

https://dspace.org/

Assessment of services rendered by a non-governmental organization on the management of cleft lip and palate in South Africa

Muthavhine, Mashudu

2021
https://hdl.handle.net/10539/33579

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

Assessment of services rendered by a non-governmental organization on the management of cleft lip and palate in South Africa

Dr Mashudu Muthavhine
Student Number: 2294141

Supervisor
Professor Phumzile Hlongwa, BOH; BDS; MDent (Ortho); PhD

Co-supervisor
Professor Mbulaheni Simon Nemutandani, BchD; MSc; MChD; PhD
Dr Helen Malherbe, BSc (Hons); MSc; PhD

A Research Report submitted for the partial fulfilment of the degree MSc (Dent), in the Department of Orthodontics, School of Oral Health Sciences, Faculty of Health Sciences, University of the Witwatersrand, Johannesburg.

2021

Declaration

I, Mashudu Lucky Muthavhine, declare that this research project "Assessment of services rendered by a non-governmental organization on the management of cleft lip and palate in South Africa" is my own work. It is being submitted for the Degree of Master of Science in Dentistry in the Department of Orthodontics at the University of Witwatersrand, Johannesburg. It has not been submitted before for any degree or any examination in any other University.



Date: 03 September 2021

Dedication

To God Almighty

For you never left nor forsaken me throughout my journey of academic development.

To my loving wife, Rabelani and children, Tshedza, Andani and Mashudu.

For all your support, patience, and sacrifices.

To my late parents, Ratshilumela, and Gladys.

For believing in me.

Abstract

Objectives: To assess the services of a single NGO, Smile Foundation (SF), operating in South Africa in managing OFC.

Methodology: A case study to obtain the organisation's background and care management approach was obtained through a structured interview with the Smile Foundation Chief Executive Officer and Operations Executive Director. Furthermore, an audit of the clinical records of OFC cases facilitated by Smile Foundation over a period of five years (2015-2019) was conducted.

Results: Smile Foundation facilitated OFC services in six provinces in 11 centres from 2015-2019. Support provided included staff renumeration costs for psychological, surgical, theatre staff and other relevant specialist health care professionals, patient transport and accommodation, outreach programmes, medical staff training, and purchase of medical equipment. Records for 515 patients undergoing a total of 564 surgical procedures were analysed. The majority of procedures were performed in the Western Cape Province over the five-year period.

Conclusions: Smile Foundation augmented 30% of OFC state-based procedures annually over a five-year period. To ensure sustainability of these services in the future, greater government commitment to OFC services is recommended.

Acknowledgements

- I am indebted to my primary supervisor, Professor Phumzile Hlongwa, for the role she has played from the conceptualization of this research project to its final finishing. Words cannot express enough the tremendous support she gave me, and unreservedly carrying me through my academic development.

- I thank and appreciate Dr Helen Malherbe, my co-supervisor. For your passion and dedication in guiding me in every aspect of my research.

- I also thank Professor Mbulaheni Simon Nemutandani, for agreeing to co-supervise this Research Report.

- I wish to gratefully acknowledge Mr Hedley Lewis and Moira Gerszt from Smile Foundation for allowing me access to the records for this project.

- Thank you Dr Vuyo Bomvana and Dr Mandla Nyakale for your guidance and moral support during my research project.

- I thank my family: - My God sent wife, Rabelani, her love, and support were the source of my strength through this journey. My two daughters, Tshedza and Andani, and son, Mashudu Jr. Your love and playfulness kept me sane during hard and challenging times.

Table of Contents

Declaration ii
Dedication iii
Abstract iv
Acknowledgements v
Table of Contents vi
List of figures viii
List of tables ix
Abbreviations x
CHAPTER 1 1
Introduction and Literature Review 1
1.1 Background 1
1.2 Development of OFC 2
1.3 Aetiology of OFC 2
1.4 Epidemiology of OFC 3
1.5 OFC laterality and gender differences 4
1.6 Individuals and parents' perception of OFC 4
1.7 Management of OFC 4
1.8 The multidisciplinary team 6
1.8.1 The Dentist 7
1.8.2 The ear, nose, and throat specialist (ENT) 7
1.8.3 The Medical Geneticist and/or Genetic counsellor 7
1.8.4 The Maxillofacial surgeon 8
1.8.5 The Orthodontist 8
1.8.6 The Paediatrician 8
1.8.7 The Plastic surgeon 8
1.8.8 The Psychologist and/or social worker 9
1.8.9 The Prosthodontist 9
1.8.10 The professional nurse 9
1.8.11 The Speech therapist 10
1.9 Non-profit organisations involved in OFC treatment 10

1.10 Role of NGOs in OFC treatment support 15
1.11 Study Rationale 16
1.12 Aim of the study 16
1.13 Research objectives 16
CHAPTER 2 17
Methodology 17
2.1 Study design 17
2.2 Study setting 17
2.3 Study population and sampling 17
2.4 Data collection 17
2.5 Data management and analysis 18
2.6 Ethical considerations 19
CHAPTER 3 20
Results 20
3.1 Qualitative results 20
3.2 Quantitative results 23
CHAPTER 4 31
Discussion and Conclusions 31
Study limitations 36
Conclusions 37
Recommendations 37
References 39
Appendix 1 - Data Collection Questionnaire 45
Appendix 2 - Participants Information Sheet 50
Appendix 3 - Informed Consent Form 52
Appendix 4 - Orofacial Clinical information 53
Appendix 5 – Ethics Clearance Certificate 55
Appendix 6 – Permission letter from Smile Foundation 56
Appendix 7 - Permission from Smile Foundation 57
Appendix 8 – Turn-it in Report 58

List of figures

Figure 1: Clinical appearance of OFC 1
Figure 2: Smile Foundation organisational structure 21
Figure 3: Age distribution for the total sample Gender 24
Figure 4: Distribution of surgical procedures per year over the study period 25
Figure 5: Distribution of type of surgical procedure for the total sample 26
Figure 6: Distribution of surgical procedure per province 28
Figure 7: Procedures per Centre per year 30

List of tables

Table 1: Globally recommended OFC Diagnosis age and CLP treatment 6
Table 2: Summary of key OFC NGOs and support they provide 11
Table 3: Descriptive statistics – Age of sample according to gender 24
Table 4: Provincial distribution of total OFC procedures 27
Table 5: Descriptive statistics of procedures performed in each centre 29

Abbreviations

ACPA	American Cleft Palate-Craniofacial Association
AFS	Alliance for Smiles
APRASA	Association of Plastic, Reconstruction and Aesthetic Surgery of South Africa
BCLP	Bilateral cleft lip and palate
CEO	Chief executive officer
CL	Cleft lip
CP	Cleft palate
CLP	Cleft lip and palate
DOH	Department of health (DOH)
ENT	Ear, nose, and throat
IRF6	Interferon regulatory factor-6
LMICS	Low- and middle-income countries
MDT	Multidisciplinary team
NGO	Non-governmental organisation
NPO	Non-profit organisation
OED	Operations executive director
OS	Operation smile
OFC	Orofacial clefts
PI	Principal investigator
PVRL1	Poliovirus receptor-like-1

SF	Smile foundation
TBX22	t-box transcription factor-2
TF	Transforming faces
UCL	Unilateral cleft lip
UCLP	Unilateral cleft lip and palate
USA	United States of America

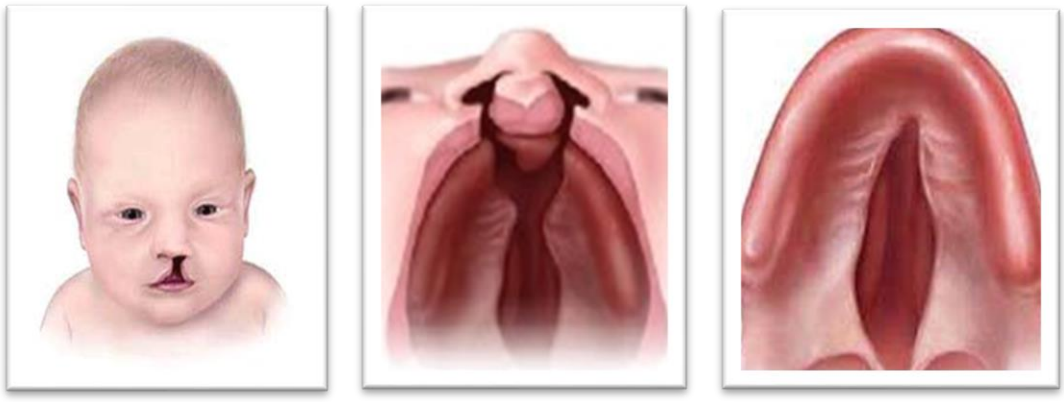
CHAPTER 1

Introduction and Literature Review

1.1 Background

Cleft lip (CL), cleft palate (CP) and cleft lip and palate (CLP) are orofacial defects referred to as orofacial clefts (OFC) that occur when the tissues that make up the lip and palate do not join completely before birth [1]. CL and CLP can be diagnosed during pregnancy by a routine ultrasound or after the baby is born. While CP is mostly diagnosed after birth, certain types of cleft palate (submucous cleft palate and bifid uvula), may be missed or misdiagnosed during pregnancy and at birth during the top to toe examination of new-born and may only be diagnosed later in life [2].

The clinical appearance of OFC anomalies may involve the lip, the palate, or both. When they occur on one side of the face (either left or right lip), they are referred to as unilateral cleft lip (UCL), and unilateral cleft lip and palate (UCLP) when they involve both lip and palate. They may occur on both sides of the face and referred to as bilateral cleft lip and palate (BCLP) [3]. Clefts that occur on the middle of the face (midline cleft) are rare [1]. Figure 1 below shows the clinical appearance of some OFC.



A) B) C)

Figure 1: Clinical appearance of OFC

A) Left unilateral cleft lip, B) Bilateral cleft lip and palate, C) Cleft palate

Source: https://www.cdc.gov/ncbddd/birthdefects/

1.2 Development of OFC

During human embryonic development, the frontonasal prominence, the two maxillary prominences, and the two mandibular prominences appear. The lower portion of the frontonasal prominence differentiates into nasal placodes (ectodermal thickenings), which are paired medial and lateral nasal prominences. The two medial nasal prominences merge with one another and with the maxillary prominences on each side to form the upper lip and premaxilla or primary palate. The secondary palate is formed by the fusion of outgrowth from the paired maxillary prominences called the palatal shelves [4]. Failure of fusion of these prominences can result in facial cleft [4]. A failed fusion between the maxillary and medial nasal or nasomedial prominence on one or both sides, will result in CL. CP results from the failure of fusion between the two palatal shelves. CL and CP can occur together, resulting in CLP[5].

1.3 Aetiology of OFC

The aetiology of OFC is multifactorial and consists of interaction between genetic and environmental factors [6, 7]. Women who: smoke during pregnancy[6]; diagnosed with diabetes before pregnancy[8]; who used medicines, such as topiramate or valproic acid to treat epilepsy during the first trimester of pregnancy[9], have an increased risk of having a baby with OFC. Heavy maternal alcohol drinking also increases the risk of CLP, in addition to causing foetal alcohol spectrum disorder[10]. It has been reported that the risk for CLP development could be tripled if folic acid and cobalamins vitamin supplements are not taken during early pregnancy[11]. A retrospective study by Pradat *et al*., found an increased risk of CLP occurrence associated with systemic corticoids use[12]. Cortisone use has been reported to delay palatal shelf elevation in mice[13].

Approximately 70% of CLP cases are non-syndromic), while 30% are syndromic and associated with other abnormalities[14], including inter alia musculoskeletal defects, cardiovascular and central nervous system anomalies[15]. Several identified genes causing syndromic CLP include T-box transcription factor-22 (*TBX22*), poliovirus receptor-like-1 (*PVRL1*), and interferon regulatory factor-6 (*IRF6*), while genes associated with non-syndromic CLP are Transforming growth factor-alpha, Drosophila msx homeobox homolog-

1, 5,10-Methylenetetrahydrofolate reductase, and Transforming growth factor-beta-3 amongst others[16]. Common OFC syndromes include Apert syndrome, Crouzon syndrome, Pierre Robin syndrome, and Treacher Collins syndrome[16].

1.4 Epidemiology of OFC

OFC are amongst the most common congenital malformation in the craniofacial complex with a birth prevalence[17].that varies according to geographic location, socio-economic status, and ethnic background. Worldwide, it is estimated that a child with OFC is born every three minutes[18]. The worldwide birth prevalence ranges from 1.47 per 1 000 live births[19] to 1.25 per 1000 live births[20].

Regional variation and racial differences in birth prevalence of OFC have been demonstrated in several studies. In Asia, North America, Europe, Oceania, South America, and Africa, birth prevalence were reported to be 1.57 (1.54-1.60), 1.56 (1.53-1.59), 1.55 (1.52-1.58), 1.33 (1.30-1.36), 0.99 (0.96-1.02), and 0.57 (0.54-0.60) per 1,000 live births, respectively[19]. It is easier to confirm birth prevalence in high income countries where infrastructure and resources allow for detailed descriptive data[20]. In low- and middle-income countries (LMICs), where birth prevalence rates maybe higher in areas that are away from health care delivery systems, it may be difficult to find complete records[19]. It has been demonstrated in several studies that the birth prevalence of OFC is highest among Asians, followed by Caucasians, and lowest in African populations[21-23].

In Africa, it has been reported that the birth prevalence rates for black Africans and Caucasians ranging from 0.3 to 1.65 per 1000 live births[19]. In Ethiopia, the birth prevalence rate was estimated at 0.20 per 1000 live births (1 per 5000 live birth)[24], Kenya at 1.7 per 1000 live births (1 per 590 live birth)[25], Nigeria at 0.5 per 1000 live births (1 per 2000 live births)[26], and Uganda at 0.8 per 1000 live births (1 per 1250 live birth)[27]. In the Democratic Republic of Congo, a study reported an incidence of non-syndromic CLP as 0.8 per 1000 live births (1 per 1250 live birth)[28]. A study in southeast Ghana to measure the birth prevalence through community self-reporting, reported the prevalence of 6.3 per 1000 people with CLP [29].

In South Africa, CLP is one of the five most common congenital anomalies notified [30, 31], with a birth prevalence rate estimated at 0.3 per 1000 live births (1 per 3333 live births) in the

public sector[32]. CLP is specified as a priority condition for monitoring both in the South African *Policy Guidelines for the Management and Prevention of Genetic Disorders, Birth Defects and Disability (2001) and National Guidelines for the Care and Prevention of the Most Common Genetic Disorders, Birth Defects and Disabilities (2005)*[33]. Results of national surveillance of OFC's, along with other priority conditions reported via the Birth Defect Notification Tool (BDNT) between 2006-2014 were published in 2016[34]. Of the 13 252 congenital anomalies reported, only 943 CLP cases were reported with a birth prevalence of 0.07 per 1000 live births[34]. The BDNT is currently undergoing revision to improve reporting quality and adherence, due to the significant under-reporting identified[34].

