Survey of Management of Children With Cleft Lip and Palate in Teaching and Specialist Hospitals in Nigeria

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Objective: A national survey was conducted to obtain an overall view of the current management of children with cleft lip and/or palate in Nigeria.

Method: Questionnaires were sent to 44 identified cleft surgeons in all teaching and specialist hospitals in Nigeria.

Results: A total of 38 respondents returned completed questionnaires. The findings are as follows: (1) a majority of the surgeons (68.4%) are "low-volume operators," undertaking 10 or fewer new cleft repairs annually; (2) 86.8% of the surgeons repair cleft lip at 3 to 4 months of age, and the most common (71%) unilateral cleft lip repair method is the rotation-advancement technique; (3) 50% of the surgeons use straight line repair for bilateral cleft lip; (4) a majority (79%) of the respondents close the soft and hard palates as a single procedure; 47.3% of respondents use the von Langenbeck technique, 21.1% use the double-opposing Z-plasty and 21.1%, the palatal pushback; (5) in the management of protruding premaxilla, 52.6% of the respondents choose adhesive tape; (6) procedures such as alveolar bone grafting, rhinoplasty, and surgical treatment for velopharyngeal incompetence are rarely done as part of cleft management; and (7) the interdisciplinary team approach is practiced by 21% of respondents.

Conclusions: Issues are raised regarding the current organization of cleft services. We hope the findings of this study will provide preliminary information needed for the eventual establishment of standard cleft management for children with cleft lip and palate deformity in Nigeria.

KEY WORDS: cleft lip and palate, hospitals, management, Nigeria

Cleft lip and/or palate (CL/P) is the most common congenital craniofacial malformation and affects between 1 per 500 to 1 per 700 live births in Europe (World Health Organization, 1998). Genetic studies on human samples have demonstrated that CL/P has a heterogeneous genetic background, and environmental factors also contribute to this malformation (Marazita et al., 1986; Wyszynski et al., 1996). For a neonate with CL/P, appropriate corrective surgery in early childhood is necessary to improve function and appearance. Subsequent impairment of facial and dental development, speech, and hearing are common and may be accompanied by socio-psychological maladjustment (Strauss, 1998). A large number of specialist services are often necessary if the child's potential is to be maximized: neonatal

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nursing services, surgery, speech, hearing, dentistry, otolaryngology, psychology and genetic counseling (Strauss and ACPA Team Standard Committee, 1998).

Nigeria is the largest of the West African coastal nations, and its population of 88,992,220 (February 1991 census) (Adekunle and Otolorinto, 2000) is divided into six regions for administrative purposes (Fig. 1) (Adekunle and Otolorinto, 2000). Attempts have been made to determine the incidence of oral clefts in Nigeria. In a hospital-based study, Iregbulem (1982) reported an incidence of 1 in every 2703 live births in Enugu, South-East Nigeria. Approaches to cleft care vary considerably among countries (Asher-McDade and Shaw, 1990; Brennan et al., 2001; Lee et al., 2003; Weinfeld et al., 2005). Little, however, is known about the provision of care for children with CL/P in most countries in sub-Saharan Africa.

To our knowledge, there has not been a previous national survey undertaken in a country in sub-Saharan Africa to assess the management of children with CL/P. The purpose of this survey was to establish a database of current cleft management of children in Nigeria.

Метнор

A preliminary registry of cleft care providers in Nigerian teaching and specialist hospitals was compiled from a



FIGURE 1 Map showing the six geographic regions of Nigeria.

number of sources, including the Nigeria cleft association list, local workshop attendance lists, and individuals identified by a literature search. A total of 44 surgeons repairing clefts were identified, and a questionnaire (Appendix) was sent to each of the surgeons asking for details of cleft care in their hospitals. The surgeons were asked about general information concerning patient caseload, the clinical specialities involved in cleft care, primary surgical protocol, techniques for cleft lip and palate repair, and use of a multidisciplinary team in cleft management. It is possible that one or two cleft surgeons working in very remote areas of the country were unintentionally left out of the survey.

The Medical and Ethics Committee of the institution approved the study, and informed consent was obtained.

RESULTS

Regional Distribution of Cleft Services

Of the 44 questionnaires sent, 38 respondents returned fully completed questionnaires, two respondents returned questionnaires with incomplete information to justify exclusion from the survey, and four questionnaires were not returned, representing a response rate of 86%. The respondents covered the six geographic regions of Nigeria: 13 in South-West, six in South-East, five in South-South, six in

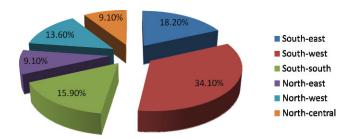


FIGURE 2 Regional distribution of cleft surgeons.

