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Comparative analysis of healthcare provision to individuals with cleft lip and/or palate at specialised academic centres in South Africa

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Background. Cleft lip and/or palate (CLP) is the most common congenital anomaly of the craniofacial complex, with an estimated worldwide prevalence of 1/500 - 700 live births. Affected children require immediate medical treatment and prolonged management by a multidisciplinary team of health professionals.
Objectives. To describe and compare healthcare provision to individuals with CLP at specialised care centres in South Africa (SA).
Methods. The study was conducted at all CLP care centres in 6 of SA's 9 provinces that provide specialised treatment and care to individuals with CLP. At each centre, the team leader was interviewed using a semi-structured questionnaire that focused on the point-of-care entry for CLP patients; type of services provided; whether treatment protocols were used, which treatment protocols were used and internal referral systems; and members of the healthcare team. Stata 13 (StataCorp., USA) was used to analyse the data.
Results. Eleven CLP team leaders participated in the study, of whom 5 were from Gauteng Province. The point-of-care for CLP patients in the majority of centres was plastic surgery (*n*=9/11; 81.8%). The majority of centres (*n*=10/11; 90.9%) followed similar treatment protocols and only 1 centre performed lip surgery at 12 - 18 months. Although all centres reported a multidisciplinary team approach for CLP care provision, there were gaps in the health professions categories, which influenced the type of treatment provided. Hence, surgical repair of the lip and palate (*n*=10/11; 90.9%) and speech therapy (*n*=7/11; 63.6%) dominated the type of treatment provided, and patients were referred to other provinces or to the private health sector for other types of treatment.
Conclusions. The gaps in services at the CLP care centres in SA need to be addressed to ensure integrated, holistic care provision.

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Cleft lip and/or palate (CLP) is the most common congenital anomaly of the craniofacial complex, with an estimated worldwide prevalence of 1/500 - 700 live births.[1] In South Africa (SA), CLP is among the 5 most common birth defects reported,[2] with an estimated prevalence of 0.1 - 0.4/1 000 live births.[2-4]

Affected children present with a number of medical problems and potential complications that include feeding difficulties, hearing loss, speech problems, disfigured appearance and dental malformation.[5-8] Hence, individuals with CLP require co-ordinated and specialised treatment offered by a multidisciplinary team,[9-11] which includes geneticists, maxillofacial surgeons, otolaryngologists, orthodontists, paediatricians, plastic surgeons, paediatric dentists, psychologists, professional nurses, social workers and speech and language therapists.[12-14] These teams are available in high-income countries (HICs).

Various clinical protocols have been proposed for the treatment of CLP.[15] Many HICs have adopted national protocols and guidelines and have centralised services and/or established centres for the management and follow-up of affected individuals.[16-19] The advantages of centralisation and/or concentration of specialised services include the following: co-ordination of treatment, standardised data collection, availability of a critical mass of experts, clinical audit and review to enhance quality of care, responsiveness to patient and family needs, and ongoing monitoring and evaluation.[12,20-26]

Globally, it is estimated that only 20% of individuals with CLP have access to comprehensive treatment that involves a team co-ordinated approach.[27] The treatment and care gaps include: lack or shortages of healthcare professionals, lack of infrastructure and delays in surgical repair of the clefts.[27,28] These gaps are most pronounced in low- and middle-income countries (LMICs) in Africa, Asia and South America.[28,29] In many LMICs, outreach programmes by non-governmental organisations (NGOs) have assisted in improving the services for individuals with CLP.[29-34] These NGOs, together with support groups that include parents of the affected children, have played an important role in the ongoing management of CLP patients. However, the majority of LMICs, including SA, lag behind in terms of national treatment protocols, standardised data collection, access to care by a multidisciplinary team, and evaluation of treatment outcomes.[12,18,35]