1.5 OFC laterality and gender differences

The proportion of bilateral cleft has been reported as 10.3% for CL and 30.2% for CLP[35]. Unilateral cleft occurring on the right side is 36.9% for CL and 41.1% for CL with CP[35]. Several studies have shown that left side clefts dominate in occurrence, CLP affects more males than females and CP affects more females often than males[32, 36, 37], despite a twenty-year study that could not identify an association between sex and laterality of CLP[38]. A study in Scotland found male dominance for left-sided clefts[39]. OFC involving the lip was observed to be higher in males than females in a ten-year study undertaken in Northeast Mexico[40] with similar results reported by Knox in Northumberland and Durham [41].

1.6 Individuals and parents' perception of OFC

Patients with CL with or without a CP or isolated CP often experience challenges with feeding, speech, ear infections, hearing problems and dental complications[1]. Psychological disturbances may arise from belief systems, cultural practices, and parental factors[42]. Mothers of affected children may have feelings of guilt and self-blame for giving birth to a child with an OFC, and sometimes may feel uncomfortable and/or shamed by courtesy stigma expressed through staring, gossiping, and excessive questioning[43]. A young adult's experience of growing up with an OFC may also impact their decision on starting a family[44].

1.7 Management of OFC

Children affected by OFC have higher morbidity and mortality throughout life than unaffected individuals[45]. The mortality rate has been reported as 36 per 1000 affected births [46].

Compared with the overall population, infants with OFC are nine times more likely to die before one year of age [47]. As a result, they require multidisciplinary care from birth until adulthood[45, 48]. Treatment of CLP is readily available in developed countries such as the United States and throughout Europe. The Initial surgery is undertaken in the first few months of life followed by ongoing comprehensive care throughout their youth[49]. The same cannot be said for developing countries with limited health resources, where there is a huge backlog of untreated patients with OFC [50]. Access to surgery and related care is a challenge in the developing world, and children with unrepaired clefts may face lives filled with hardship, solitude, and shame[49].

South Africa has acquired more than six decades in treating children with CLP and other types of craniofacial anomalies[51]. However, the country is still amongst the majority of LMICs that lag behind in terms of access to care provided by a multidisciplinary team, with national treatment protocols, standardised data collection, and evaluation of treatment outcomes[51]. Although there is still no universal protocol for OFC repair and ongoing clinical management, the common elements of clinical standards and the sequence of treatment of CLP are detailed in Table 1[32]

Table 1: Globally recommended OFC Diagnosis age and CLP treatment

Chronologic age of diagnosis	CLP intervention and Treatment
Preconception care[52]	• Identification of risks factors associated with adverse pregnancy outcome
Pre-natal[53]	• Ultrasound diagnosis • Surgical plans, procedures information, and termination of pregnancy option • Prenatal genetic counselling
At birth	• Genetic counselling • Feeding plate • Pre-surgical infant orthopaedics • Psycho-social counselling
3-months	• Surgical lip repair
6-12 months	• Surgical palate repair • Grommets • Speech therapy • Routine dental treatment
5-7 years	• Alveolar bone graft • Speech therapy • Dental treatment/maxillary expansion/bone graft/dental arch alignment • Speech therapy
10-14 years	• Orthodontic treatment/maxillary expansion/bone graft/maxillary protraction
16-18 years	• Orthodontics treatment/orthognathic surgery/maxillary advancement • Psychology counselling
18-20 years	• Prosthodontic replacement of missing teeth • Nose revision

Source: Adopted from P Hlongwa et al [2019]

1.8 The multidisciplinary team

Treatment of individuals with OFC requires a coordinated and specialised treatment offered by a multidisciplinary team (MDT)[54-56]. This includes, inter alia, medical geneticists, genetic counsellors, maxillofacial surgeons, otolaryngologists (ENT), orthodontists, paediatricians, plastic surgeons, paediatric dentists, psychologists, professional nurses, social workers and

speech and language therapists and psychosocial support through linkages to non-profit organisations (NPOs)[57].

1.8.1 The Dentist

In early care, pre-surgical orthopaedics and naso-alveolar moulding using a dental plate, and sometimes lip strapping, may make the primary surgery a much easier procedure[58]. OFC conditions are characterized by supernumerary teeth, crowding, tooth cavities, missing teeth, impactions, and delayed eruptions. Therefore, a dentist working closely with the orthodontist and plastic surgeon to manage these conditions is necessary[59]. The dentist's role in managing patients with OFC include; dental examination, preventive management, restorative care, pulp therapy space management, minor orthodontic treatment, extractions and communication with cleft team[59].

1.8.2 The ear, nose, and throat specialist (ENT)

The tensor veli palatini muscle of the palate can be dysfunctional in patients with CP. Disruption of the function of this muscle may result in dysfunction of the eustachian tube in the ear. As a result, there can be an increased risk of otitis media with middle ear effusions[60]. Otitis media may lead to hearing loss and associated speech and language delays[61]. Regular ENT and audiology consultations are important for children with OFCs.

1.8.3 The Medical Geneticist and/or Genetic counsellor

When a child is diagnosed with CLP, appropriate counselling including the aetiology and embryology of clefts, detailed family history and the risk of recurrence should be provided by a medical geneticist or genetic counsellor[62]. When a patient is suspected to have an identifiable congenital syndrome, referral to a medical geneticist and/or genetic counsellor is required for appropriate evaluation, diagnosis, (by the medical geneticist) counselling, and testing when appropriate[63].

1.8.4 The Maxillofacial surgeon

Alveolar bone graft and orthognathic surgery are the two main procedures performed by the maxillofacial surgeon in patients with OFC[64]. Alveolar bone grafting is a procedure involving the filling of the alveolar bone cleft to increase maxilla stability, alveolar bone morphology, and allow the eruption of teeth into the grafted area[65]. Ideally, the procedure is performed between the ages of nine to twelve [64]. Orthognathic surgery is performed after the completion of facial growth to correct sagittal maxillomandibular jaws discrepancies that are often observed in patients with OFC[64]. Other procedures performed by maxillofacial surgeons or plastic surgeons include lip repair and nasal reconstruction[66].

1.8.5 The Orthodontist

Orthodontists play a significant role as part of the craniofacial team developing the treatment plan. From birth to seven years of age, the orthodontist constructs neonatal maxillary orthopaedics to bring the maxilla into alignment[59]. Around the age of seven, treatment may include a face mask to protract the maxilla[59]. At approximately eight years of age, the maxillary arch transversal deficiency and the posterior crossbite may be corrected by rapid maxillary expansion. This expansion also helps to prepare the maxillary arch to receive the secondary alveolar bone graft[59]. Around nine to eleven years of age, concurrent with alveolar bone grafting, fixed appliances may be placed on the maxillary arch to eliminate crossbite and malpositioned teeth[67]. In the permanent dentition stage, from ten to thirteen years of age or even older, there is a determination of whether orthognathic surgery is indicated[59].

1.8.6 The Paediatrician

The paediatrician plays an essential role in diagnosis and referral, acute care, ongoing medical care, as well as serve as a link between the speciality team and the family[68].

1.8.7 The Plastic surgeon

OFC conditions may present with distortions of skin, muscle, cartilage, mucous membranes, teeth, and bone, which are elements that affect communication, facial expression, and

appearance[69]. To restore an aesthetically natural appearance, plastic surgeons perform surgeries that correct cleft lip, as well as the associated nasal deformity[63]. Palatal repair is recommended at about the time the child begins to develop plosives b, d, and g in speech, at about age 11 to 12 months, rather than first waiting for maxillary growth as was undertaken historically and consequently affected speech development[63].

1.8.8 The Psychologist and/or social worker

Children with CLP are likely to be socially isolated, experience stigma and teased by their peers because of the less socially acceptable facial appearance. These children may have speech and language difficulties, and a poor self-image[70]. Reconstructive surgery alone will not solve all the patients' psychological problems. Therefore, post-operative psychological support is as important as pre-operative psychological management as these children will still need to be treated for previous psychosocial problems[42]. Caregivers may also need psychosocial support because of the burden of raising a child with OFC and the risk of burnout [43].

1.8.9 The Prosthodontist

A prosthodontic prosthesis may be necessary for conditions that include un-operated palates, failure of surgery to close the CP, a wide cleft with insufficient local tissue available to repair, individuals with soft palate and pharynx neuromuscular deficit, contra-indication to surgery due to medical condition, delayed surgery, patients that need removable and fixed prosthesis for missing teeth[71].

1.8.10 The professional nurse

Paediatric nursing care for children with OFC begins in infancy and continues through adolescence, by the provision of relevant care for these patients, caregivers, and families. This care includes educating patients and parents about the development and intervention options available to promote informed decision making, as in addition to meeting their immediate medical needs. The OFC paediatric nurse has a basic overview of normal lip and palate development, modes of inheritance, genetic and environmental causes, classification of clefts,

pathophysiology of clefts, incidence, genetic counselling, prenatal diagnosis, and corrective surgery[72].

1.8.11 The Speech therapist

Patients with OFC often present with distorted speech sounds resulting from oral anatomical structure anomalies[73]. Speech therapy is effective in increasing consonant proficiency in patients with cleft[74]. Consonant sounds like p, b, t, d, k, g, are the most challenging to pronounce in individuals with CLP [75]. Hypernasality often occurs when both the oral and nasal cavities vibrate and enhance the sound during a speech, caused mainly by velopharyngeal insufficiency which may be treated by speech therapy [76]. Surgery may be necessary for 20% of children to achieve intelligible speech[76]. In addition, articulation challenges that are caused by a short or narrow maxillary arch are often difficult to correct[76]. Speech and language therapy is essential, especially in the early period of speech development, especially by/before? age three, where abnormal speech patterns has been detected[77].

1.9 Non-profit organisations involved in OFC treatment

The term non-profit organisations (NPOs) are used interchangeably with non-governmental organisations (NGOs), and for the purpose of this study, the term NGOs will be used. There are several NGOs involved in CLP management based in different locations worldwide with a variety of mandates. Many countries NGOs focuses on providing cleft support nationally and to localised country regions. Others have an international focus on LMICs. An overview of the main OFC NGOs operating globally are detailed in Table 2. Note: this focuses on the key OFC NGOs (i.e., those with footprints in Africa) and does not claim to be a complete list.

Table 2: Summary of key OFC NGOs and support they provide

NGO	Country of origin	Countries operating	Mandate and services
1. Smile Train (ST)[78]	USA	More than 90 countries globally	Provides comprehensive cleft care, empower local professionals and awareness services internationally, including 33 African countries. In South Africa, the NGO only support orthodontic care.
2. Operation Smile (SO)[18]	USA	More than 30 countries globally	International provision of cleft surgery support, dental support, speech and nutrition therapy, and post-operative care. In some places, they supply medical equipment. South Africa is one of the 10 African countries that SO supports, but cleft support is limited and not clearly defined.
3. American Cleft Palate-Craniofacial Association (ACPA)[79]	USA	Not specified	Provides educational and research opportunities for cleft and craniofacial professionals around the world. ACPA also offers educational materials about cleft and craniofacial care, to affected individuals and their families.
4. Alliance for Smiles (AfS)[80]	USA	China and Nigeria	Provides free surgical CLP repair, dental care, speech therapy, orthodontics, and sometimes psychological counselling in Asia and Africa (Nigeria). No cleft support has been recorded in South Africa
5. Transforming face (TF)[81]	Canada	Nine countries globally	Collaborates with local health and community centres to ensure cleft care and supportive services in nine countries, including two African countries, Uganda, and Ethiopia. No cleft support has been recorded in South Africa
6. Project Harar[82]	UK	Ethiopia	Provides outreach programmes that recruit and transport patients with cleft to their nearest hospitals for surgery, provide them with nutritional support and refer to speech therapists
7. Smile Foundation (SF)[83]	SA	South Africa	Collaborates with 11 South Africa's Academic Hospitals to augment corrective facial reconstructive surgery and treatments services. SF also encourage, informs, and educate parents of children with clefts throughout the reconstructive process. In addition, SF supports the teams of qualified medical staff with skills development programs, as well as the purchasing of medical equipment for plastic and reconstructive surgery in the departments within the hospitals.

1.9.1 International NGOs

Smile Train

Smile Train (ST) is an international children's charity organisation established to relieve the burden of OFC. Based in the United States of America (USA), ST operates in more than 90 countries, partnering with more than 1100 hospitals and over 2100 medical professionals, and has supported more than 1.5 million OFC surgeries to date[84]. Services of ST include surgeries, nutrition services, orthodontic and general dental care, speech therapy and emotional support. This NGO also provides surgical training resources, workshops, and education opportunities to cleft surgeons, anaesthetists, nurses, and related medical professionals around the world[78].

Through donor funding, ST supports local centres with equipment and supplies that are essential for OFC surgery, including safety equipment like pulse oximeters, surgical instruments, sutures etc. ST also raises awareness about OFC, achieved through radio adverts and programmes, fliers, newspaper ads, collaborations with other organizations for patient referrals, and SMS/text message alerts, to reach patients in the most remote of areas across the globe. While OFC surgery is provided for free, many patients also require transport support to or from the hospitals to access treatment. Special programmes that offer small stipends to those in need to cover travel costs are also implemented by this organisation worldwide[78].

ST supports OFC treatment in 33 African countries and has achieved 100 000 cleft surgical cases in Kenya in 2018 alone[85]. In South Africa, Smile Train funds the Wentworth Foundation based in Durban, to provide orthodontic treatment to individuals with CLP who are referred from surrounding hospitals[86].

Operation Smile

Operation Smile (OS) is an international NGO that provides cleft support to more than 30 countries, involving more than 6000 volunteers across 60 countries[18]. Based in the USA, OS provides surgery and dental care for more than 19 000 children each year by deploying more than 100 medical missions annually[18]. Over two weeks, each mission includes health screenings, speech and nutrition therapy, dental care, surgery, and post-operative care. In some locations, they also supply medical equipment, and in others, deploy specialized medical

volunteers that work with the local community to create solutions[18]. They have also established 31 centres in 16 countries that provide free services year-round for those individuals who require more complex treatment than a single mission can provide. These services include dentistry, orthodontics, psychosocial care, speech therapy, nutritional counselling, orthognathic and cleft surgeries[18].

