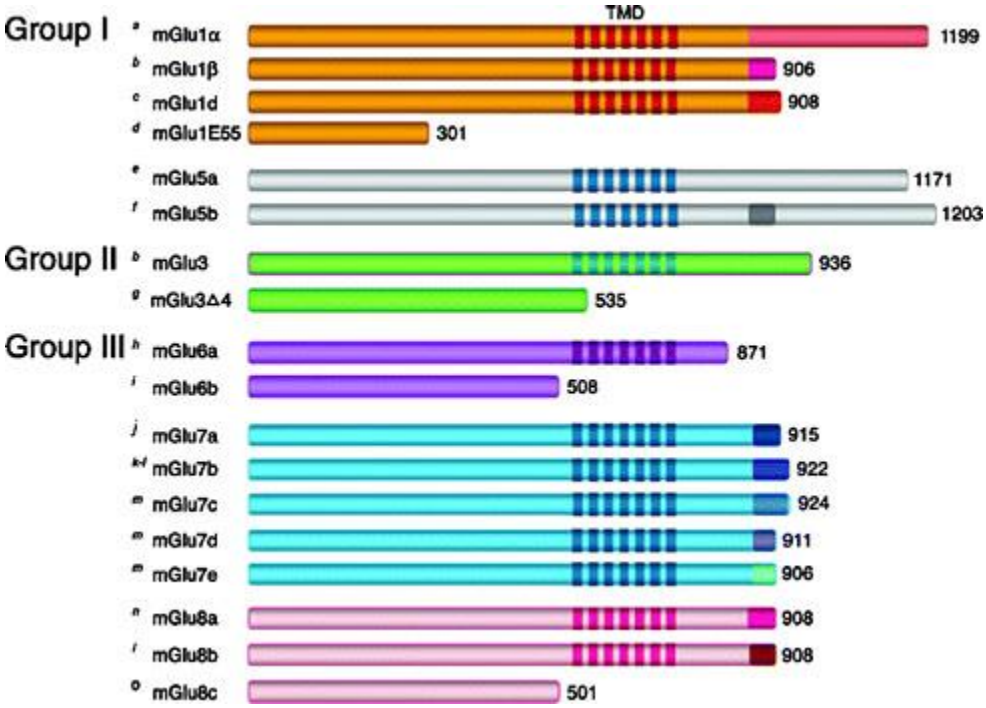
Halftone Art



- Definition: Photographs, drawings, or paintings with fine shading, etc.

- If any magnification is used in the photographs, indicate this by using scale bars within the figures themselves.
- Halftones should have a minimum resolution of 300 dpi.

Combination Art



- Definition: a combination of halftone and line art, e.g., halftones containing line drawing, extensive lettering, color diagrams, etc.
- Combination artwork should have a minimum resolution of 600 dpi.

Color Art

- Color art is free of charge for online publication.
- If black and white will be shown in the print version, make sure that the main information will still be visible. Many colors are not distinguishable from one another when converted to black and white. A simple way to check this is to make a xerographic copy to see if the necessary distinctions between the different colors are still apparent.
- If the figures will be printed in black and white, do not refer to color in the captions.
- Color illustrations should be submitted as RGB (8 bits per channel).

Figure Lettering

- To add lettering, it is best to use Helvetica or Arial (sans serif fonts).
- Keep lettering consistently sized throughout your final-sized artwork, usually about 2–3 mm (8–12 pt).
- Variance of type size within an illustration should be minimal, e.g., do not use 8-pt type on an axis and 20-pt type for the axis label.
- Avoid effects such as shading, outline letters, etc.
- Do not include titles or captions within your illustrations.

Figure Numbering

- All figures are to be numbered using Arabic numerals.
- Figures should always be cited in text in consecutive numerical order.
- Figure parts should be denoted by lowercase letters (a, b, c, etc.).

- If an appendix appears in your article and it contains one or more figures, continue the consecutive numbering of the main text. Do not number the appendix figures,
  "A1, A2, A3, etc." Figures in online appendices (Electronic Supplementary Material) should, however, be numbered separately.