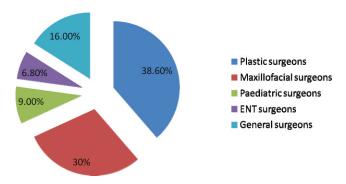


FIGURE 3 Percentage distribution of specialists involved in cleft surgery.

North-West, four in North-East and four in North-Central (Fig. 2). A majority (39.5%) of these respondents were plastic surgeons, followed by oral and maxillofacial surgeons (29%) (Fig. 3). Other surgical specialities identified as undertaking cleft surgery include general surgeons (15.7%), pediatric surgeons (10.5%), and ear, nose, and throat surgeons (5.3%).

High-Volume and Low-Volume Operators

The majority of surgeons were "low-volume operators," undertaking 10 or fewer new cleft lip repairs annually (Table 1). Only nine (23.7%) surgeons undertook more than 30 new cleft lip repairs annually. All the high-volume operators were either plastic surgeons or oral and maxillofacial surgeons, and all had passed the national professional fellowship examination in their respective subspecialty.

Timing of Surgery and Treatment Protocols

Lip repairs are usually done by 3 to 4 months of age by a majority (86.8%) of the respondents; although, five respondents wait until 6 to 9 months of age before repair. Ninety-two percent repair both sides of a bilateral cleft lip as a single procedure. A total of 32 surgeons (84.2%) repair the palate at approximately 18 months of age, and a majority (79%) close the soft and hard palates in the same surgical procedure. Only four surgeons refer children with cleft palate for ear, nose, and throat consultation. Undernourishment and difficulty in gaining weight in

TABLE 1 The Median Number of Cleft Lip and Palate Repairs Undertaken by High-, Medium- and Low-Volume Operators

		Type of Operator	
	High Volume (≥30 Lip Repairs a Year)	Medium Volume (<30 but >10 Lip Repairs a Year)	Low Volume (≤10 Lip Repairs a Year)
Median number			
of lip repairs	38	13	4.8
Median number			
of palate repairs	31	11.3	2.6
Total number			
of surgeons	9	3	26

TABLE 2 Unilateral Cleft Lip Management

Age of Definitive Repair	n (%)	
3–4 mo	33 (86.8)	
5–6 mo	4 (10.5)	
>6 mo	1 (2.7)	
Technique		
Rotation advancement	27 (71.1)	
Straight line	4 (10.5)	
Triangle flap	7 (18.4)	

children with CL/P were the most common reasons for delayed surgical correction of cleft lip or palate.

Surgical Techniques

The most common technique of unilateral cleft lip repair is the rotation-advancement technique of Millard, used by 71% of the surgeons (Table 2). The straight line repair is the least frequently used for unilateral cleft (10.5%). However, 50% of the surgeons use straight line repair for bilateral cleft lip. Only four surgeons occasionally use lip adhesions for bilateral cleft lip. Twenty-two (58%) of the 38 surgeons routinely repaired the vermilion border in cleft lip surgery, and only four routinely correct associated nasal deformity as part of cleft lip surgery (Table 3).

The most common technique of cleft palate repair most respondents reported (47.3%) is the von Langenbeck, followed by double-opposing Z-plasty (21.1%) and palatal pushback (21.1%). Only four surgeons reported using the vomer flap procedure (Table 4). No obvious relationship was demonstrable between surgeons' subspecialty and the choice of technique for lip and palate repair.

Alveolar Cleft Repair

Only five (13%) of the respondents stated that their cleft care includes alveolar cleft repair. The gingivoperiosteoplasty method and bone-graft method for alveolar cleft repair were reported by three and two of these respondents, respectively.

Management of Protruding Premaxilla

The protruding premaxilla is being managed as follows: adhesive tape by 52.6% of the respondents, head cap by

TABLE 3 Bilateral Cleft Lip Management

Age of Definitive Repair	n (%)	
3–4 mo	28 (73.7)	
5–6 mo	7 (18.4)	
>6 mo	3 (7.9)	
Technique		
Rotation advancement	12 (31.6)	
Straight line	19 (50)	
Triangle flap	7 (18.4)	

TABLE 4 Cleft Palate Management

Stage	n (%)	
Single-stage method	30 (78.9)	
Two-stage method	8 (21.1)	
Technique		
von Langenbeck	18 (47.3)	
Double-opposing Z-plasty	8 (21.1)	
Palatal pushback	8 (21.1)	
Vomer flap	4 (10.5)	

21.1%, lip adhesions by 10.5%, presurgical orthopedics by 10.5%, and the presurgical nasal alveolar molding devices by 5.3%.

Cleft Team and Ancillary Staff

Of the respondents, 30 (79%) work in isolation, and only eight (21%) practice the team approach to management. Of the eight documented cleft teams, only four have an orthodontist available as part of the team, and none had a speech pathologist or a speech therapist. Only two teams have separate, designated record keeping of cleft cases.