Notwithstanding improvements in the treatment of CLP individuals in the preceding decades,[36-38] there is still no universal protocol for repair and ongoing clinical management. However, treatment modalities in the management of CLP are often based on chronological age and dentofacial development.[38] The common elements of clinical standards and sequence of treatment of CLP are shown in Table 1.[38] At birth, genetic counselling is given to parents, and feeding of the baby is evaluated. Depending on the size of the cleft, feeding plates are recommended within the first week after birth to assist with feeding. Presurgical infant orthopaedic treatment for approximation of the segment is also performed prior to cleft lip repair. As the child grows, several follow-up consultations for management of the cleft are done until treatment is completed during adulthood.[12]

Table 1. Treatment sequence in the management of cleft lip and/or palate

Chronological age/dentofacial development	Cleft lip and/or palate treatment
At birth	Genetic counselling
	Feeding plate
	Presurgical infant orthopaedics
	Psychosocial 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	Orthodontic treatment/orthognathic surgery/maxillary advancement
	Psychology counselling
19 - 20 years	Prosthodontic replacement of missing teeth
	Nose revision

Adapted from De Ladeira and Alonso.[38]

There is a dearth of studies on the management of CLP in SA, except for a 1953 study performed in a Johannesburg private hospital, which found that the clinical management of patients with CLP was uncoordinated.[39] The aim of our study was to compare the treatment and care of individuals with CLP at all the CLP care centres in SA. This is part of a larger doctoral study on the epidemiology and care of individuals with CLP in SA.

Methods

This cross-sectional study was conducted during 2015 and 2016 in all the 11 specialised, academic CLP care centres in 6 of SA's 9 provinces.

Confidentiality was maintained throughout the study. The same unique identification number was allocated to each centre and CLP team leader to ensure anonymity. The data containing unique numbers and centre information were kept on a password-protected computer.

The CLP team leaders were interviewed using a semi-structured questionnaire that contained 29 questions divided into 3 parts, i.e. centre information, treatment and management of CLP and team members. The questions focused on the entry point of care for CLP individuals, type of services and treatment provided at each centre, treatment protocols followed in the clinical management of individuals with CLP, continuum of care, internal referral systems, as well as healthcare professionals who were members of the team. The interviews were complemented with a record review of all individuals with CLP who visited the academic centres from 1 January 2013 to 31 December 2014.

At each centre, the principal investigator conducted the interviews in a private room. Each interview lasted an average of 30 minutes. The responses of participants were written down verbatim in the space provided in the questionnaire, typed out and saved as individual Microsoft Word documents. The data were imported into Stata 13 (StataCorp., USA) for descriptive analysis.

Ethical approval

The Human Research Ethics Committee (medical) of the University of the Witwatersrand, Johannesburg, provided ethical approval (ref. no. M150536). Approval was also obtained from the CLP care centres and permission was obtained from the relevant healthcare authorities. We adhered to standard ethical procedures, which included study information sheets, voluntary participation, informed consent, anonymity of responses and confidentiality. The participants' consent was in written form.

Results

A 100% response rate was obtained and all 11 team leaders from CLP care centres participated in the study: 5 in Gauteng Province, 2 in Western Cape Province and 1 in each of the provinces of the Eastern Cape, Free State, KwaZulu-Natal and Limpopo. Most of the centres have been in existence for >5 years. The descriptive characteristics of the centres are shown in Table 2.

One centre in Gauteng had no records for the 2-year review period. The number of individuals with CLP at the 10 centres totalled 699, ranging from 16 to 144. The team leaders reported that individuals with CLP were treated primarily by plastic surgeons. Most centres reported a team approach for the provision of CLP treatment.

The treatment provided by each centre is shown in Table 3. The dominant modes of treatment were surgical repair and speech therapy. Other treatment modalities were less common. Although centre 9 is listed as a CLP specialised centre, it does not provide any active treatment but serves as an NGO booking and follow-up centre for surgical repairs.