Figure Captions

- Each figure should have a concise caption describing accurately what the figure depicts. Include the captions in the text file of the manuscript, not in the figure file.
- Figure captions begin with the term Fig. in bold type, followed by the figure number, also in bold type.
- No punctuation is to be included after the number, nor is any punctuation to be placed at the end of the caption.
- Identify all elements found in the figure in the figure caption; and use boxes, circles, etc., as coordinate points in graphs.
- Identify previously published material by giving the original source in the form of a reference citation at the end of the figure caption.

Figure Placement and Size

- Figures should be submitted separately from the text, if possible.
- When preparing your figures, size figures to fit in the column width.
- For most journals the figures should be 39 mm, 84 mm, 129 mm, or 174 mm wide and not higher than 234 mm.
- For books and book-sized journals, the figures should be 80 mm or 122 mm wide and not higher than 198 mm.

Permissions

If you include figures that have already been published elsewhere, you must obtain permission from the copyright owner(s) for both the print and online format. Please be aware that some publishers do not grant electronic rights for free and that Springer will not be able to refund any costs that may have occurred to receive these permissions. In such cases, material from other sources should be used.

Accessibility

In order to give people of all abilities and disabilities access to the content of your figures, please make sure that

- All figures have descriptive captions (blind users could then use a text-to-speech software or a text-to-Braille hardware)
- Patterns are used instead of or in addition to colors for conveying information (colorblind users would then be able to distinguish the visual elements)
- Any figure lettering has a contrast ratio of at least 4.5:1

ELECTRONIC SUPPLEMENTARY MATERIAL

Springer accepts electronic multimedia files (animations, movies, audio, etc.) and other supplementary files to be published online along with an article or a book chapter. This feature can add dimension to the author's article, as certain information cannot be printed or is more convenient in electronic form.
Before submitting research datasets as electronic supplementary material, authors should read the journal's Research data policy. We encourage research data to be archived in data repositories wherever possible.

Submission

- Supply all supplementary material in standard file formats.
- Please include in each file the following information: article title, journal name, author names; affiliation and e-mail address of the corresponding author.
- To accommodate user downloads, please keep in mind that larger-sized files may require very long download times and that some users may experience other problems during downloading.

Audio, Video, and Animations

- Aspect ratio: 16:9 or 4:3
- Maximum file size: 25 GB
- Minimum video duration: 1 sec
- Supported file formats: avi, wmv, mp4, mov, m2p, mp2, mpg, mpeg, flv, mxf, mts, m4v, 3gp

Text and Presentations

- Submit your material in PDF format; .doc or .ppt files are not suitable for long-term viability.
- A collection of figures may also be combined in a PDF file.

Spreadsheets

- Spreadsheets should be submitted as .csv or .xlsx files (MS Excel).

Specialized Formats

- Specialized format such as .pdb (chemical), .wrl (VRML), .nb (Mathematica notebook), and .tex can also be supplied.

Collecting Multiple Files

- It is possible to collect multiple files in a .zip or .gz file.

Numbering

- If supplying any supplementary material, the text must make specific mention of the material as a citation, similar to that of figures and tables.
- Refer to the supplementary files as “Online Resource”, e.g., "... as shown in the animation (Online Resource 3)", “... additional data are given in Online Resource 4”.
- Name the files consecutively, e.g. “ESM_3.mpg”, “ESM_4.pdf”.

Captions

- For each supplementary material, please supply a concise caption describing the content of the file.

Processing of supplementary files

- Electronic supplementary material will be published as received from the author without any conversion, editing, or reformatting.