Velopharyngeal Incompetence

In the absence of speech pathologists, eight surgeons indicated that they manage velopharyngeal incompetence (VPI). Diagnosis of VPI was based on the existence of a continued speech defect after palatal repair. Pharyngeal flaps were used by all eight surgeons to address VPI.

DISCUSSION

With an 86% response rate and the inclusion of cleft surgeons from all six regions of the country, the results obtained from this study provide valuable insight into the current status of cleft management of children with cleft deformity in Nigeria in particular and sub-Saharan Africa in general. Other features provide clues as to how cleft management might be improved in this part of the world.

A wide spectrum of surgical conditions affects African children. The majority of patients that present for management in the hospital, however, fall into three major diagnostic categories: injuries, congenital anomalies, and surgical infections (Bickler and Sanno-Duanda, 2000). Compared with recent reports from Nordic countries and the U.K., where cleft services tend to be centralized (Shaw et al., 2001), the present report shows that cleft management in Nigeria is being provided via large numbers of local hospitals. Our findings show that a majority of surgeons work in isolation and are low-volume operators, undertaking 10 or fewer new cleft lip repairs annually (Williams et al., 1996). Only nine (23.7%) surgeons reported undertaking more than 30 new cleft lip repairs annually. In six instances in the present survey, three surgeons were found

to operate clefts independently at the same hospital. There has been increasing evidence from multicenter studies that decentralized care, especially by low-volume operators, may be associated with inferior outcomes (Shaw et al., 1992; Shaw et al., 2001). The consequences of this are greater hardship for patients and families and possible increased health costs due to additional operations and ancillary care necessary to improve treatment outcome (Shaw et al., 2001).

There is currently a large variation in the timing of cleft surgery and the surgical techniques that are used (Osborn et al., 1983; Lee et al., 2003; Weinfeld et al., 2005). In the present report, a majority of children with cleft lip undergo repairs between 10 and 12 weeks of age, possibly due to safety and the presence of adequate lip tissue associated with this later intervention (Lee and Kim, 2003). This suggests that most of the cleft surgeons in Nigeria are following the Wilhelmsen and Musgrave (1966) "rule of ten." In the present report, the rotation-advancement method was found to be the most common method for cleft lip repair, applied more frequently than evidenced in the reports from Korea (Lee and Kim, 2003) and the U.K. (Asher-McDade and Shaw, 1990). However, contrary to the findings in most recent studies (Lee et al., 2003; Weinfeld et al., 2005), straight line repair was found to be the method of choice by most surgeons for bilateral cleft lip.

An overwhelming majority of the surgeons in the present report perform one-stage palatal repair. This observation represents a possible difference from the findings in the Eurocleft report, which shows that approximate 50% of patients undergo one-stage soft and hard palate repair at various times, either in isolation or in combination with lip procedures (Shaw et al., 2001). With the absence of a wellestablished health insurance system in most countries in the developing world, the financial implications of care for patients may often influence the type of health services provided; a cleft care protocol that might bring about a cost reduction in service provision may, therefore, be more attractive in such environment. Another notable finding of the present study is the use of the von Langenbeck simple closure technique as the most frequent procedure for palatal closure.

The present survey reflects a significant use of adhesive tape among the respondents for protruding maxilla. Only 5.3% make use of nasoalveolar molding devices compared with 48.3% reported in European countries (Asher-McDade and Shaw, 1990; Shaw et al., 2001; Weinfeld et al., 2005). Shortage of appropriate professionals to fabricate nasoalveolar molding devices and the inability of parents to afford the cost of this appliance were provided by surgeons as the frequent reasons for using simple adhesive tape for protruding maxilla. Health problems such as undernourishment, anemia, and failure to thrive, which can be directly attributed to feeding difficulties in the patients, are the most common challenges encountered during preoperative evaluations and were responsible for

most surgical delays. Cleft care professionals in developing countries, in most instances, are not empowered with enough information to give education on lactation and other feeding techniques to parents with children with CL/P (Olasoji et al., 2005). Specific educational programs on feeding interventions for parents and cleft care providers could improve nutritional intake in these children and could move the schedule for surgical procedures forward. The concept of secondary surgical procedures such as alveolar bone grafting, rhinoplasty, surgery for correction of VPI, and orthognathic surgery have not really taken root in Nigeria, as observed in this survey. This probably is due to limited personnel and resources for such secondary procedures.

It has been recognized for some time that the preferred form of delivery of care for children with CL/P is by interdisciplinary cleft team (Strauss, 1999). Our survey showed that only eight (21%) of the surgeons performing cleft procedures run joint multidisciplinary clinics. The national shortage of specialists such as orthodontists, otorhinolaryngologists, and speech professionals was given by a majority of the respondents as the main reason for not running cleft clinics (Fig. 3). The need to address the acute shortage of essential staffing for ancillary care of children with clefts through specific training programs becomes more imperative in this environment.