Approximately 85% of the OS medical volunteers are from the LMICs in which OS serve[18]. Majority of these professionals have received training and education from OS, which invests in health system strengthening and partnering with universities, NPOs and governments to deliver surgery to those who need it most. In Africa, OS operates in 10 countries including South Africa. In Rwanda, OS's intervention is crucial to mitigate challenges included lack of expertise - with only two reconstructive plastic surgeons and 18 anaesthesiologists serving 12 million people. Patients are forced to wait years for treatment because of a lack of adequately trained surgical teams[18].

In South Africa, OS has made announcements to conduct cleft medical missions in Witbank, Mpumalanga, in 2019 and 2020[18].

American Cleft Palate-Craniofacial Association

The American Cleft Palate-Craniofacial Association (ACPA) supports team care in the USA and around the world by striving to enhance and expand educational opportunities for interested professionals who want to improve in clinical care and research advancements in cleft and craniofacial conditions. ACPA also offers educational materials about cleft and craniofacial care, to affected individuals and their families[79].

Alliance for Smiles

Alliance for Smiles (AfS) is an NGO based in San Francisco, that provide free surgical repairs for CLP in Asia and Africa. With five treatment centres located in China, and one treatment centre in Lagos, Nigeria, AfS conducts two-week surgical missions and provides training for local health care providers to serve the under-served patients with CLP. They have established permanent treatment centres where patients receive follow-up and ongoing care, utilizing the USA treatment protocols. Patients with OFC receive ongoing treatment in dentistry, speech therapy, orthodontics, and sometimes psychological counselling[80].

Transforming faces

Transforming Faces (TF) is an NGO based in Canada, that collaborates with local health and community centres to ensure cleft care services. This organisation provides OFC services in nine countries, including two African countries, Uganda, and Ethiopia. In Uganda, this NGO focuses on children from rural districts with a high incidence of CLP by mobilizing and providing transportation. Through radio talk shows and community workshops, they promote community awareness about comprehensive OFC care. They also provide hospital-based nutritional support, counselling, and speech therapy. TF launched a new three-year project in 2020 to expand comprehensive OFC care services for patients with CLP and their families. Relevant resources and qualified professionals to deliver surgery and speech therapy are often unavailable to patients living in remote locations and have to travel long distances to access relevant care. Another challenge is the lack of knowledge of the importance of comprehensive cleft care[81].

In Ethiopia, TF works in Yekatit 12 Hospital, the only comprehensive OFC care centre providing services that include surgery, speech therapy, dentistry, orthodontics, and social work, where cleft care was provided by TF for 492 patients in 2019. TF has partnered with other cleft organizations to expand access to services such as speech therapy. This includes partnering with Project Harar and CURE Ethiopia Children's Hospital, through which speech therapy was delivered to patients in rural parts of the country. TF has played a significant role in the establishment of the first formal degree programme for speech therapy at Addis Ababa University. In 2019, 16 speech therapy students graduated, bringing the country's total of formally trained speech professionals from two to 18. This number of qualified speech therapists is still inadequate to serve a country with a population of over 100 million[81].

Project Harar

Project Harar is a United Kingdom (UK) based organisation working in Ethiopia to help children affected by OFC in remote communities of Ethiopia. Their outreach workers recruit and transport patients with OFC to their nearest hospitals for surgery, provide them with nutritional support and refer patients to speech therapists[82].

1.9.2 South African NGO

In South Africa, NGOS are regulated by the Non-profit Organisations Act 71 of 1997 and requires that all legal NPO entities (voluntary associations, trusts or a Section 21 Company's) register and submit financials and narrative reports annually to the NPO Directorate of the Department of Social Development[87].

Smile Foundation

SF is a registered NPO (NPO 063-498) and became a Section 21 Company in 2007 [88] under the Companies Act 61 of 1973 (followed by the new Companies Act of 2008) [89]. SF is also a registered Public Benefit Organization (PBO 930030006) with the South African Revenue Services (SARS) under the Income Tax Act [90]. PBO status entails two tax benefits: tax-exemption for the organization, and tax relief for the organization's donors under Section 18A [90].

Smile Foundation (SF) is a South African NGO that collaborates with 11 country's Academic Hospitals to augment corrective facial reconstructive surgery and treatments services. SF also assist parents of children with OFC by encouraging, informing, and educating them throughout the reconstructive process. In addition, this organisation supports the teams of qualified medical staff by investing in furthering skills development programmes, and purchasing of medical equipment for plastic and reconstructive surgery in the hospital departments [91].

1.10 Role of NGOs in OFC treatment support

In every community, there are uninsured individuals who need surgical care[92]. More so in many LMICs that rely on the NGO sector to assist with surgical intervention due to a lack of proper surgical expertise and infrastructure [93].NGOs offer surgery days of donated care to reduce the burden of uncompensated care[92] and partner with local hospitals to provide surgical care[94]. It is believed that they rely on donated funds to assist the hospitals[92]. Literature has pointed out the need for evaluation of effectiveness and cost-effectiveness of NGOs in the delivery of surgical care in LMICs[95]. The limitation of OFC data is evident in Africa where insufficient funding and resources affects the ability to perform research[96]. In addition to poor healthcare infrastructure and increased mortality of children with clefts[97, 98], unrecorded home births, record-keeping challenges and OFC stigma also contribute to limited data of OFC care[99].

1.11 Study Rationale

Limited OFC treatment in LMIC such as South Africa, has encouraged many international OFC NGOs to assist with surgical correction and related services[96]. As detailed in Table 2 under non-profit organisations involved in OFC treatment, many of these organisations have demonstrated the positive impacts of their interventions [100]. Through NGO's interventions, evidence of volunteers gaining new skills in CLP care, individuals with CLP able to access local support network, able to cope with CLP related challenges, and gaining self-confidence, have been demonstrated[101].

In South Africa, gaps exist in the types of treatment provided across the specialised academic CLP care centres, including lack of MDT members, and a reported absence of standardised treatment protocols [51]. NGOs such as OS and SF help fill the gap to meet these needs by facilitating reconstructive and corrective plastic surgery[102]. While these NGOs have been operating in the country for a number of years and facilitating the care and correction of many patients affected by OFC. There is little evidence of their impact as only limited studies have been undertaken to analyse OFC services they provide[21]. Evidence is needed to quantify the role and achievements of these NGOs in South Africa and prompted the question: "how are NGOs positively managing cleft lip and palate individuals in South Africa?". For convenience and easier access to data, this study evaluated the data of a national NGO in South Africa to analyse its involvement and impact in the management of CLP.

1.12 Aim of the study

The study aimed to assess the services of a single NGO, Smile Foundation (SF), operating in South Africa in managing OFC.

1.13 Research objectives

The research objectives of this study are:

1. To describe the geographic centres in South Africa where SF is operating.
2. To describe the types of OFC services rendered by SF.
3. To identify the number and types of OFC procedures facilitated SF.

CHAPTER 2

Methodology

2.1 Study design

This observational, descriptive study explored two components: 1) A case study on SF to obtain the organisation's background and care management approach (qualitative data); 2) An audit of the clinical outputs (quantitative data), using a cross-sectional study design to analyse records of OFC cases facilitated by SF from 1 January 2015 to 31 December 2019.

2.2 Study setting

Due to the COVID-19 pandemic, the study setting was changed to online to minimise personal contact and exposure risk. To obtain the background and a management approach of SF (qualitative data), a semi-structured interview was conducted online using the Zoom meeting platform on the 4th of December 2020. Quantitative data was accessed and analysed from an electronic database for the study period.

2.3 Study population and sampling

The Chief Executive Officer (CEO) and the Operations Executive Director (OED) of SF were interviewed by the PI to obtain information data related to the mandate of SF and the management approach. Furthermore, electronic records of OFC surgeries facilitated by SF in South Africa during the five-year study period were made available to the PI and reviewed.

2.4 Data collection

Qualitative data

A semi-structured interview questionnaire was adapted from an existing questionnaire on CLP care developed by Hlongwa et al in 2015[51]. The revised questionnaire (Appendix 1)

contained 13 questions divided into two parts: 1) NGO background information and scope of operations and 2) care management for OFC patients.

Permission was granted by the SF CEO for the study to be conducted and both the CEO and EOD were provided with the participant information sheet (Appendix 2) and informed consent (Appendix 3) obtained from both of them to participate ahead of the interview Zoom session. The interview was conducted by the PI in English, which is the official business language in South Africa. The interview took place online using the Zoom platform and lasted 40 minutes. All responses were entered into the structured questionnaire during the interview, and a video recording was undertaken concurrently, following consent, for accuracy of data collection. Section 1 of the questionnaire focused on: reasons for the existence of SF in South Africa, type of business, organisational structure, vision and mission, operational start date, provinces, and hospitals where care centres are situated and the frequency of care involvement of these centres. Section 2 evaluated care and management, including: the point of care for individuals with CLP, services rendered, treatment protocol, care provision approach, and team members involved in surgical repairs.

Quantitative data

A structured, pre-tested record review form [32] (Appendix 4), was used to obtain demographic, clinical and treatment information. Data records for the study period shared by SF were de-identified and centres were allocated unique numbers for anonymity and confidentiality. Specific clinical data sought by the PI included: the number of patients with OFC who received treatment per year, patient race, gender and age, OFC associated anomalies, cleft anatomical location, cleft laterality, and the type of surgeries performed.

2.5 Data management and analysis

Qualitative data

The responses provided by the interviewees during the semi-structured interview were written down in the space provided in the questionnaire and were transcribed post-interview and saved in a Microsoft Word © format. The interview recording was also transcribed into Microsoft

Word © format and compared with the typed verbatim responses for accuracy. The responses were transferred to an Excel spreadsheet for analysis.

Quantitative data

The data provided by SF was exported and captured into Excel software and then imported into STATA® 14 for descriptive statistical analyses. Frequencies, percentages, tables, and graphs were used to display data. All statistical tests were conducted at a 95% confidence level.

2.6 Ethical considerations

Ethical approval was obtained from the Human Research Ethics Committee (Medical) of the University of the Witwatersrand in Johannesburg to conduct the study (Ethics number: M200909) (Appendix 5). Permission for data collection was also obtained from the CEO of the SF (Appendix 6). Confidentiality and anonymity were maintained throughout the study, and information that could identify patients was not used.

CHAPTER 3

Results

3.1 Qualitative results

The Chief Executive Officer (CEO) and the Operations Executive Director (OED) of SF were interviewed by the PI to obtain information regarding the organization's involvement in South Africa.

Interview outputs on the role of SF and care management approach

The mandate of SF

SF was registered as an NPO in South Africa on 1 September 2008. SF aims to augment the Department of Health (DOH) by supporting children born with facial and cleft deformities to obtain reconstruction surgeries, using a multidisciplinary approach. The surgeries are not performed by SF themselves, as their role is to support and facilitate the critical requirements for CLP treatment and care.

Vision and Mission

SF's vision is to transform the lives of children born with facial conditions in South Africa. The organisation's mission is to bring people together for the purpose of providing expert surgical intervention, creating greater social involvement, and enabling sensible corporate social investment to make a difference in the lives of children born with facial anomalies.

History and Scope

The SF is an NPO that started its operations in South Africa in the year 2000 as Star Smile Fund, becoming a registered NPO eight years later. The organisation started its operations in one centre in Gauteng (centre 8). SF was formed following a personal request from former President Nelson Mandela to help a little girl living with a facial anomaly known as Moebius

Syndrome. The Star Smile Fund's growth necessitated establishment as a Section 21 and re-launched as the Smile Foundation in 2007[88].

Centres where SF is involved

By the year 2019, the SF facilitated OFC services in six provinces: Gauteng (four centres); Limpopo (one centre under Gauteng's DOH); Free State (one centre); Eastern Cape (one centre); Western Cape (centre); Northern Cape (one centre); and Kwazulu Natal (one centre).

Organisational structure

Figure 2 shows SF's organisational structure.

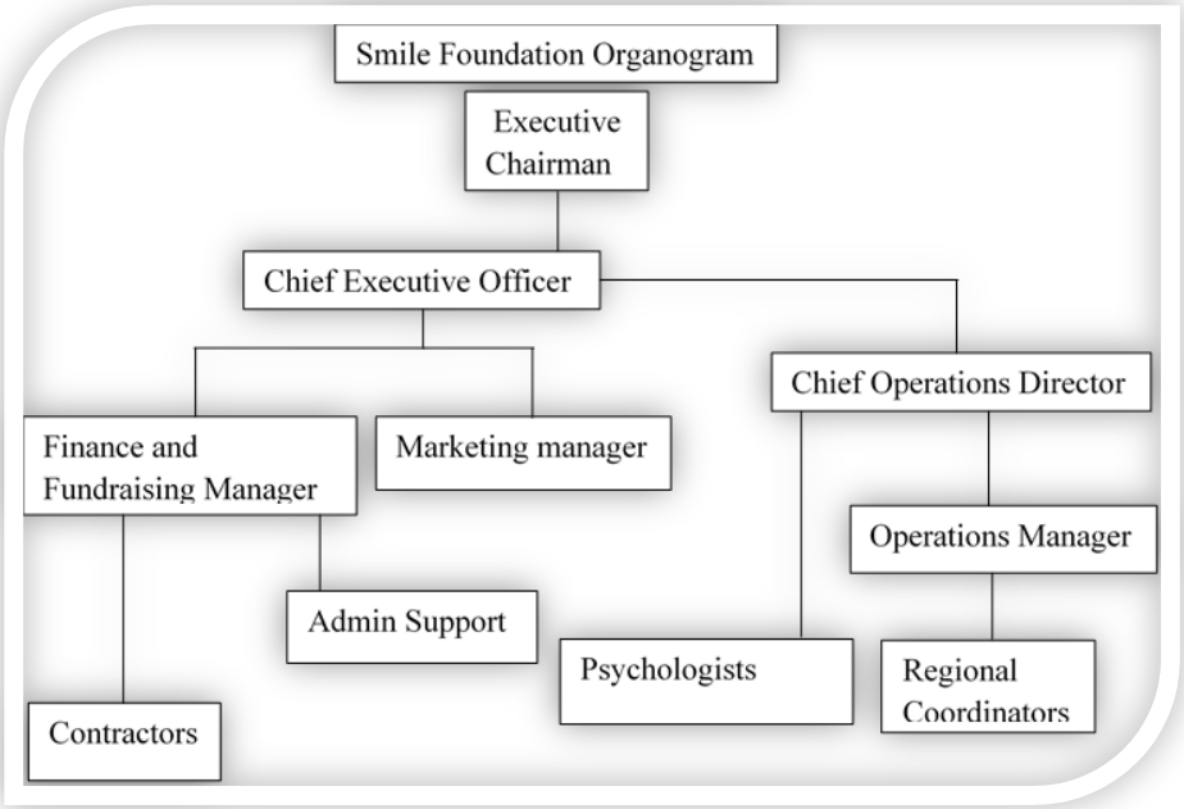


Figure 2: Smile Foundation organisational structure

Source: Smile Foundation

Facility Support

SF provides support to the centres in three keyways:

1)'Smile Week' is held annually at each centre where surgeons, nurses, psychosocial debriefing, mother to mother support, post-trauma counselling and various stakeholders (hospital, patients, media, donor) collaborate in a multidisciplinary approach during this week. Transportation and accommodation support is provided for patients to attend these focus weeks. In this week, major reconstructive surgical procedures are performed (between 20 and 30 surgeries), costs on pre-and-post operative therapy are covered, consumables used during surgery are provided, mobilisation of operating theatres and additional nurses to supplement nursing staff, entertainment, and educational and other gifts for the children are distributed.