Six centres provided their treatment protocols for analysis (Table 4). The protocols showed that 2 centres used feeding plates and presurgical orthopaedic treatment. All 6 centres followed a set protocol for timing of the surgical procedure for the lip and palate. The sequencing of lip surgery was similar in 5 centres. Palate surgery and alveolar bone graft were performed at similar times in all 6 centres. While centres 4 and 5 reported availability of orthodontic treatment, age of treatment was not indicated. Similarly, orthognathic treatment was performed at 4 centres and speech therapy was offered by all 6 centres; however, no specific age of delivery was indicated. Other treatments were not provided according to the standard protocol (Table 1), were not provided when needed and were not available in most centres.

Table 5 shows the categories of healthcare professionals (HCP) at each CLP care centre. In most centres, plastic surgeons (91%) and speech therapists (72.7%) formed part of the CLP team. Other HCPs, such as ear, nose and throat surgeons, paediatric dentists, paediatric surgeons, psychologists and social workers, were not available in most of the centres. Although professional nurses were present in 7 centres (63.6%), in only 2 centres were these nurses actively involved with CLP treatment; they played primarily administrative roles in the other 5 centres. Only 2 centres reported that parents' support groups were incorporated in CLP management.

Table 2. Descriptive characteristics of the cleft lip and/or palate centres

Characteristics	Sample size, *n* (%)
Cleft lip and/or palate team leader (*N*=11)	
Plastic surgery	9 (81.8)
Maxillofacial surgery	1 (9.1)
Orthodontics	1 (9.1)
Type of care provided (*N*=11)	
Individual specialist	1 (9.1)
Team approach	6 (54.6)
Hybrid approach	3 (27.3)
Other	1 (9.1)
Cleft lip and/or palate per academic centre (*n*=699)	
Centre 2	93 (13.3)
Centre 3	33 (4.7)
Centre 4	52 (7.4)
Centre 5	141 (20.2)
Centre 6	79 (11.3)
Centre 7	70 (10.0)
Centre 8	62 (8.9)
Centre 9	16 (2.3)
Centre 10	122 (17.5)
Centre 11	31 (4.4)

Discussion

This is one of the first comparative studies to analyse treatment and care of individuals with CLP in the SA public sector. Our study found variations across the 11 care centres in the number of CLP patients treated, type of treatment provided, availability of written protocols and composition of healthcare teams.

The number of individuals with CLP treated during the 2-year period of the review ranged from 16 to 141. Centre 9 did not meet the criteria for a specialised centre, as it only provided booking and follow-up services to CLP individuals. Experts have pointed to the importance of sufficient case-loads to ensure competent clinical care and to secure adequate resources for comprehensive care.[40] A 2001 review in the UK led to a government directive to provide care from a single regional centre, with a comprehensive specialist team and a guideline that 1 surgeon should be responsible for 40 - 50 new patients requiring primary surgery per year.[18,41] Similarly, cleft care has also been centralised in Brazil[42,43] and in the Scandinavian countries of Denmark, Finland, Norway and Sweden.[21] However, the concept of regionalisation or concentration of CLP services is under-developed in SA, exacerbated by the relatively low priority of congenital anomalies within the overall context of the country's quadruple disease burden.

The current study found that the dominant modalities of treatment were surgical repair of the lip and palate (*n*=10/11; 90.9% of centres)

Table 3. Types of treatment for cleft lip and/or palate provided by each academic centre (*N*=11)

	Treatment									
Centre	FP	PSO	ST	GC	LPR	GDT	ORT	OGT	ABG	ENT
1	No	No	Yes	Yes	Yes	No	No	No	No	No
2	No	No	Yes	No	Yes	No	No	No	Yes	No
3	No	No	No	No	Yes	No	No	No	No	No
4	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes	Yes	No
5	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	No
6	No	No	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes
7	No	No	Yes	Yes	Yes	Yes	Yes	No	No	Yes
8	No	No	Yes	Yes	Yes	Yes	Yes	Yes	Yes	No
9	No	No	No	No	No	No	No	No	No	No
10	No	No	Yes	Yes	Yes	No	No	Yes	No	No
11	No	No	No	No	Yes	No	No	No	No	No
Total, *n*	2	2	8	6	10	4	5	5	5	2