The need for a major reorganization of cleft services in Nigeria is not in question; however, such reorganization must take into account the geographic accessibility of services to patients, considering the low socioeconomic status of a majority of the populace. Cleft services are best provided in a well-equipped setting, with a team of fully trained, multidisciplinary professionals practicing on a regular basis (Pannbacker et al., 1992; Strauss, 1999). One of the key findings in the present survey is that, with the exception of the North-Central region of Nigeria, each region had at least one high-volume operator repairing clefts. The geographic division of the country into regions could possibly facilitate the establishment of cleft centers and associations among specialists within and between regions. It might, therefore, seem sensible to consider the establishment of regional tertiary cleft centers in the areas of these high-volume operators so that expertise and resources can be concentrated and put to optimal use. However, what constitutes an ideal minimum caseload for primary cleft repairs per surgeon is still being debated; the American Cleft Palate-Craniofacial Association (1993) stated that the major factor in surgical outcome is the skill, training, and experience of the surgeon. The centers producing the best results, when comparing facial growth in the Eurocleft study (Shaw et al., 1992), had the fewest number of surgeons operating.

Strategies for improving cleft care in African children, we believe, should include geographically accessible regional cleft centers, and the African government must assume the central role in this respect.

CONCLUSION

The need for reorganization of cleft services in Nigeria is not in question; success is rooted in the geographic accessibility of services to patients, staff development, and the optimal use of limited resources and practitioners. Government support, partnership, and collaboration with established cleft centers in the Western world are key factors.

We hope that the findings in this study will provide preliminary information for the eventual establishment of standard management for children born with CL/P in Nigeria and other countries in sub-Saharan Africa in the years ahead.

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APPENDIX CLEFT MANAGEMENT QUESTIONNAIRE

1.	What is the name of the hospital at which you are practising?
2.	Please circle the region of Nigeria where the hospital is located: South-West South-East South-South North-East North-West North-Central
3.	Please circle your professional subspecialty: Ear, Nose, and Throat surgery General surgery Maxillofacial surgery Paediatric surgery Plastic surgery Others (specify)
4.	What is your highest degree? Please circle as appropriate: Bachelor's Master's Doctorate D.D.S MBBS FWACS/FMCDS Others (specify)
5.	Do you have a Cleft Team in your hospital?

6. Which of the following professionals are involved in cleft management in your hospital? Please circle as many as applicable:

Dentist
Nutritionist
Orthodontist
Speech therapist/pathologist

	Surgeon Others (specify)	12.	Do you routinely repair the vermilion during lip surgery? Yes No
7.	Do you keep separate designated record for cleft cases? Yes No	13.	Please circle your preferred method in the repair of bilateral cleft lip:
8.	Please indicate the average number of new cases of these procedures you do each year:		Both sides done together as a single procedure Each side done as a separate procedure
	Unilateral cleft lip repairBilateral cleft lip repair	14.	Please circle your preferred method in the repair of cleft palate:
	Cleft palate repair		Hard and soft palates are done as single procedure Hard and soft palates are done as separate procedure
	Midface secondary surgery	15.	Do you routinely refer children with cleft palate for ear examination?YesNo
9.	Please indicate at what age of patient you routinely do the following procedures: Cleft lip repair	16.	Please circle the preferred method for treating protruding premaxilla:
	Cleft palate repair		Adhesive tape Head cap
10.	Please circle the preferred technique for these procedures:		Lip adhesion
	a. <u>Unilateral cleft lip repair</u>		Nasal alveolar moulding device Presurgical orthopaedics
	Millard rotation advancement		Others (specify)
	Straight line	17	Places give reason for the chains of method in
	Triangular flap Others (specify)	1/.	Please give reason for the choice of method in Question 16:
	b. Bilateral cleft lip repair	10	Do you maytingly access nationt for valenhammacal
	Millard rotation advancement Triangular flap	10.	Do you routinely assess patient for velopharyngeal function?YesNo
	Straight line Others (specify)	19.	Please circle your preferred method of assessing velopharyngeal function (if applicable):
	c. Cleft palate repair		Voice and resonance evaluation
	von Langenbeck Palatal pushback		Nasoendoscopy Videofluoroscopy
	Double-opposing Z-plasty	20	Please circle your preference method of treating
	Vomer flap Others (specify)	20.	velopharyngeal incompetence (if applicable):
11			Speech therapy
11.	Do you routinely repair the nose during cleft lip surgery? Yes No		Speech appliances Surgery (specify type)
	Surgery105100		Surgery (speeny type)