2) Financial Support: SF facilitates the reduction of hospital surgical backlogs on a weekly basis. This is largely by financial support by covering relevant staff and material costs, such as the salaries of specific health care professionals (e.g., an anaesthetist, three nurses and psychosocial support on Thursdays at centre 2 and supporting burn patients (Centre 3) every second Saturday). Financial support is also provided to cover treatment for urgent, ad hoc cases e.g., a one-day specific case of craniofacial surgery that requires financial assistance. Other support services that SF facilitates include the purchase of theatre equipment when required, support groups, awareness campaigns and outreach programmes.

3) SF Hospital Coordinators: SF hospital coordinators visit centres in most regions on a weekly basis according to the support required for the surgeons and their teams. The SF coordinators welcome the parents of the patients at the centres the day before surgery is performed. A SF psychologist also provides pre-and post-surgery counselling. The specific degree of support depends on the needs of the centre, as some centres already have strong psychosocial support in-house and do not require additional SF support. In addition to surgeries and psychology, SF support ENT services, and relevant allied health care support including speech therapy, occupational therapy, feeding plates and bottles in certain cases.

Capacity Building

Training of key hospital personnel is another component of SF support provided. Different donors are approached by SF to fund specific projects and programs. Through SF, sponsorships would cover registration fees for all the registrars attending the annual conference of The Association of Plastic, Reconstruction and Aesthetic Surgery of South Africa (APRASA). Upon request, sometime SF would pay an international speaker and his/her travel costs.

There are also opportunities for registrars to access online journals and books which are fully paid by a major donor through APRASSA. Registrars from Academic hospitals are also given the opportunity to attend the Smile Weeks in order to expand on their current skills base. There are also annual registrar's symposiums which SF offer financial support for the registrars to observe or assist in the theatres during the various surgeries, not limited to CLP, and learn new techniques from different consultants. In some cases when budget permits, SF funds international fellowship trips for newly qualified consultants through the South African College of Medicine. Nurses are also offered skills development courses.

3.2 Quantitative results

These results describe the OFC clinical data of patients that were facilitated by SF. These data include age distribution, gender makeup, number of surgical procedures facilitated, distribution of the various surgical procedure types and distribution of surgical procedure per centre and per province.

The OFC sample

The sample included 515 hospital records of patients (238 females, 276 males, and one missing gender) who received CLP surgical treatment. Figure 3 shows the distribution of the age of the total sample.

Age Distribution of the sample

The majority of the children with OFC received their surgical treatment under the age of 10 years (95.73%, n=493), with most surgeries performed before the age of five years (71.65%, n=369).

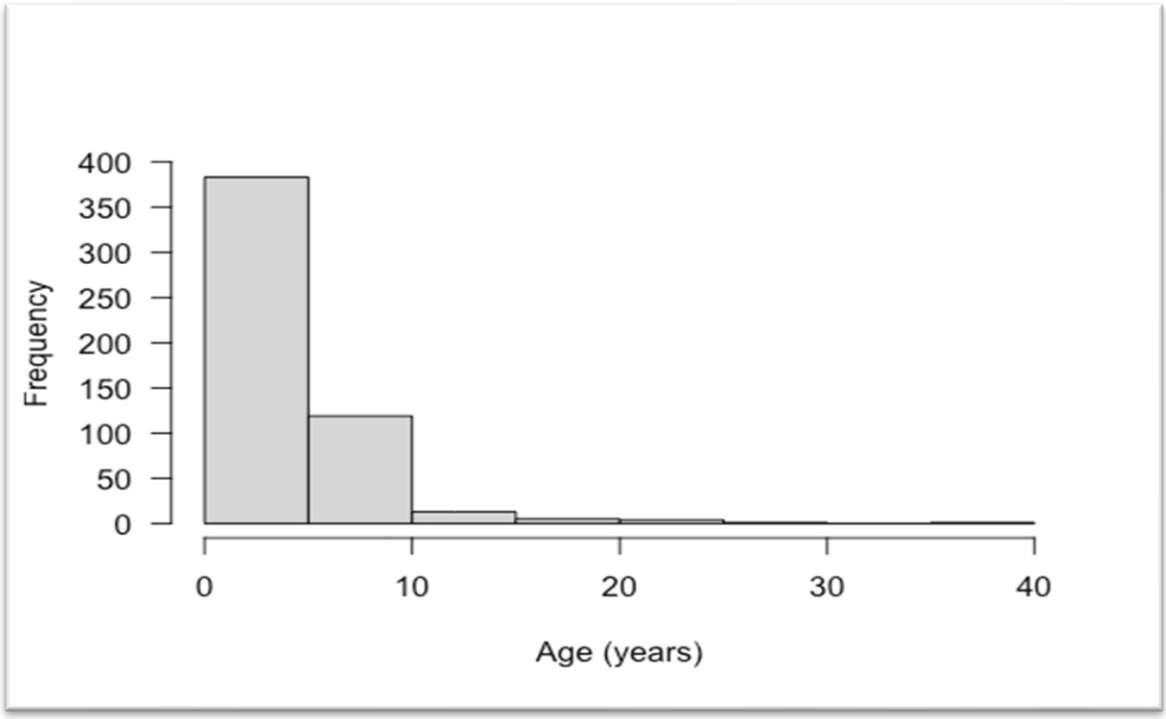


Figure 3: Age distribution for the total sample Gender

Table 3 shows the gender makeup of the sample/cohort. The median age for the female subjects was 3.1 years and the median age for the male subjects was 2.9 years. There were more male subjects (53.70%) than females (46.30%) in this sample resulting in a ratio of 1.2

Table 3: Descriptive statistics – Age of sample according to gender

Gender	Sample size (n)	1st Quartile	Median Age In Years	Range In Years	3RD Quartile
Male	276	0.0	2.9	0 to 30	6.0
Female	238	0.0	3.1	0 to 36	6.0

Surgical procedures undertaken during study period

Figure 4 shows the number of patients per year who received surgical procedures facilitated by SF during the study period. There was a varied distribution of the number of patients treated in different years. Most patients (n= 207) received surgical procedures during the year 2016, while the least number of patients (n= 27) surgical procedures were performed during the year 2019.

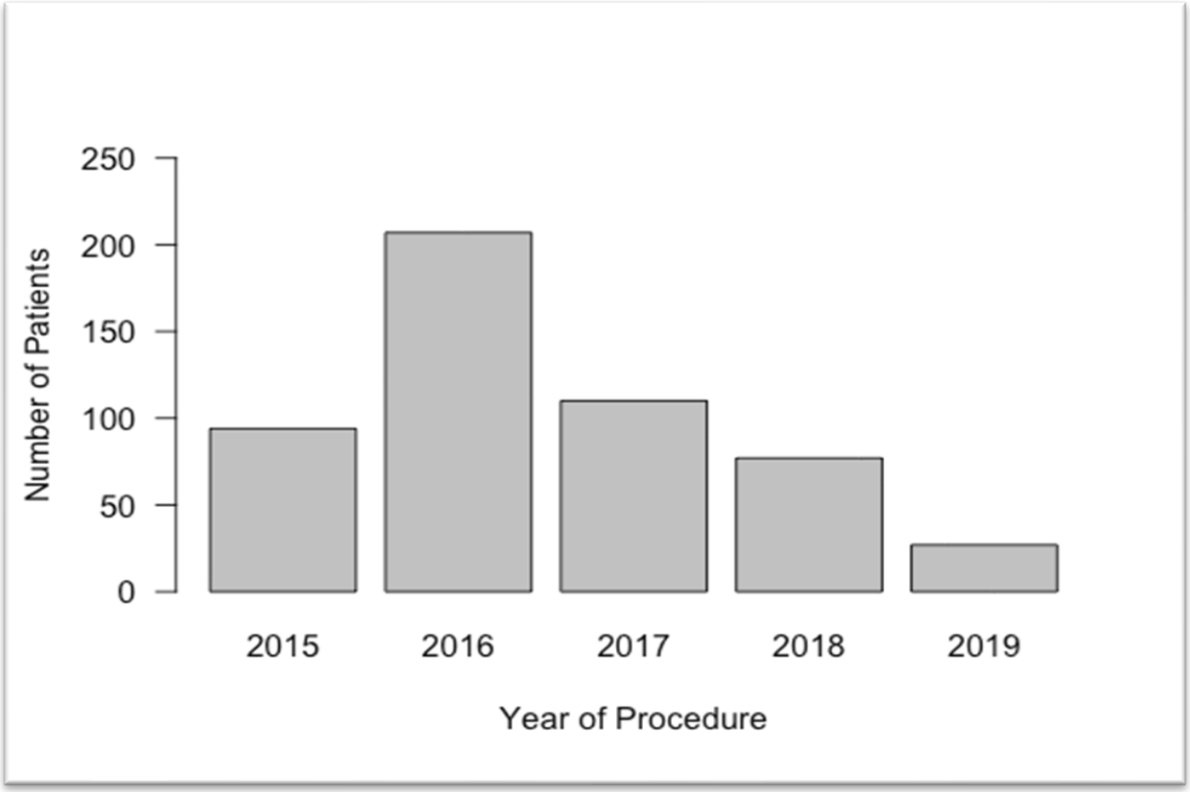


Figure 4: Distribution of surgical procedures per year over the study period

Types of Procedures Undertaken

Figure 5 shows the distribution of the various surgical procedure types facilitated by SF during the five-year period from 2015 to 2019. There was a varied distribution of the type of surgical procedures for the total sample in different years. CP surgery was the most frequently performed procedure, accounting for 48.40% (n=273), followed by CL surgery accounting for 191% (n=33.87) during the period of this study. Other procedures, such as grommets, accounted for 10.82% (n=61). Combined CLP surgery was the least frequently performed procedure during the study period (6.91%, n=39).

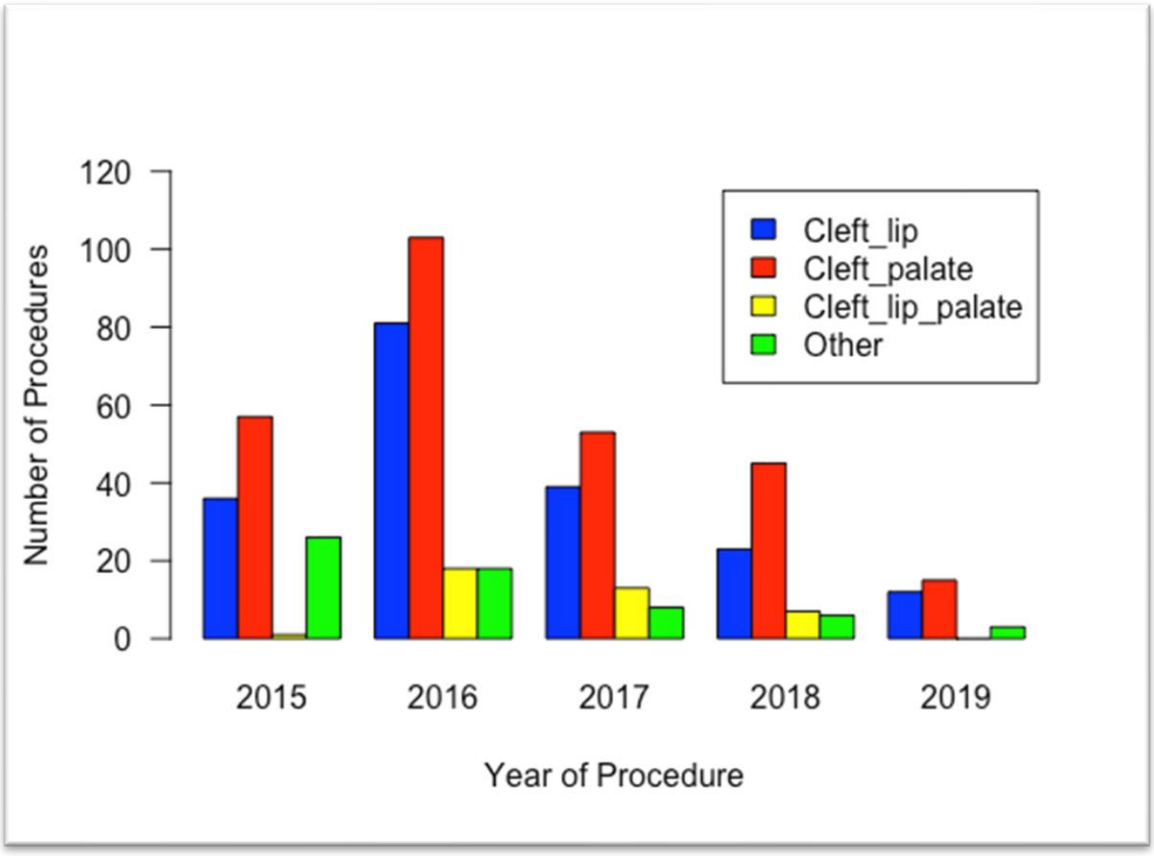


Figure 5: Distribution of type of surgical procedure for the total sample

Procedures performed in different Provinces

Table 4 shows the provincial distribution of the total OFC surgical procedures performed during the study period. The highest number of surgical procedures were performed in the Western Cape Province which had three centres (*n* = 240, 42.55%), and the lowest number of surgical procedures were performed in Kwazulu-Natal (*n* = 1, 0.18%), which had one centre.