Accessibility

In order to give people of all abilities and disabilities access to the content of your supplementary files, please make sure that

- The manuscript contains a descriptive caption for each supplementary material
- Video files do not contain anything that flashes more than three times per second (so that users prone to seizures caused by such effects are not put at risk)

ETHICAL RESPONSIBILITIES OF AUTHORS

This journal is committed to upholding the integrity of the scientific record. As a member of the Committee on Publication Ethics (COPE) the journal will follow the COPE guidelines on how to deal with potential acts of misconduct.
Authors should refrain from misrepresenting research results which could damage the trust in the journal, the professionalism of scientific authorship, and ultimately the entire scientific endeavour. Maintaining integrity of the research and its presentation can be achieved by following the rules of good scientific practice, which include:

- The manuscript has not been submitted to more than one journal for simultaneous consideration.
- The manuscript has not been published previously (partly or in full), unless the new work concerns an expansion of previous work (please provide transparency on the re-use of material to avoid the hint of text-recycling (“self-plagiarism”)).

- A single study is not split up into several parts to increase the quantity of submissions and submitted to various journals or to one journal over time (e.g. “salami-publishing”).
- No data have been fabricated or manipulated (including images) to support your conclusions
- No data, text, or theories by others are presented as if they were the author’s own (“plagiarism”). Proper acknowledgements to other works must be given (this includes material that is closely copied (near verbatim), summarized and/or paraphrased), quotation marks are used for verbatim copying of material, and permissions are secured for material that is copyrighted.

  Important note: the journal may use software to screen for plagiarism.
- Consent to submit has been received explicitly from all co-authors, as well as from the responsible authorities - tacitly or explicitly - at the institute/organization where the work has been carried out, **before** the work is submitted.
- Authors whose names appear on the submission have contributed sufficiently to the scientific work and therefore share collective responsibility and accountability for the results.
- Authors are strongly advised to ensure the correct author group, corresponding author, and order of authors at submission. Changes of authorship or in the order of authors are **not** accepted **after** acceptance of a manuscript.
- Adding and/or deleting authors **at revision stage** may be justifiably warranted. A letter must accompany the revised manuscript to explain the role of the added and/or deleted author(s). Further documentation may be required to support your request.
- Requests for addition or removal of authors as a result of authorship disputes after acceptance are honored after formal notification by the institute or independent body and/or when there is agreement between all authors.
- Upon request authors should be prepared to send relevant documentation or data in order to verify the validity of the results. This could be in the form of raw data, samples, records, etc. Sensitive information in the form of confidential proprietary data is excluded.

If there is a suspicion of misconduct, the journal will carry out an investigation following the COPE guidelines. If, after investigation, the allegation seems to raise valid concerns, the accused author will be contacted and given an opportunity to address the issue. If misconduct has been established beyond reasonable doubt, this may result in the Editor-in-Chief’s implementation of the following measures, including, but not limited to:

- If the article is still under consideration, it may be rejected and returned to the author.
- If the article has already been published online, depending on the nature and severity of the infraction, either an erratum will be placed with the article or in severe cases complete retraction of the article will occur. The reason must be given in the published erratum or retraction note. Please note that retraction means that the paper is **maintained on the platform**, watermarked "retracted" and explanation for the retraction is provided in a note linked to the watermarked article.
- The author’s institution may be informed.

COMPLIANCE WITH ETHICAL STANDARDS

To ensure objectivity and transparency in research and to ensure that accepted principles of ethical and professional conduct have been followed, authors should include information regarding sources of funding, potential conflicts of interest (financial or non-financial), informed consent if the research involved human participants, and a statement on welfare of animals if the research involved animals.

Authors should include the following statements (if applicable) in a separate section entitled “Compliance with Ethical Standards” when submitting a paper:

- Disclosure of potential conflicts of interest
- Research involving Human Participants and/or Animals
- Informed consent

Please note that standards could vary slightly per journal dependent on their peer review policies (i.e. single or double blind peer review) as well as per journal subject discipline. Before submitting your article check the instructions following this section carefully.

The corresponding author should be prepared to collect documentation of compliance with ethical standards and send if requested during peer review or after publication.
The Editors reserve the right to reject manuscripts that do not comply with the above-mentioned guidelines. The author will be held responsible for false statements or failure to fulfill the above-mentioned guidelines.