FP = feeding plate; PSO = presurgical orthopaedics; ST = speech therapy; GC = genetic counselling; LPR = lip and palate repair; GDT = general dental treatment; ORT = orthodontic treatment; OGT = orthognathic treatment; ABG = alveolar bone graft; ENT = ear, nose and throat treatment; No = treatment not offered; Yes = treatment offered.

Table 4. Comparison of cleft lip and/or palate treatment protocols across 6 centres*

Centre	Feeding plate	Presurgical orthopaedics	Lip repair, months	Palate repair, months	Alveolar bone graft, years
2	No	No	3	6 - 12	6 - 8
4	Birth	Birth	3 - 6	12 - 18	8 - 10
5	Birth	Birth	12 - 18	6 - 12	8 - 10
6	No	No	3	6 - 12	8 - 10
7	No	No	3	12 - 18	8 - 10
8	No	No	3	9	7 - 11

*These centres provided written protocols for analysis.

Table 5. Members of cleft lip and/or palate team in each academic centre

	Team members											
Centre	Gen	PS	Orth	ENT	MFS	PD	Paed	Psych	PN	SW	ST	PSG
1	1	1	0	0	0	0	0	0	1	1	1	1
2	0	1	0	0	1	0	0	0	1	1	1	0
3	0	1	0	0	0	0	0	0	1	0	0	0
4	0	1	1	0	1	1	0	0	0	0	1	0
5	1	0	1	0	1	1	0	0	1	0	1	1
6	1	1	1	1	1	0	0	0	1	0	1	0
7	1	1	1	1	0	1	0	1	0	0	1	0
8	1	1	1	0	1	1	1	0	1	0	1	0
9	0	1	0	0	0	0	0	0	1	0	0	0
10	1	1	0	0	0	0	0	1	0	0	1	0
11	0	1	0	0	0	0	1	0	0	0	0	0
Total, *n* (%)	6 (54.5)	10 (91)	5 (45.5)	2 (18.2)	5 (45.5)	4 (36.4)	2 (18.1)	2 (18.2)	7 (63.6%)	2 (18.2)	8 (72.7)	2 (18.2)

Gen = geneticist; PS = plastic surgeon; Orth = orthodontist; ENT = ear, nose and throat surgeon; MFS = maxillofacial surgeon; PD = paediatric dentist; Paed = paediatric surgeon; Psych = psychologist; PN = professional nurse; SW = social worker; ST = speech therapist; PSG = parent support group; 1 = member of team; 0 = not member of team.

and speech therapy (*n*=7/11; 63.6%). Two of the centres that did not provide speech therapy were located in the country's rural provinces, and 1 centre (centre 9) did not provide any active treatment for CLP. This underscores the inequities in access to specialised services in the SA public health sector.[44] Speech therapy is an essential service for CLP. This gap would need to be addressed, either through efforts to recruit speech therapists to these centres, formal referral to and/or partnerships with urban CLP centres, or public/private partnerships.

This study found that feeding plates were underutilised, as 2 centres (*n*=2/11; 18.2%) reported their use, while the other 9 centres (*n*=9/11; 81.8%) did not use them for CLP patients. Notwithstanding the controversies regarding the use of feeding plates,[45-47] feeding difficulties following the birth of babies with CLP are common and the feeding plate can assist with closure of the cleft to facilitate sucking and to prevent choking. A study among caregivers of CLP children found that feeding difficulties were one of the most distressing aspects in caring for the children,[48] highlighting the importance of adequate support and assistance to caregivers regarding feeding methods.