Table 4: Provincial distribution of total OFC procedures

Province and centres	Number (*n*)	Percentage (%)
Western Cape	240	42.55%
Gauteng	198	35.11%
Eastern Cape	59	10.46%
Free State	56	9.93%
Northern Cape	10	1.77%
Kwazulu-Natal	1	0.18%
Total	564	100%

Figure 6 shows the distribution of various surgical procedures performed per province. There was a varied distribution of the surgical procedures in each province for the total sample during the study period. Western Cape had the highest numbers in 2016, followed by Gauteng and Eastern Cape, both with the highest numbers in 2016 as well. Free State had their highest numbers in 2015. Northern Cape performed their highest numbers in 2017. In KZN Province, only one procedure was performed in 2018.

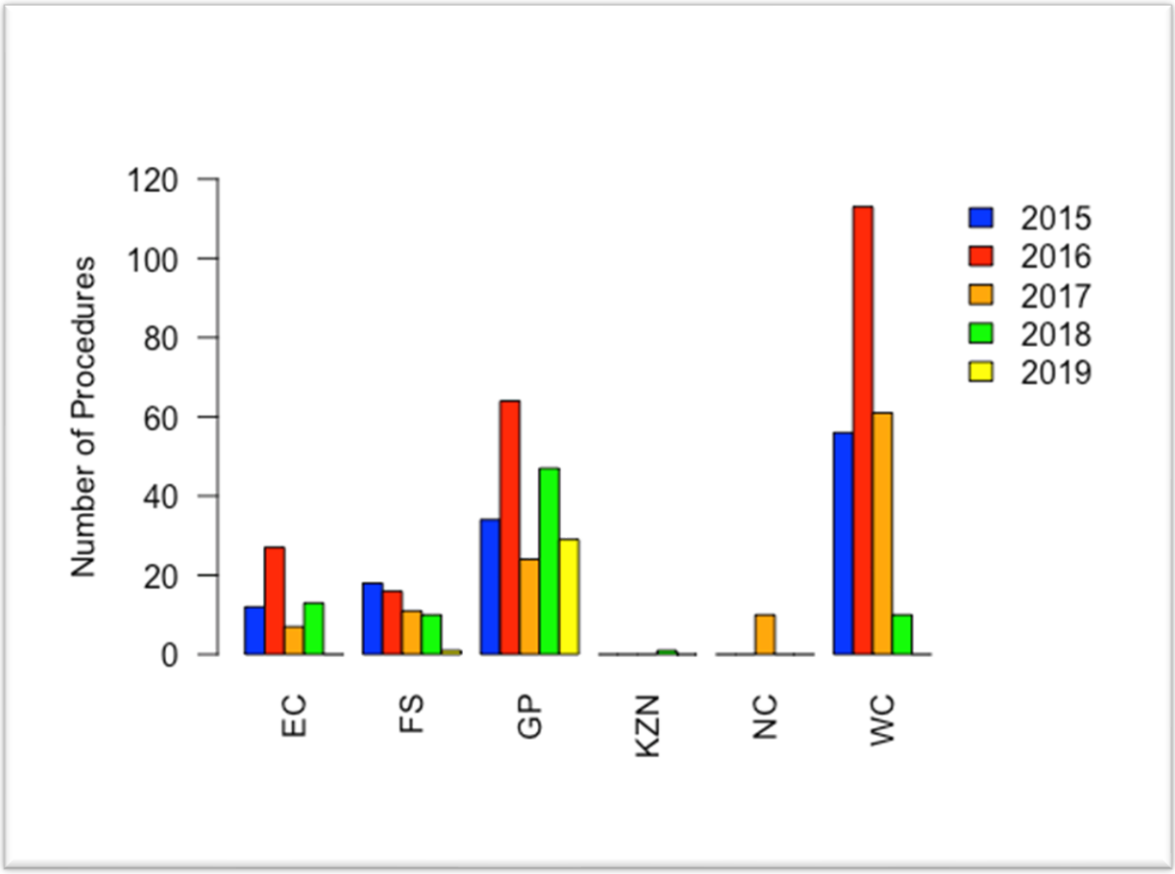


Figure 6: Distribution of surgical procedure per province

Procedures per Centre

Table 5 shows the breakdown of CLP surgical procedures performed per treatment centre. Most surgical procedures were performed in centre 2 (*n* = 138, 24.47%) in WCP and the lowest number of surgical procedures were performed in centre 10 (*n* = 1, 0.18%) in KZN.

Table 5: Descriptive statistics of procedures performed in each centre

Centre	Number (*n*)	Percentage (%)
Centre 1 (FS)	56	9.93%
Centre 2 (WC)	138	24.47%
Centre 3 (WC)	69	12.23%
Centre 4 (WC)	33	5.85%
Centre 5 (GP)	24	4.26%
Centre 6 (GP)	83	14.72%
Centre 7 (GP)	50	8.87%
Centre 8 (GP)	41	7.27%
Centre 9 (NC)	10	1.77%
Centre 10 (KZN)	1	0.18%
Centre 11 (EC)	59	10.46%
TOTAL	564	100%

FS = Free State, WC = Western Cape, GP = Gauteng, NC = Northern Cape, KZN = Kwazulu Natal, EC = Eastern Cape

Figure 7 shows the procedures performed per centre per year of the study period. Centre 2 had the most number of procedures performed in 2015, 2016 and 2017. Contrary, most centres had fewer procedures performed in 2018 and 2019.

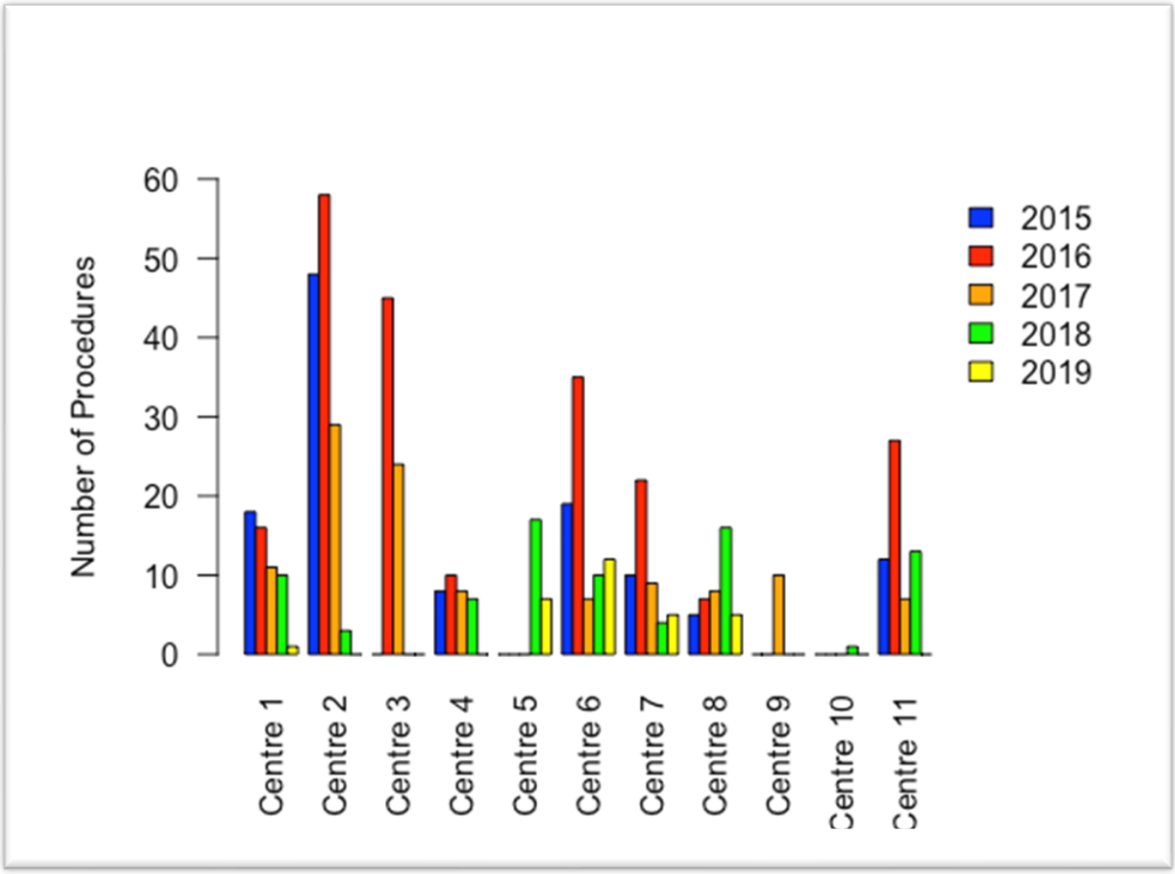


Figure 7: Procedures per Centre per year

CHAPTER 4

Discussion and Conclusions

This is one of the first studies to assess the services of a single NGO, Smile Foundation (SF), operating in South Africa in managing OFC. Globally, evidence that quantifies the role and achievements of NGOs is lacking, and this is also the case in South Africa where there are no published studies that evaluate the impact of these organisations in augmenting state services. It has been reported that in most sub-Saharan African countries, hospital-based registries provide reliable OFCs estimates[103]. Hence, we analysed 515 hospital records which SF had collected from the South African hospitals they have support from 2015 to 2019.

4.1 FS Organisational background and areas of services

The results of the interview revealed that SF assists with augmenting the DOH services by supporting children born with OFC and other deformities with reconstruction surgeries. Many NGOs have reported partnering with local hospitals to provide surgical care elsewhere in the world for OFC care [94]. This is the same approach taken by SF in South Africa, with a primary focus to address the medical backlog in the public healthcare system for the management of children affected by treatable facial abnormalities including CLP, burn survivors, Moebius syndrome (facial paralysis) and other conditions through life changing reconstructive surgeries [83].

Several NGOs usually organise surgery days of donated care to reduce the burden of uncompensated care[92]. Contributing factors for backlogs in surgical care may be the high cost for surgery and the limited state budget. In every community, some uninsured individuals need surgical care[92]. It has been reported that about 83% of South Africa's population is catered by the public health sector, while the private health sector provides care to about 17% (minority) of the population with private health insurance[104]. Lack of MDT members and gaps in the type of CLP treatment provided in South Africa's specialised care centres have also been reported[51]. All these factors may have contributed to backlogs of CLP surgical care

experienced in the care centres. There is an obvious role for NGOs such as FS to assist the state in reducing the backlog experienced by the public sector.

A European synthesis report recommended that surgeons undertake no less than 40 new primary OFC surgeries per year in European countries[105]. In South Africa, 699 patients with CLP were treated by 10 plastic surgeons in the public sector over a two years, equating to 349 surgeries per year [51]. On average, each plastic surgeon treated 35 individuals annually with CLP in total in the South African state sector, including both primary and follow up interventions. While applying a recommendation from a majority of high-income region is of limited value for an LMIC, it serves to highlight the service delivery gap in South Africa, which is further constrained by the lack of adequate numbers of surgeons. The findings of the present study demonstrate that SF supports an average of 103 of the 349 annual state surgeries in South Africa – thus augmenting more than a third (30%) of the national state provision of CLP surgical services. This is a significant contribution by an individual NPO such as SF. Both SF and the public sector have reported backlogs of untreated CLP cases. Based on this evidence, there is a clear need for extra capacity and funding in the management of CLP in South Africa to ensure all CLP patients are treated appropriately and timeously.

As outlined by the SF CEO, the organisation facilitates OFC care using a multidisciplinary approach. This approach has been recommended by several studies that emphasize the importance of coordinated and specialised treatment offered by a multidisciplinary team[54-56]. SF facilitates OFC services in 11 centres within six of South Africa's nine provinces. An earlier study conducted to determine the epidemiology and clinical profile of individuals with CLP in South Africa, collated data in ten of these centres[32], which provide services for 83% of the population seeking CLP care in South Africa's public health (state) system[32]. This highlights the alignment of the SF scope of coverage with these state centre services, focusing on multidisciplinary care for individuals born with CLP [32].

4.2 Care and management

Although SF aims to facilitate comprehensive OFC care, there are some service gaps in terms of the specialist disciplines supported. These include medical geneticists, genetic counsellors, orthodontists, prosthodontist, paediatricians, and paediatric dentists, which are recommended as part of comprehensive care[79]. Orthodontic and orthognathic treatment for patients with a

cleft is available and performed in some of the SA's centres[32], but SF reported that these treatments are not their vision to support.

Other support services that SF facilitates include the purchase of theatre equipment when required, training and workshops of medical staff members on an ongoing basis, support groups, and awareness campaigns and outreach programmes. International OFC NGOs such as ST and OS offer similar services to local centres[18, 78].

4.3 Orofacial clinical information

A total of 564 surgical repairs were supported by SF during a five-year period (2015-2019). The predominance of males with OFC reported in this study is similar to results reported by several other studies [26, 36, 37]. However, an epidemiological study done in South Africa has reported a female predominance[32]. This difference may be a result of a cross-sectional study design used, which can easily exclude the other gender in the reports.

Most of the patients supported by SF for OFC surgical treatments were under the age of ten years, with most surgeries performed before the age of five years. With lip repair, surgery is recommended at three months of age, palatal repair at about 11-12 months and alveolar bone graft at five to seven years [63, 106]. Further analysis is required to evaluate the age at which specific procedures facilitated are undertaken to verify the proportion undertaken within the appropriate age ranges in our study.

Annual variation in OFC procedures

The variation in the number of procedures facilitated by SF annually (ranging from 207 in 2016 to 27 in 2019), may be due to several reasons. The centres where SF facilitates CLP surgeries have sole discretion in decision making around the type and number of surgical procedures performed. The annual variation in SF facilitated surgeries may also be due to an emphasis by SF on other life-changing reconstructive surgical procedures for patients with other mandated conditions. It may also indicate that a large proportion of CLP patients did not receive the treatment required in that year, further contributing to the CLP surgical backlog. Further

investigations are required to analyse if more procedures were performed by the state the same year and to identify underlying reasons for the significant decline in the number of procedures performed in 2019 as compared to the previous years.

Incomplete OFC Data

This study found incomplete, or no data recorded for many patients facilitated by SF. The SF CEO indicated that this may be due to two reasons: 1) Patients do not always sign consent for SF to have their information used; 2) SF was not using a comprehensive data collection tool. The CEO acknowledged this shortfall and further suggested the use of the data collection tool used in this study for future baseline use by the organisation (Appendix 4). Shortages of funding, resources, and poor healthcare infrastructure, known challenges in Africa, may also account for incomplete OFC datasets (i.e., uncounted cases of CLP)[96-98].

