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Pattern and Outcome of Cardiac Interventions Performed within and Outside Nigeria for Children with Structural Heart Diseases in Sokoto, North-western Nigeria

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Authors' contributions

This work was carried out in collaboration among all authors. Author UMS conceptualized and designed the study, wrote the protocol and wrote the first draft of the manuscript. Author UMW managed the analyses of the study and literature search. Author KOI assisted in data analysis and writing of the initial draft. Author BIG managed the literature searches and review of the manuscript. All authors read and approved the final manuscript.

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ABSTRACT

Background: Management of structural heart diseases is quite challenging in Nigeria, due to limited availability and access to cardiac care services. There is paucity of data on the pattern and outcome of cardiac interventions in children from the study area.
Aims: To determine the pattern of cardiac interventions and the early outcome (within six-months) of such interventions in children with structural heart diseases (SHD) seen in our hospital.
Study Design: Descriptive cross-sectional study.

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Place and Duration of Study: Department of Paediatrics, Usmanu Danfodiyo University Teaching Hospital (UDUTH), Sokoto, between August 2012 to September 2019.
Methodology: All children with SHDs seen at our hospital, but had Cardiac surgery or interventional cardiac procedures within and outside Nigeria were recruited. Relevant information including type of lesion, procedure performed and the outcome within the first six months and length of hospital stay was analyzed.
Results: Only 63 out of 780 (8.1%) children with structural heart diseases, aged 6 months-15 years (Mean age 6.5± 5.0 years, M: F = 1.3:1), had cardiac interventions. The interventions were surgical in 61 ((96.8%) and catheter-based in 2 (3.2%) patients; with 49 (77.8%) of these performed abroad. Patent Ductus arteriosus 14 (22.2%), Tetralogy of Fallot 14 (22.2%), ventricular septal defect 13 (20.6%) and atrial septal defect 6 (9.5%) were the commonest lesions for which interventions were undertaken. Early post-operative complications included pulmonary hypertensive crises, pericardial and pleural effusion, complete heart block and left branch pulmonary artery stenosis warranting re-intervention. During the first six months after the interventions, three (4.8%) patients died while 60 (95.2%) survivors were followed up. However, 25 (39.7%) of them were thereafter lost to follow.
Conclusion: Majority of the cardiac interventions were surgical, performed predominantly in older children with CHDs; and associated with good outcome. However, most of them were undertaken outside the country, highlighting the need to ensure these services are available and accessible locally.

Keywords: Structural heart diseases; cardiac intervention; outcome; children; Sokoto.

1. INTRODUCTION

Structural heart diseases (SHDs), comprising both congenital and acquired heart lesions, are associated with significant morbidity and mortality [1]. Though the prevalence of Congenital heart diseases (CHDs) at birth is generally similar across the globe (3-4 per 1000 live birth), more of the burden is seen in resource poor countries of Sub-Saharan Africa and other developing nations due to the high fertility rate that prevails [2,3]. Besides CHDs, countries in sub-Saharan Africa also suffer additional burden of preventable acquired heart diseases (AHDs), particularly rheumatic fever and Rheumatic heart diseases (RHD), which have been largely eradicated in developed nations [4,5]. Factors responsible for this include poor social and living conditions of the populace, late presentation, harmful sociocultural practices, misconceptions, and delay in making diagnosis due to lack of skill and facilities [4,6,7]. There is also limited capacity for carrying out lifesaving cardiac interventions in many developing nations, leading to a wide gap between population demand and access to these services [8,9].

The age of elective surgery for CHDs has declined steadily in developed countries, and surgeries are now performed as early as the neonatal period with less risk of adverse effects [10]. Such care is not readily available for children born with CHDs in developing countries [2]. The development of cardiac surgery in Nigeria has been very slow, since it was first carried out in 1974 in Enugu, South eastern part of the country [11]. Patients requiring intervention are often referred abroad in order to access such services while those who could not afford treatment faces a life of continuous suffering due to repeated ill health and hospitalization (1). A study by Ekure et al. [9[has shown that more than 80% of Nigerian children with CHD have no access to corrective surgery and only 20.5% of all the interventions were undertaken in the country. In the last decade, however, these cardiac interventions have become more frequent in the country through the support of non-governmental donor agencies (NGOs) and visiting foreign surgeons, who provide short term periodic cardiac missions [7,11].

Early intervention in children with structural heart diseases is associated with good surgical outcome and improved quality of life; while delayed diagnosis and intervention may be associated with increased perioperative morbidity and mortality [12,13]. Unlike developed nations, many children in middle- and low-income countries including the study area can only access the desired services or cardiac interventions at a relatively older age, due to the aforementioned challenges [14]. Since the outcome of interventions may potentially be different, it is necessary to audit the status of our patients who underwent surgery both within and outside Nigeria. This study was therefore carried out in Sokoto, North-western Nigeria to

determine the socio-demographic characteristics, pattern and early post-operative outcome of children with SHDs, who underwent surgical or catheter-based cardiac interventions.