The comparison of treatment protocols was only possible for 6 of the 11 centres. Five of the centres did not have a written protocol, which means that care might not be standardised in these centres, which could influence the quality of care provided. CLP care centres should be encouraged to develop standard treatment protocols. The findings of this study could be used to develop standard treatment guidelines, with the involvement of all 11 CLP centres, drawing on the experiences of the centres with protocols and international best practice.[21,42,43]

The timing for lip repair was 3 - 6 months in most centres. However, for our sample, the median age at consultation was ~3 months, with a high interquartile range of 3 weeks - 13 months; i.e. a sizeable portion of these children presented after 6 months, when ideally their first operation should have been done. This delayed age of consultation has also been reported in other LMICs.[49-51] Lip surgery provides comfort to the parents of CLP children, as they are able to show their children in public and avoid stigmatisation. Strategies need to be developed to ensure that the majority of children with CLP have access to lip surgery within the first 6 months of life. This will assist with the reduction of possible physical impairment and societal courtesy stigma and discrimination.[48,52]

There was variation in the number of team members at CLP centres, ranging from 2 to 8. In most centres, there was a scarcity of certain specialists. There are existing guidelines on health professionals who should constitute members of CLP teams.[12] Although the SA context would need to be taken into account, these guidelines should inform the constitution of the multidisciplinary team. Our study found that professional nurses were underutilised as members of the team. In other settings, professional nurses play an important role, especially in assisting mothers with feeding.[12,53,54]

The current study revealed that only 2 centres reported incorporating CLP parent support groups in their management. Other studies found that parents' involvement and participation in care decisions are important in CLP care outcomes, as their knowledge and support enhance compliance with treatment, retention and reduction of complications.[48,55,56]

Study limitations

Although our study generated new knowledge on the types of treatment available at all public sector CLP centres, it was largely descriptive. We did not evaluate the following: types of surgery used for treatment, quality of care, outcomes of treatment, cost of treatment and follow-up treatment provided to CLP individuals. These are study limitations, but point to areas for further research. Nonetheless, the knowledge generated by this study can be used to develop standard treatment guidelines for individuals with CLP, enhance the discourse on appropriate treatment modalities and contribute to the development of collaborative partnerships across the 11 centres.

Conclusions

Although the treatment of children with CLP and other types of craniofacial anomalies in SA has been ongoing for >6 decades,[39] comprehensive care is still lacking. The study has shown that there are gaps in the types of treatment provided across the 11 centres, that not all members of the multidisciplinary team are present, and that standardised treatment protocols are absent. These gaps are more pronounced in centres located in the rural provinces of SA.

Recommendations

The study findings underscore the need for the National Department of Health (NDoH) to review and update the national policy on congenital anomalies, which has not been revised since 2005. The NDoH should also ensure equitable access to appropriate treatment and care for individuals with CLP in SA, regardless of geographical

location. This might be achieved through regionalisation of centres and developing a standard treatment protocol for CLP, in consultation and with the involvement of health professionals at these 11 centres. Such regionalisation will ensure the existence of a critical mass of health professionals at each specialist facility, foster collaboration, enhance the education and training of healthcare providers on congenital anomalies, enable sharing of experiences and mutual learning, optimise resource utilisation and facilitate research on treatment models and outcomes. Ultimately, these initiatives will contribute to quality-of-care improvements for all individuals with CLP.

Appendix 1. The dataset supporting the conclusions of this article is not currently available, as it is part of PH's PhD thesis. The questionnaire used to collect the data is available from the corresponding author on request.

Declaration. Article based on a study done by PH in partial fulfilment of her PhD thesis.

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Author contributions. LCR is the primary supervisor of the PhD and TCD the co-supervisor. PH and LCR conceptualised and designed the PhD study. PH collected all the data, PH and LCR analysed the data. All authors contributed to writing and editing the manuscript, and read and approved the final version.

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