Variation in procedures

Throughout the present study period (2015-2019), CP repair was the most common surgical procedure performed, followed by CL, and the least performed was a combination CLP. Since the birth prevalence of OFC varies between regions and racial groupings, it has also been reported elsewhere that the most frequently occurring type of cleft is CLP, followed by CP, and lastly CL[107, 108]. However, a South African study reported CP as the most commonly occurring, followed by CLP and lastly CL[32]. The reasons for the higher number of CP repair procedures as compared to CLP and CL, are not explained by the present study, which was limited to the surgeries undertaken by SF only. Further investigation is required to evaluate the ratio of types of CLP procedures nationally, i.e., of total SF facilitated procedures, state-funded surgeries and procedures undertaken in the private sector. Data collated by the organisation is limited to procedure data provided by the implementing centres and hospitals.

Provincial Coverage

This study found that the highest number of surgical procedures were performed in the WC (*n* = 240, 42.55%), despite the province accounting for only 10% of births[109]. This province

has been reported to have the second highest birth prevalence of CLP (1 per 1000 live births) in South Africa, which may be due in part to a high number of referrals from other provinces[32]. In the present study, the higher number of surgical repairs in the province may be attributed to the greater capacity provided through three provincial centres resulting in cross-border referrals from other provinces, compared with other provinces, e.g., FS, with only one centre, and less capacity. Further analysis is required to quantify what proportion SF facilitated procedures represent of the total CLP in the province during the same period.

Our findings showed that GP ranked second, in the number of CLP procedures performed per province. GP has the highest provincial population and number of births, accounting for 22% of the country's population, despite being the smallest geographically[109]. The lowest CLP birth prevalence rates have been reported in the province (0.5 per 1000 live births)[32].

KZN was the province where the least surgical procedures were facilitated by SF during the study period (n=1). This province has an estimated CLP birth prevalence rate of 0.2 per 1000 live births[32]. KZN is the second largest province in terms of population and annual births (21%) and is a geographically extensive province[109], and thus a higher number of procedures would be expected. It is likely that the majority of CLP surgical procedures are being supported directly by the provincial DOH or by another NGO, ST, at the Wentworth Centre in Durban, making it unnecessary for SF to enable as many CLP procedures in the province. This requires further confirmation and investigation to ensure that CLP patients are being treated in the province by ST and not remaining undiagnosed, untreated or unreferred in the province.

While LP is supported by SF, it serves only as a referral centre for GP centres, as indicated by the SF EOD. Specialist services at regional hospitals in rural provinces, such as LP (with 134,056 births per year)[109], are poor or unavailable, with patients referred to tertiary services in other provinces, such as those in GP, that are far away[110]. Due to the long traveling distance and cost of accessing health services, some patients from rural provinces may not access the referral facilities or the care centres[111]. Therefore, the low number of surgical procedures would be recorded in these rural provinces, and some patients may remain untreated, with lethal consequences.

The number of procedures facilitated by SF varied between the provinces across the timespan of the study. The WC, GP and ECP recorded the highest numbers of CLP surgical procedures facilitated in 2016, the FS in 2015, NCP s in 2017, and the only one KZN procedure was facilitated by SF in 2018. The present study did not assess the reasons for this variation but it may be related to the differences in availability of funds since SF relies on donor money to support patients[92]. Further investigation is needed to quantify the proportion of SF facilitated procedures at a provincial level across the five-year period of this study.

OFC Centres

With regards to the centres, the study found that the highest number of surgical procedures was performed in centre 2 (n = 138, 24.47%) in WC and the lowest number of surgical procedures were performed in centre 10 (n = 1, 0.18%) in KZN. As for the variation in procedures facilitated between provinces, the unstable NGO funding environment may also account for the diverse number of procedures facilitated in individual centres over the study period[92]. In addition, referral rates of CLP patients to the different centres may have varied, along with directly state-funded services (i.e., the proportion of CLP funded by provincial DOH), and SF may also have focused on specific centres during the study period. Inadequate surgical infrastructure in rural areas [93], lack of MDT members, and absence of standardised treatment protocols in certain centres have been reported in South Africa [51]. Further investigation is required to clarify the causes of this variation.

Study limitations

This study was focused on a single NGO (SF) operating in South Africa. There are other NGOs, mainly international, involved in supporting individuals born with CLP in the country which this study did not include. Therefore, the results of our study do not claim to represent total services rendered or facilitated by all NGOs in South Africa.

The interview took only 40 minutes on Zoom meeting. Although the participants (CEO and EOD) provided the PI with follow up supporting documents, the interview time was very limited to extract credible qualitative data.

The SF relies on the hospitals for patient data that are not recorded or collected by SF for the purpose of research. This accounts for incomplete data over the study period. Missing information includes patient race, associated anomalies, cleft laterality (incomplete), and type of surgeries performed (incomplete). Records missing these data prevented the analysis of these specific clinical components.

The outcomes of treatment, care-givers inputs and health care practitioner's experiences were not evaluated in our study. These are potential areas of future investigation to further supplement SF's evidence of impact in augmenting OFC care in South Africa.

Conclusions

This study provides an indication of the scale of support provided by SF in the ongoing treatment of individuals with OFC in South Africa, which to date has remained largely unquantified and relied on anecdotal reports. SF provides financial and material support, undertakes public awareness, advocacy, and other services for OFC care in South Africa. This study indicates that SF supports almost a third (30%) of South Africa's state OFC surgical services annually – and without this NGO, many of those born with OFC would not receive the prompt care they required, resulting in increased child mortality and morbidity. The need for greater investment in OFC services countrywide is highlighted, particularly in rural areas where service shortfalls of SF have been reported.

Recommendations

Further studies documenting and comparing the CLP services offered by South African NGOs are recommended. The findings of this study and similar future work will assist with demonstrating the evidence-based contribution and impact of CLP NGOs in the country. Specific recommendations from this study include:

- The data capturing system used by SF requires review to ensure more comprehensive data capturing to promote more complete data records. This would enable a more accurate representation of SF's contribution, which may currently be underreported due to incomplete records. Inclusion of race, associated anomalies, cleft laterality, different procedures, referral processes and distribution of surgery type are also suggested for future data capturing.
- To ensure the sustainability of these services in the future, greater government commitment to OFC services is recommended. Due to the unpredictability of the NGO funding base and the increased pressure due to competing health priorities (e.g., COVID-19 pandemic), sustainable and expanded government services in South Africa will be vital to ensure services do not fall short in the future in the event of inadequate SF funding.

References

1. CDC: **Facts about cleft lip and cleft palate** [https://www.cdc.gov/ncbddd/birthdefects/cleftlip.html] Access date: 02 December 2019
2. CDC: **Facts about cleft lip and cleft palate** [https://www.cdc.gov/ncbddd/birthdefects/cleftlip.html] Access date: 16 February 2021
3. CDC: **Cleft lip and palate|Birth defects surveillance toolkit** [https://www.cdc.gov/ncbddd/birthdefects/surveillancemanual/photo-atlas/cleft.html] Access date: 16 Fecruary 2021
4. Moore KL, Persaud TVN, Torchia MG: **The developing human-e-Book: Clinically oriented embryology**: Elsevier Health Sciences; 2018.
5. Bernheim N, Georges M, Malevez C, De Mey A, Mansbach A: **Embryology and epidemiology of cleft lip and palate**. *B ENT* 2006:11.
6. Zeiger JS, Beaty TH, Liang K-Y: **Oral clefts, maternal smoking, and TGFA: a meta-analysis of gene-environment interaction**. *The Cleft Palate Craniofacial Journal* 2005, **42**(1):58-63.
7. Kawalec A, Nelke K, Pawlas K, Gerber H: **Risk factors involved in orofacial cleft predisposition–review**. *Open Medicine* 2015, **10**(1):163-175.
8. Correa A, Gilboa SM, Besser LM, Botto LD, Moore CA, Hobbs CA, Cleves MA, Riehle-Colarusso TJ, Waller DK, Reece EA: **Diabetes mellitus and birth defects**. *American Journal of Obstetrics and Gynecology* 2008, **199**(3):237. e231-237. e239.
9. Margulis AV, Mitchell AA, Gilboa SM, Werler MM, Mittleman MA, Glynn RJ, Hernandez-Diaz S, Study NBDP: **Use of topiramate in pregnancy and risk of oral clefts**. *American Journal of Obstetrics and Gynecology* 2012, **207**(5):405. e401-405. e407.
10. Munger RG, Romitti PA, Daack-Hirsch S, Burns TL, Murray JC, Hanson J: **Maternal alcohol use and risk of orofacial cleft birth defects**. *Teratology* 1996, **54**(1):27-33.
11. Shaw GM, Nelson V, Carmichael SL, Lammer EJ, Finnell RH, Rosenquist TH: **Maternal periconceptional vitamins: interactions with selected factors and congenital anomalies?** *Epidemiology* 2002:625-630.
12. Pradat P, Robert-Gnansia E, Di Tanna GL, Rosano A, Lisi A, Mastroiacovo P, database acttM: **First trimester exposure to corticosteroids and oral clefts**. *Birth Defects Research Part A: Clinical and Molecular Teratology* 2003, **67**(12):968-970.
13. Diewert VM, Pratt RM: **Cortisone-induced cleft palate in A/J mice: Failure of palatal shelf contact**. *Teratology* 1981, **24**(2):149-162.
14. Schutte BC, Murray JC: **The many faces and factors of orofacial clefts**. *Human molecular genetics* 1999, **8**(10):1853-1859.
15. Stoll C, Alembik Y, Dott B, Roth M: **Associated malformations in cases with oral clefts**. *The Cleft Palate-Craniofacial Journal* 2000, **37**(1):41-47.
16. Kohli SS, Kohli VS: **A comprehensive review of the genetic basis of cleft lip and palate**. *Journal of Oral and Maxillofacial Pathology* 2012, **16**(1):64.
17. Christianson A, Howson CP, Modell B: **March of Dimes: Global report on birth defects, the hidden toll of dying and disabled children**. *March of Dimes* 2005.
18. Operation Smile: **Operation Smile** [https://www.operationsmile.org/] Access date: 07 February 2021
19. Panamonta V, Pradubwong S, Panamonta M, Chowchuen B: **Global birth prevalence of orofacial clefts: a systematic review**. *Journal of Medical Association Thailand* 2015, **98**(Suppl 7):11-21.
20. Mossey PA, Modell B: **Epidemiology of oral clefts 2012: An international perspective**. *Cleft Lip and Palate* 2012, **16**:1-18.
21. Tanaka SA, Mahabir RC, Jupiter DC, Menezes JM: **Updating the epidemiology of cleft lip with or without cleft palate**. *Plastic and Reconstructive Surgery* 2012, **129**(3):511-518.

22. Chung C, Myrianthopoulos N: **Racial and prenatal factors in major congenital malformations**. *American Journal of Human Genetics* 1968, **20**(1):44.
23. Erickson JD: **Racial variations in the incidence of congenital malformations**. *Annals of Human Genetics* 1976, **39**(3):315-320.
24. Eshete M, Butali A, Deressa W, Pagan-Rivera K, Hailu T, Abate F, Mohammed I, Demissie Y, Hailu A, Dawson DV: **Descriptive epidemiology of orofacial clefts in Ethiopia**. *The Journal of Craniofacial Surgery* 2017, **28**(2):334.
25. Khan A: **Congenital malformations in African neonates in Nairobi**. *The Journal of Tropical Medicine and Hygiene* 1965, **68**(11):272-274.
26. Butali A, Adeyemo W, Mossey P, Olasoji H, Onah I, Adebola A, Akintububo A, James O, Adeosun O, Ogunlewe M: **Prevalence of orofacial clefts in Nigeria**. *The Cleft Palate-Craniofacial Journal* 2014, **51**(3):320-325.
27. Kesande T, Muwazi LM, Bataringaya A, Rwenyonyi CM: **Prevalence, pattern and perceptions of cleft lip and cleft palate among children born in two hospitals in Kisoro district, Uganda**. *BMC Oral Health* 2014, **14**(1):1-7.
28. Mbuyi-Musanzayi S, Kayembe TJ, Kashal MK, Lukusa PT, Kalenga PM, Tshilombo FK, Devriendt K, Reychler H: **Non-syndromic cleft lip and/or cleft palate: Epidemiology and risk factors in Lubumbashi (DR Congo), a case-control study**. *Journal of Cranio-Maxillofacial Surgery* 2018, **46**(7):1051-1058.
29. Agbenorku P, Agbenorku M, Iddi A, Abude F, Sefenu R, Matondo P, Schneider W: **A study of cleft lip/palate in a community in the South East of Ghana**. *European Journal of Plastic Surgery* 2011, **34**(4):267-272.
30. Kromberg J, Jenkins T: **Common birth defects in South African blacks**. *South African Medical Journal* 1982, **62**(17):599-602.
31. Joint W, Organization WH: **Management of birth defects and haemoglobin disorders: report of a joint WHO-March of Dimes meeting, Geneva, Switzerland, 17-19 May 2006**. 2006.
32. Hlongwa P, Levin J, Rispel LC: **Epidemiology and clinical profile of individuals with cleft lip and palate utilising specialised academic treatment centres in South Africa**. *PLOS ONE* 2019, **14**(5):01-14.
33. National Department of Health: **National Guidelines for the Care and Prevention of the Most Common Genetic Disorders, Birth Defects and Disabilities**. In. Pretoria 2005.
34. Lebese V, Aldous C, Malherbe H: **South African congenital disorders data, 2006-2014**. *South African Medical Journal* 2016, **106**(10):992-995.
35. IPDTOC WG: **Prevalence at birth of cleft lip with or without cleft palate: data from the international perinatal database of typical oral clefts**. *The Cleft Palate-Craniofacial Journal* 2011, **48**(1):66-81.
36. FitzPatrick D, Raine P, Boorman J: **Facial clefts in the west of Scotland in the period 1980-1984: epidemiology and genetic diagnoses**. *Journal of Medical Genetics* 1994, **31**(2):126-129.
37. Carroll K, Mossey PA: **Anatomical variations in clefts of the lip with or without cleft palate**. *Plastic Surgery International* 2012:01-06.
38. Gregg T, Leonard A, Hayden C, Howard K, Coyle C: **Birth prevalence of cleft lip and palate in Northern Ireland (1981 to 2000)**. *The Cleft Palate-Craniofacial Journal* 2008, **45**(2):141-147.
39. Bellis T, Wohlgemuth B: **The incidence of cleft lip and palate deformities in the south-east of Scotland (1971-1990)**. *British Journal of Orthodontics* 1999, **26**(2):121-125.
40. Blanco-Davila F: **Incidence of cleft lip and palate in the northeast of Mexico: a 10-year study**. *Journal of Craniofacial Surgery* 2003, **14**(4):533-537.
41. Knox G, Braithwaite F: **Cleft lips and palates in Northumberland and Durham**. *Archives of Disease in Childhood* 1963, **38**(197):66.