DISCLOSURE OF POTENTIAL CONFLICTS OF INTEREST

Authors must disclose all relationships or interests that could influence or bias the work. Although an author may not feel there are conflicts, disclosure of relationships and interests affords a more transparent process, leading to an accurate and objective assessment of the work. Awareness of real or perceived conflicts of interests is a perspective to which the readers are entitled and is not meant to imply that a financial relationship with an organization that sponsored the research or compensation for consultancy work is inappropriate. Examples of potential conflicts of interests **that are directly or indirectly related to the research** may include but are not limited to the following:

- Research grants from funding agencies (please give the research funder and the grant number)
- Honoraria for speaking at symposia
- Financial support for attending symposia
- Financial support for educational programs
- Employment or consultation
- Support from a project sponsor
- Position on advisory board or board of directors or other type of management relationships
- Multiple affiliations
- Financial relationships, for example equity ownership or investment interest
- Intellectual property rights (e.g. patents, copyrights and royalties from such rights)
- Holdings of spouse and/or children that may have financial interest in the work

In addition, interests that go beyond financial interests and compensation (non-financial interests) that may be important to readers should be disclosed. These may include but are not limited to personal relationships or competing interests directly or indirectly tied to this research, or professional interests or personal beliefs that may influence your research.
The corresponding author collects the conflict of interest disclosure forms from all authors. (**Please note that each author should complete a disclosure form.**) Examples of forms can be found

- here:

The corresponding author will include a summary statement **on the title page that is separate from their manuscript,** that reflects what is recorded in the potential conflict of interest disclosure form(s).
See below examples of disclosures:
Funding: This study was funded by X (grant number X).
Conflict of Interest: Author A has received research grants from Company A. Author B has received a speaker honorarium from Company X and owns stock in Company Y. Author C is a member of committee Z.
If no conflict exists, the authors should state:
Conflict of Interest: Author A, Author B, and Author C declare that they have no conflict of interest.

RESEARCH INVOLVING HUMAN PARTICIPANTS AND/OR ANIMALS

1) Statement of human rights

When reporting studies that involve human participants, authors should include a statement that the studies have been approved by the appropriate institutional and/or national research ethics committee and have been performed in accordance with the ethical standards as laid down in the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards.

If doubt exists whether the research was conducted in accordance with the 1964 Helsinki Declaration or comparable standards, the authors must explain the reasons for their approach, and demonstrate that the independent ethics committee or institutional review board explicitly approved the doubtful aspects of the study.
The following statements should be included in the text before the References section:
Ethical approval: "All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards."
For retrospective studies, please add the following sentence:
"For this type of study formal consent is not required."

2) Statement on the welfare of animals

The welfare of animals used for research must be respected. When reporting experiments on animals, authors should indicate whether the international, national, and/or institutional guidelines for the care and use of animals have been followed, and that the studies have been approved by a research ethics committee at the institution or practice at which the studies were conducted (where such a committee exists).
For studies with animals, the following statement should be included in the text before the References section:
Ethical approval: "All applicable international, national, and/or institutional guidelines for the care and use of animals were followed."
If applicable (where such a committee exists): "All procedures performed in studies involving animals were in accordance with the ethical standards of the institution or practice at which the studies were conducted."
If articles do not contain studies with human participants or animals by any of the authors, please select one of the following statements:
"This article does not contain any studies with human participants performed by any of the authors."
"This article does not contain any studies with animals performed by any of the authors."
"This article does not contain any studies with human participants or animals performed by any of the authors."