42. Fadeyibi IO, Coker OA, Zacchariah MP, Fasawe A, Ademiluyi SA: **Psychosocial effects of cleft lip and palate on Nigerians: the Ikeja-Lagos experience**. *Journal of Plastic Surgery and Hand Surgery* 2012, **46**(1):13-18.
43. Hlongwa P, Rispel LC: **"People look and ask lots of questions": caregivers' perceptions of healthcare provision and support for children born with cleft lip and palate**. *BMC Public Health* 2018, **18**(1):01-10.
44. NM Stock NR: **Starting a Family: The Experience of Parents With Cleft Lip and/or Palate**. *The Cleft Palate–Craniofacial Journal* 2015, **Vol. 52 No. 4**:425–436.
45. Ngai CW, Martin WL, Tonks A, Wyldes MP, Kilby MD: **Are isolated facial cleft lip and palate associated with increased perinatal mortality? A cohort study from the West Midlands Region, 1995–1997**. *The Journal of Maternal-Fetal & Neonatal Medicine* 2005, **17**(3):203-206.
46. Kang S-L, Narayanan CS, Kelsall W: **Mortality among infants born with orofacial clefts in a single cleft network**. In.: SAGE Publications Sage CA: Los Angeles, CA; 2012.
47. Carlson L, Hatcher KW, Vander Burg R: **Elevated infant mortality rates among oral cleft and isolated oral cleft cases: a meta-analysis of studies from 1943 to 2010**. *The Cleft Palate-Craniofacial Journal* 2013, **50**(1):2-12.
48. Christensen K, Juel K, Herskind AM, Murray JC: **Long term follow up study of survival associated with cleft lip and palate at birth**. *British Medical Journal* 2004, **328**(7453):01-04.
49. Hubli EH, Noordhoff MS: **Smile Train: changing the world one smile at a time**. *Annals of Plastic Surgery* 2013, **71**(1):04-05.
50. Bermudez L, Carter V, Magee W, Sherman R, Ayala R: **Surgical outcomes auditing systems in humanitarian organizations**. *World Jurnal of Surgery* 2010, **34**(3):403-410.
51. Hlongwa P, Dandajena T, Rispel L: **Comparative analysis of healthcare provision to individuals with cleft lip and/or palate at specialised academic centres in South Africa**. *South African Medical Journal* 2019, **109**(6):426-430.
52. CDC: **Recommendations to improve preconception health and health care-United States** [https://www.cdc.gov/mmwr/preview/mmwrhtml/rr5506a1.htm] Access date: 16 April 2021
53. Han HH, Choi EJ, Kim JM, Shin JC, Rhie JW: **The importance of multidisciplinary management during prenatal care for cleft lip and palate**. *Archives of Plastic Surgery* 2016, **43**(2):153.
54. Kennedy T, Miller S, Tooze F, Harding R, Davis T, Graham 3rd W: **The team approach to treatment of the cleft lip and palate**. *American Family Physician* 1978, **18**(1):74-84.
55. Wellens W, Poorten V: **Keys to a successful cleft lip and palate team**. *B ENT* 2006:3.
56. Akinmoladun V, Obimakinde O, Okoje V: **Team approach to management of oro. facial cleft among African practitioners: A survey**. *Nigerian Journal of Clinical Practice* 2013, **16**(1):86-90.
57. Association ACP-C: **Parameters for evaluation and treatment of patients with cleft lip/palate or other craniofacial differences**. *The Cleft Palate-Caniofacial Journal* 2018, **55**(1):137-156.
58. Proffit WR, Fields HW, Larson B, Sarver DM: **Contemporary Orthodontics-e-book**. Philadelphia: Elsevier Health Sciences; 2018.
59. Abu-Hussein M, Watted N, Emodi O, Zere E: **Role of pediatric dentist-orthodontic In cleft lip and cleft palate patients**. *Journal of Dental and Medical Sciences* 2015, **1**:2.
60. Broen PA, Moller KT, Carlstrom J, Doyle SS, Devers M, Keenan KM: **Comparison of the hearing histories of children with and without cleft palate**. *The Cleft Palate-Craniofacial Journal* 1996, **33**(2):127-133.
61. Hayes D: **Hearing loss in infants with craniofacial anomalies**. *Otolaryngology Head and Neck Surgery* 1994, **110**(1):39-45.
62. Harper PS: **Practical genetic counselling** A Hodder Arnold Publication; 2004.

63. Kasten EF, Schmidt SP, Zickler CF, Berner E, Damian L, Christian G, Workman H, Freeman M, Farley MD, Hicks TL: **Team care of the patient with cleft lip and palate**. *Current Problems in Pediatric and Adolescent Health Care* 2008, **38**(5):138-158.
64. Freitas JAdS, Garib DG, Trindade-Suedam IK, Carvalho RM, Oliveira TM, Lauris RdCMC, Almeida ALPFd, Neves LTd, Yaedú RYF, Soares S: **Rehabilitative treatment of cleft lip and palate: experience of the Hospital for Rehabilitation of Craniofacial Anomalies-USP (HRAC-USP)-part 3: Oral and Maxillofacial Surgery**. *Journal of Applied Oral Science* 2012, **20**(6):673-679.
65. Jackson I, Vandervord J, McLennan J, Christie F, McGregor J: **Bone grafting of the secondary cleft lip and palate deformity**. *British Journal of Plastic Surgery* 1982, **35**(3):345-353.
66. Pai BCJDDS, M.S.; Hung, Yi-Tan M.S.; Wang, Ruby S. H. C.C.C.-S.L.P.; Lo, Lun-Jou M.D.: **Outcome of patients with complete unilateral cleft lip and palate: 20-Year follow-up of a treatment protocol**. *Journal of the American Society of Plastic Surgeons* 2019, **143**(2):359-367.
67. Graber LW, Vanarsdall Jr, Robert L: **Orthodontics: Current principles and techniques-e-book**. Philadelphia: Elsevier; 2011.
68. Lewis CW, Jacob LS, Lehmann CU: **The primary care pediatrician and the care of children with cleft lip and/or cleft palate**. *Pediatrics* 2017, **139**(5).
69. McCarthy J: **Plastic surgery** 1st Edition. Philadelphia: WB Saunders; 1990.
70. Bernstein NR, Kapp K: **Adolescents with cleft palate: body-image and psychosocial problems**. *Psychosomatics* 1981, **22**(8):697-703.
71. Dhakshaini M, Pushpavathi M, Garhnayak M, Dhal A: **Prosthodontic management in conjunction with speech therapy in cleft lip and palate: A review and case report**. *Journal of International Oral health* 2015, **7**(Suppl 2):106.
72. Bender PL: **Genetics of cleft lip and palate**. *Journal of Pediatric Nursing* 2000, **15**(4):242-249.
73. Kummer AW: **Speech therapy for errors secondary to cleft palate and velopharyngeal dysfunction**. In: *Seminars in Speech and Language: 2011*; 2011: 191.
74. Alighieri C, Bettens K, Bruneel L, Sseremba D, Musasizi D, Ojok I, Van Lierde K: **Comparison of motor-phonetic versus phonetic-phonological speech therapy approaches in patients with a cleft (lip and) palate: a study in Uganda**. *International Journal of Pediatric Otorhinolaryngology* 2020, **131**:109849.
75. Vyas T, Gupta P, Kumar S, Gupta R, Gupta T, Singh HP: **Cleft of lip and palate: A review**. *Journal of Family Medicine and Primary Care* 2020, **9**(6):2621.
76. Habel A, Sell D, Mars M: **Management of cleft lip and palate**. *Archives of Disease in Childhood* 1996, **74**(4):360-366.
77. Kaufman FL: **Managing the cleft lip and palate patient**. *Pediatric Clinics of North America* 1991, **38**(5):1127-1147.
78. Smile Train: **Smile Train | Cleft lip and palate children's charity** [www.smiletrain.org] Access date: 06 February 2021
79. Association ACP-C: **American Cleft Palate-Craniofacial Association** [https://acpa-cpf.org/] Access date: 07 February 2021
80. Alliance for Smiles: **Alliance for smiles** [https://www.allianceforsmiles.org/] Access date: 07 February2 021
81. Faces T: **Transforming Faces** [https://transformingfaces.org/] Access date: 07 February 2021
82. Harar P: **Project Harar** [https://www.projectharar.org/] Access date: 07 February 2021
83. Foundation S: **Funding proposal submitted by smile foundation**. In. Edited by Foundation S: Smile Foundation; 2019.
84. Angela S. Volk M, Matthew J. Davis, BSa, Priya Desai, MPHb,, Larry H. Hollier Jr M: **The history and mission of Smile Train, a global cleftC harity**. *Oral and Maxillofacial Surgery Clinics of North America* 2020, **32**:481-488.

85. Smile Train: **Smile Train celebrated Its 100,000th cleft surgery milestone in Africa** [https://www.smiletrain.org] Access date: 08 February 2021
86. Train S: **Why Yona has six babies named for her** [https://www.smiletrain.org/stories/why-yona-has-six-babies-named-her] Access date: 14022021
87. South African Government: **Nonprofit Organisations Act 71 of 1997** [https://www.gov.za/documents/nonprofit-organisations-act] Access date: 05 September 2021
88. Foundation S: **Smile Foundation overview**. In.: Smile Foundation 2018.
89. South African Government: **Companies Act of 2008**
90. South African Government: **Income Tax Act 58 of 1962**.
91. Foundation S: **What we do** [https://smilefoundationsa.org/about-us/] Access date: 14 February 2021
92. Casey KM: **The global impact of surgical volunteerism**. *Surgical Clinics of North America* 2007, **87**(4):949-960.
93. Martiniuk AL, Manouchehrian M, Negin JA, Zwi AB: **Brain gains: A literature review of medical missions to low and middle-income countries**. *BMC Health Services Research* 2012, **12**(1):1-8.
94. Louis M, Dickey RM, Hollier Jr LH: **Smile Train: Making the grade in global cleft care**. *Craniomaxillofacial Trauma & Reconstruction* 2018, **11**(1):001-005.
95. Shrime MG, Sleemi A, Ravilla TD: **Charitable platforms in global surgery: A systematic review of their effectiveness, cost-effectiveness, sustainability, and role training**. *World Journal of Surgery* 2015, **39**(1):10-20.
96. Conway JC, Taub PJ, Kling R, Oberoi K, Doucette J, Jabs EW: **Transforming Faces**. *BMC Pediatrics* 2015, **15**(1):01-09.
97. Morrison G, Cronje A, Van Vuuren I, Op't Hof J: **The incidence of cleft lip and palate in the Western Cape**. 1985.
98. Adekeye E, Lavery K: **Cleft lip and palate in Nigerian children and adults: a comparative study**. *British Journal of Oral and Maxillofacial Surgery* 1985, **23**(6):398-403.
99. Wilson J, Hodges A: **Cleft lip and palate surgery carried out by one team in Uganda: Where have all the palates gone?** *The Cleft Palate-Craniofacial Journal* 2012, **49**(3):299-304.
100. Reeve M-E, Groce NE, Persing JA, Magge SN: **An international surgical exchange program for children with cleft lip/cleft palate in Manaus, Brazil: Patient and family expectations of outcome**. *Journal of Craniofacial Surgery* 2004, **15**(1):170-174.
101. Stock NM, Guest E, Stoneman K, Ridley M, Evans C, LeRoy C, Anwar H, McCarthy G, Cunniffe C, Rumsey N: **The contribution of a charitable organization to regional cleft lip and palate services in England and Scotland**. *The Cleft Palate-Craniofacial Journal* 2020, **57**(1):14-20.
102. Smile Foundation: **Hospitals Smile works with Smile Foundation** [https://smilefoundationsa.org/hospitals-smile-works-with/] Access date: 12 December 2019
103. Butali A, Pagán-Rivera K, Dawson DV, Munger R, Eshete MA, Adeyemo WL, Mossey PA: **Descriptive epidemiology of orofacial clefts in Africa using data from 46,502 Smile Train surgeries**. 2017.
104. Statistics SA: **General household survey 2015** [https://www.statssa.gov.za/publications/P0318/P03182015.pdf] Access date: 30 March 2021
105. Shaw WC, Semb G, Nelson P, Brattström V, Mølsted K, Prahl-Andersen B, Gundlach KK: **The Eurocleft project 1996–2000: Overview**. *Journal of Cranio-Maxillofacial Surgery* 2001, **29**(3):131-140.
106. de Ladeira PRS, Alonso N: **Protocols in cleft lip and palate treatment: systematic review**. *Plastic Surgery International* 2012, **2012**.

107. Nagase Y, Natsume N, Kato T, Hayakawa T: **Epidemiological analysis of cleft lip and/or palate by cleft pattern**. *Journal of Maxillofacial and Oral Surgery* 2010, **9**(4):389-395.
108. Yılmaz HN, Özbilen EÖ, Üstün T: **The prevalence of cleft lip and palate patients: a single-center experience for 17 years**. *Turkish Journal of Orthodontics* 2019, **32**(3):139.
109. Statistics South Africa: **Statistical Release P0305: Recorded live births. Pretoria** [http://www.statssa.gov.za/publications/P0305/P03052019.pdf] Access date: 28 April 2021
110. Gaede B, Versteeg M: **The state of the right to health in rural South Africa**. *South African Health Review* 2011, **2011**(1):99-106.
111. Schneider H: **Phase 1 results access challenges in TB, ART and maternal health services**. In: *Johannesburg: Researching Equity in Access to Health Care Project (REACH).* 2009.

Appendix 1 - Data Collection Questionnaire

STATEMENT OF CONSENT

I have been given an information sheet and I understand the objectives of the study.

I further understand that my

responses will be kept confidential and that it is up me whether or not to complete this questionnaire.

It has been explained

to me that even if I choose not to complete this questionnaire, I should still return the questionnaire to the researchers and

indicate No in the space below.

My refusal to participate will in no way prejudice me.

I agree voluntarily to complete the questionnaire **(please tick).** ☐ **Yes**

☐ **No**

IF YOU AGREE TO PARTICIPATE, PLEASE ANSWER ALL OF THE FOLLOWING QUESTIONS.

SECTION 1: Organisational background and areas of service

1.1	What is the main reason for your existence in South Africa?		
1.2	What type of business is your organisation?		
1.3	Do you have an outline copy of your organisational structure?	If yes, would you please provide us with the outline of your organisational structure?	
1.4	What is the Vision of the organization?		
1.5	What is the Mission of the organization?		
1.6	When did you start operating in South Africa?		
1.7	In which Provinces are you providing services?	Centres:	How many times do you visit these centres per year?
	☐ Eastern Cape		
	☐ Free State		
	☐ Gauteng		

	☐ Kwazulu Natal		
	☐ Limpopo		
	☐ Mpumalanga		
	☐ Northern Cape		
	☐ North West		
	☐ Western Cape		

SECTION 2: Care and management

2.1	Which point of care do you provide for individuals with CLP?	☐ Plastic surgery ☐ Paediatric Surgery ☐ Dentistry ☐ Maxillo-facial & oral surgery ☐ Orthodontics ☐ Speech Therapy ☐ Other, please specify ----------
2.2	Which of the following services do you provide in all the centres (Tick all that apply)	
a) Feeding plate		☐ No ☐ Yes
b) Speech Therapy		☐ No ☐ Yes
c) Genetic counselling		☐ No ☐ Yes
d) Lip and palate repair		☐ No ☐ Yes
e) General dental treatment		☐ No ☐ Yes
f) Orthodontic treatment		☐ No ☐ Yes
g) Orthognathic treatment		☐ No ☐ Yes
h) Other please specify ----------		☐ No ☐ Yes

2.3	Is there a specific treatment protocol for CLP?	☐ No ☐ Yes
If yes, please could we have a copy of the protocol?		☐ No ☐ Yes
2.4	Is there a copy for internal referrals?	☐ No ☐ Yes
If yes, please could we have a copy of the protocol?		☐ No ☐ Yes
2.5	How could you describe care provision at the centre?	☐ Individual specialist ☐ Team approach ☐ Hybrid approach ☐ Other (please specify) --- --- ---
2.6	Which of the following groups are members of the team at the centre? Tick all that apply	
a) Geneticist/Genetic Counsellor		☐ No ☐ Yes
b) Plastic Surgeon		☐ No ☐ Yes
c) Orthodontist		☐ No ☐ Yes
d) ENT Surgeon		☐ No ☐ Yes
e) Maxillo-facial Surgeon		☐ No ☐ Yes
f) Paediatric Dentist		☐ No ☐ Yes
j) Other, please specify ---		☐ No ☐ Yes

THANK YOU FOR PARTICIPATING

Appendix 2 - Participants Information Sheet

Assessment of services rendered by a non-governmental organization on the management of cleft lip and palate in South Africa

Introduction

Good day. My name is Mashudu Muthavhine. I am conducting a study on cleft lip and palate (CLP) in order to determine the services rendered by a non-governmental organization on the management of cleft lip and palate in South Africa. The study is being done for a postgraduate degree.

Why are we doing the study?

The purpose of the study is to obtain information on management of CLP from the CEO of the Smile Foundation. We hope that the information obtained from the study will be used to assist us to improve the provision of CLP treatment services in the country.

How will we do the study?

The study will obtain information of all records of OFC surgeries facilitated by Smile Foundation in South Africa during a five-year period, from 1 January 2015 to 31 December 2019. The researcher would like to ask the CEO questions regarding their organisational background and services rendered for CLP. It should take around 40 minutes to answer these questions. A structured, pre-tested record review form will be used to obtain demographic, clinical and treatment information of OFC.

How do I know that the information I give will be kept confidential?

The information that you provide to me will be kept confidential and will be used for the purpose of this study only. Everything that you say will be treated as private and confidential. I will complete the questionnaire with the answers that you give. The answers given by you will be looked at and the combined information will be written-up in a form of a report.

Did you get permission to carry out the study?

This research is still in the Protocol development stage. Permission to carry out this project will be obtained from the University of the Witwatersrand Research Ethics Committee once it has been approved by the university.

Will there be any benefits from participating?

There will be no direct benefits to participate in the interviews, as participation is voluntary. This means you have the right to refuse to answer the questionnaire or if you do agree to answer the questionnaire you can leave out the answers you are uncomfortable with and you can stop the interview at any time.

Will there be any harm from participating?

We don't foresee any risks to you taking part in the research apart from taking up your time. If you feel upset during the interview, we can stop the interview at any time. It is not a problem if you don't want to be interviewed. However, we would really appreciate it if you do share your thoughts and feelings about the questions we will be asking you. We hope that the information we will get from you will be used to help us to improve treatment services for individuals with CLP.

Who do I contact if I want to ask more questions?

We will be happy to answer any questions you have about this study. If you have any questions about your rights as a study participant or questions or concerns about any aspect of the study, you may contact the Chairperson of the Ethics Committee, Professor Clement Penny, on (011) 717 1234. If you have questions about the research, you may also contact the principal investigator: Mashudu Muthavhine on 084 602 6227 or the supervisor: Professor Phumzile Hlongwa on 072 664 4664.

Thank you for your assistance with this study.

Appendix 3 - Informed Consent Form

I,…….

of ……..
(NGO)

Hereby confirm that I have read the Participant Information Sheet, and that I do agree to be interviewed for the research project described there. I understand that my participation is voluntary, that I may refuse to answer specific questions, or withdraw at any time if I so wish, and that my views will remain confidential.

Signature:

Appendix 4 - Orofacial Clinical information

Province: _____

	Centre:		2015	2016	2017	2018	2019
1	Number of OFC individuals who received treatment per centre per year						
2	Gender	Males					
		Females					
3	Race	Black					
		White					
		Coloured					
		Indian					
4	Average age at time of treatment						
5	Number of associated anomalies						
6	Cleft anatomical location	Cleft lip					
		Cleft lip and palate					
		Cleft palate					
		Midline					
		Other, please specify ----------					
7	Cleft laterality	Unilateral: Left					
		Unilateral: Right					
		Bilateral					
		Palate only					
		Midline					

		Other, please specify ----------					
8	Type of surgeries performed	Primary lip/ nose Unilateral					
		Primary lip/ nose Bilateral					
		Lip/ nose revision					
		Primary cleft palate					
		Secondary cleft palate					
		Fistula repair					
		Alveolar bone graft					
		Other, please specify ----------					

Appendix 5 – Ethics Clearance Certificate

UNIVERSITY OF THE WITWATERSRAND JOHANNESBURG

R14/49 Dr M Muthavhine

HUMAN RESEARCH ETHICS COMMITTEE (MEDICAL)
CLEARANCE CERTIFICATE NO. M200909

NAME: (Principal Investigator)	Dr M Muthavhine
DEPARTMENT:	School of Oral Health Sciences Department of Orthodontics Dental School University
PROJECT TITLE:	Assessment of services rendered by a non-governmental organization on the management of cleft lip and palate in South Africa
DATE CONSIDERED:	2 October 2020
DECISION:	Approved unconditionally
CONDITIONS:	
SUPERVISOR:	Profs P Hlongwa & S Nemutandani & Dr H Malherbe
APPROVED BY:	Dr CB Penny, Chairperson, HREC (Medical)
DATE OF APPROVAL:	20 November 2020

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

DECLARATION OF INVESTIGATORS

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

Principal Investigator Signature

24/11/2020
Date

Appendix 6 – Permission letter from Smile Foundation

09 November 2020

Department: Orthodontics
Protocol Ref No: M200909

Dear: Dr Mashudu Muthavhine

The Smile Foundation would be privileged to partner with you and your department to showcase the impact that the NGO sector is making.

This letter serves to confirm that permission is granted for the above study.

We look forward to engaging further.

Kind regards,



Hedley Lewis
CEO

Appendix 7 - Permission from Smile Foundation

To: Hedley Lewis
(The Chief Executive Officer)
Smile Foundation

From: Dr Mashudu Lucky Muthavhine
(Investigator)
Wits Oral Health Centre

Re: Permission to do research at Smile Foundation

TITLE OF STUDY: Assessment of services rendered by a non-governmental organization on the management of individuals with cleft lip and palate in South Africa

This request is lodged with you in terms of the requirements of the Promotion of Access to Information Act. No. 2 of 2000, Section 32 of the constitution.

I am a researcher/ MSc student at the University of Witwatersrand Oral Health Centre. My supervisor is Prof P Hlongwa ,and co-supervisors Prof M.S. Nemutandani and

Dr H Malherbe.

I herewith request permission on behalf of all of us to conduct a study on the above topic in your organisation. This study involves access to patient records.

The researchers request access to the following information: Clinical files records and data bases.

We intend to publish the findings of the study in a professional journal and/ or to present them at professional meetings like symposia, congresses, or other meetings of such a nature.

We intend to protect the personal identity of the patients by assigning each individual a random code number.

We undertake not to proceed with the study until we have received approval from the Faculty of Health Sciences Research Ethics Committee, University of Witwatersrand.

Yours sincerely

10/12/2020

Signature of the Principal investigator

Appendix 8 – Turn-it in Report

2294141:Turnitin_content2.docx

ORIGINALITY REPORT

13%	8%	8%	4%
SIMILARITY INDEX	INTERNET SOURCES	PUBLICATIONS	STUDENT PAPERS

PRIMARY SOURCES

1	www.samj.org.za Internet Source	2%
2	Phumzile Hlongwa, Jonathan Levin, Laetitia C. Rispel. "Epidemiology and clinical profile of individuals with cleft lip and palate utilising specialised academic treatment centres in South Africa", PLOS ONE, 2019 Publication	1%
3	www.smiletrain.org Internet Source	1%
4	Kasten, E.F.. "Team Care of the Patient with Cleft Lip and Palate", Current Problems in Pediatric and Adolescent Health Care, 200805 Publication	1%
5	www.operationsmile.org Internet Source	1%
6	Eric H. Hubli. "Smile Train : Changing the World One Smile at a Time", Annals of Plastic Surgery, 01/2012 Publication	1%

7	Idowu O Fadeyibi, Olurotimi A Coker, Matthiew P Zacchariah, Adedolapo Fasawe, Samuel A Ademiluyi. "Psychosocial effects of cleft lip and palate on Nigerians: The Ikeja-Lagos experience", Journal of Plastic Surgery and Hand Surgery, 2012 Publication	<1 %
8	www.shout-africa.com Internet Source	<1 %
9	Bender, P.L.. "Genetics of cleft lip and palate", Journal of Pediatric Nursing, 200008 Publication	<1 %
10	Submitted to Varsity College Student Paper	<1 %
11	hdl.handle.net Internet Source	<1 %
12	Mitchell, J.C.. "Management of cleft lip and palate in primary care", Journal of Pediatric Health Care, 200001/02 Publication	<1 %
13	Angela S. Volk, Matthew J. Davis, Priya Desai, Larry H. Hollier. "The History and Mission of Smile Train, a Global Cleft Charity", Oral and Maxillofacial Surgery Clinics of North America, 2020 Publication	<1 %

14	www.cdc.gov Internet Source	<1 %
15	José Alberto de Souza Freitas, Daniela Gamba Garib, Marchini Oliveira, Rita de Cássia Moura Carvalho Lauris et al. "Rehabilitative treatment of cleft lip and palate: experience of the Hospital for Rehabilitation of Craniofacial Anomalies - USP (HRAC-USP) - Part 2: Pediatric Dentistry and Orthodontics", Journal of Applied Oral Science, 2012 Publication	<1 %
16	Peter A Mossey, Julian Little, Ron G Munger, Mike J Dixon, William C Shaw. "Cleft lip and palate", The Lancet, 2009 Publication	<1 %
17	pdfs.semanticscholar.org Internet Source	<1 %
18	bmcpublichealth.biomedcentral.com Internet Source	<1 %
19	Tim Goodacre, Marc C. Swan. "Cleft lip and palate: current management", Paediatrics and Child Health, 2012 Publication	<1 %
20	www.ajol.info Internet Source	<1 %
21	Submitted to Flinders University Student Paper	

		<1%
22	mafiadoc.com Internet Source	<1%
23	www.coursehero.com Internet Source	<1%
24	worldwidescience.org Internet Source	<1%
25	Submitted to University of Debrecen Student Paper	<1%
26	Houriiyah Tegally, Eduan Wilkinson, Marta Giovanetti, Arash Iranzadeh et al. "Detection of a SARS-CoV-2 variant of concern in South Africa", Nature, 2021 Publication	<1%
27	Submitted to Imperial College of Science, Technology and Medicine Student Paper	<1%
28	Joost Engel, Elien Mahler, Lucien Anteunis, Edmond Marres, Gerhard Zielhuis. "Why are NICU infants at risk for chronic otitis media with effusion?", International Journal of Pediatric Otorhinolaryngology, 2001 Publication	<1%
29	Submitted to Ocean County College Student Paper	<1%

30	P. S. Fry. "Success, failure, and self-assessment ratings.", Journal of Consulting and Clinical Psychology, 1976 Publication	<1 %
31	Submitted to St Peter's College Student Paper	<1 %
32	www.cavalierhealth.org Internet Source	<1 %
33	acpa-cpf.org Internet Source	<1 %
34	insights.ovid.com Internet Source	<1 %
35	Stephanie J. Schrag, Sara Zywicki, Monica M. Farley, Arthur L. Reingold et al. "Group B Streptococcal Disease in the Era of Intrapartum Antibiotic Prophylaxis", New England Journal of Medicine, 2000 Publication	<1 %
36	arcabc.ca Internet Source	<1 %
37	pt.scribd.com Internet Source	<1 %
38	K A Jellinger. "Unusual presentation of a primary spinal lymphoma", Journal of Neurology, Neurosurgery & Psychiatry, 2002 Publication	<1 %

39	Qiang Liu, Ming-Liang Yang, Zeng-Jian Li, Xiao-Feng Bai, Xu-Kai Wang, Li Lu, Yu-Xin Wang. "A Simple and Precise Classification for Cleft Lip and Palate: A Five-Digit Numerical Recording System", The Cleft Palate-Craniofacial Journal, 2017 Publication	<1 %
40	d-scholarship.pitt.edu Internet Source	<1 %
41	open.library.ubc.ca Internet Source	<1 %
42	www.slideshare.net Internet Source	<1 %
43	Helen A Thomason. "Craniofacial Defects and Cleft Lip and Palate", Encyclopedia of Life Sciences, 03/15/2009 Publication	<1 %
44	Michael J Dixon. "Orofacial Clefting", Encyclopedia of Life Sciences, 01/27/2006 Publication	<1 %
45	P Hlongwa, T C Dandajena, L C Rispel. "Comparative analysis of healthcare provision to individuals with cleft lip and/or palate at specialised academic centres in South Africa", South African Medical Journal, 2019 Publication	<1 %

46	V Lebese, C Aldous, H L Malherbe. "South African congenital disorders data, 2006 - 2014", South African Medical Journal, 2016 Publication	<1%

Exclude quotes	On	Exclude matches	Off
Exclude bibliography	On		