INFORMED CONSENT

All individuals have individual rights that are not to be infringed. Individual participants in studies have, for example, the right to decide what happens to the (identifiable) personal data gathered, to what they have said during a study or an interview, as well as to any photograph that was taken. Hence it is important that all participants gave their informed consent in writing prior to inclusion in the study. Identifying details (names, dates of birth, identity numbers and other information) of the participants that were studied should not be published in written descriptions, photographs, and genetic profiles unless the information is essential for scientific purposes and the participant (or parent or guardian if the participant is incapable) gave written informed consent for publication. Complete anonymity is difficult to achieve in some cases, and informed consent should be obtained if there is any doubt. For example, masking the eye region in photographs of participants is inadequate protection of anonymity. If identifying characteristics are altered to protect anonymity, such as in genetic profiles, authors should provide assurance that alterations do not distort scientific meaning.
The following statement should be included:
Informed consent: "Informed consent was obtained from all individual participants included in the study."
If identifying information about participants is available in the article, the following statement should be included:
"Additional informed consent was obtained from all individual participants for whom identifying information is included in this article."

RESEARCH DATA POLICY

The journal encourages authors, where possible and applicable, to deposit data that support the findings of their research in a public repository. Authors and editors who do not have a preferred repository should consult Springer Nature's list of repositories and research data policy.

- List of Repositories
- Research Data Policy

General repositories - for all types of research data - such as figshare and Dryad may also be used.
Datasets that are assigned digital object identifiers (DOIs) by a data repository may be cited in the reference list. Data citations should include the minimum information recommended by DataCite: authors, title, publisher (repository name), identifier.

- DataCite

Springer Nature provides a research data policy support service for authors and editors, which can be contacted at **researchdata@springernature.com**.

This service provides advice on research data policy compliance and on finding research data repositories. It is independent of journal, book and conference proceedings editorial offices and does not advise on specific manuscripts.

- Helpdesk

AFTER ACCEPTANCE

Upon acceptance of your article you will receive a link to the special Author Query Application at Springer's web page where you can sign the Copyright Transfer Statement online and indicate whether you wish to order OpenChoice and offprints.
Once the Author Query Application has been completed, your article will be processed and you will receive the proofs.

Copyright transfer

Authors will be asked to transfer copyright of the article to the Publisher (or grant the Publisher exclusive publication and dissemination rights). This will ensure the widest possible protection and dissemination of information under copyright laws.

- Creative Commons Attribution-NonCommercial 4.0 International License

Offprints

Offprints can be ordered by the corresponding author.

Color illustrations

Publication of color illustrations is free of charge.

Proof reading

The purpose of the proof is to check for typesetting or conversion errors and the completeness and accuracy of the text, tables and figures. Substantial changes in content, e.g., new results, corrected values, title and authorship, are not allowed without the approval of the Editor.
After online publication, further changes can only be made in the form of an Erratum, which will be hyperlinked to the article.

Online First

The article will be published online after receipt of the corrected proofs. This is the official first publication citable with the DOI. After release of the printed version, the paper can also be cited by issue and page numbers.

OPEN CHOICE

In addition to the normal publication process (whereby an article is submitted to the journal and access to that article is granted to customers who have purchased a subscription), Springer provides an alternative publishing option: Springer Open Choice. A Springer Open Choice article receives all the benefits of a regular subscription-based article, but in addition is made available publicly through Springer's online platform SpringerLink.

- Open Choice

Copyright and license term – CC BY

Open Choice articles do not require transfer of copyright as the copyright remains with the author. In opting for open access, the author(s) agree to publish the article under the Creative Commons Attribution License.

- Find more about the license agreement

ENGLISH LANGUAGE EDITING

For editors and reviewers to accurately assess the work presented in your manuscript you need to ensure the English language is of sufficient quality to be understood. If you need help with writing in English you should consider:

- Asking a colleague who is a native English speaker to review your manuscript for clarity.
- Visiting the English language tutorial which covers the common mistakes when writing in English.
- Using a professional language editing service where editors will improve the English to ensure that your meaning is clear and identify problems that require your review. Two such services are provided by our affiliates Nature Research Editing Service and American Journal Experts.

- English language tutorial
- Nature Research Editing Service
- American Journal Experts

Please note that the use of a language editing service is not a requirement for publication in this journal and does not imply or guarantee that the article will be selected for peer review or accepted.
If your manuscript is accepted it will be checked by our copyeditors for spelling and formal style before publication.