2. MATERIALS AND METHODS

2.1 Study Area

This study was carried out at the Paediatric department of Usmanu Danfodiyo University Teaching Hospital (UDUTH), Sokoto. This is a tertiary health facility located in Sokoto State, which lies in the extreme Northwestern Nigeria. The hospital serves as a referral center for patients from Zamfara, Katsina, Kebbi and Niger states; and from neighboring countries of Niger and Benin Republics of West African sub-region. The Pediatric Cardiology unit runs a regular weekly outpatient clinic and echocardiography sessions. Patients with cardiac diseases requiring medical care are managed in the unit while those requiring cardiac surgical interventions are often referred abroad. Since 2009, our hospital signed an MoU with Madras Medical Mission (MMM), Chennai-India which facilitated the training of personnel and treatment of our patients at India. Patients diagnosed at our centre with structural heart diseases and requiring cardiac interventions, were referred to MMM for treatment unless otherwise decided by their parents/caregivers. After the interventions, the patients were usually referred back to our institution, with a copy of their case note, for continuation of post-operative care and follow up. Of recent, our hospital has witnessed its first open heart surgery anchored by our local team with the support of visiting foreign cardiac mission team. Our local team, who had training at MMM, is also set to perform such interventions independently in the near future.

2.2 Study Design

It was a descriptive cross-sectional study conducted between August 2012 and September 2019. Patients were recruited consecutively over the study period, as cardiac interventions were performed or as they presented to our facility after such interventions were already performed.

2.3 Study Subjects

The study subjects comprised children aged 15 years and below who had cardiac intervention, either cardiac/open heart surgery or interventional cardiac procedures, within or outside Nigeria. Included for the study were all patients with congenital or acquired heart diseases seen at our hospital and who had either palliative or definitive interventions. Patients who were referred for cardiac surgery and had cardiac catheterization, but declared inoperable due to raised pulmonary artery pressure were excluded. Also excluded were patients in whom records on the nature of intervention carried out was lacking or where information regarding patients' post-operative or post interventional procedure outcomes was not available.

Relevant demographic (age, gender, socio-economic class) and clinical information was documented for each recruited subject using a structured study proforma. Social class was class determined according to Oyedeji's method [15].

For patients who were being followed up at the clinic prior to intervention, preoperative cardiac evaluation was performed by chest x-ray, electrocardiography and transthoracic echocardiography. A 12-lead ECG was performed in all the patients prior to echocardiography while echocardiography was done using Sonoscape SSI 5000 echo-machine (Sonoscape Yizhe, Shenzen, China). Cardiac imaging was performed with the patients in left lateral position and through standard echocardiography widows (Apical, subcostal, parasternal long and short axes, and suprasternal notch views). Two-dimensional echo was used to evaluate structural abnormalities while flow direction and pressure gradients were assessed using Colour flow doppler and continuous wave (CW) doppler respectively. Ventricular function was measured by M mode.

Following diagnosis of cardiac lesions, parents/guardians were counselled on the need for intervention. Affected patients were continued on conservative management and follow up at our clinic, pending the time the parents could afford surgical or catheter-based interventions. All patients who were referred and had intervention abroad were re-evaluated at the clinic upon their return. Information on the type of procedure performed, outcome of the intervention, length of hospital stay, medications and complications were retrieved from the post-operative case records of the patients.

2.4 Outcome Measures

Outcome measures included post-operative mortality within six months of the procedure and mean duration of hospital stay which was calculated from date of admission to date of hospital discharge. Other outcome measures were post-operative complications and re-admission/reintervention after discharge.

2.5 Data Analysis

Data was entered into a Statistical Package for Social Sciences IBM® SPSS version 22.0 (SPSS Inc, USA). for cleaning and analysis. Quantitative data were expressed as means and standard deviation while categorical variables were summarized using frequencies and percentages. Chi-square or where necessary, Fisher's Exact test, was used to compare associations between categorical variables such as relationship between type of CHD (cyanotic or acyanotic) and PICU stay (prolonged or not prolonged). Student T test was also used to compare difference of means between continuous variable e.g mean preoperative weight compared with their mean weight at six months postoperatively. A P-value of <0.05 was considered statistically significant.

2.6 Ethical Consideration

The study was performed after obtaining informed consent from the parents plus assent for children above 7 years. Ethical approval was obtained from the Hospital Ethics and Research Committee of UDUTH, Sokoto.

3. RESULTS

3.1 Sociodemographic and Clinical Characteristics

The total number of children with structural heart diseases seen over the study period was 780, with 633 (81.2%) of them being CHDs and 147 (18.8%) being AHDs. Cardiac interventions (either surgical or catheter-based) were performed in only 63 (8.1%) patients out of the total children. These comprised 35 males (55.6%) and 28 females (44.4%), given a male to female ratio of 1.3:1. Their mean age was 6.5 ± 5.0 years (range = 6months-15 years). Majority 29 (46.0%) of the patients that had interventions belonged to middle socio-economic class (SEC), while the remaining 22 (34.9%) and 12 (19.1%) subjects were from the high and low SEC respectively (Table 1). More than half of the patients were self-sponsored and had their interventions at India whereas, only 14(22.2%) interventions were carried out locally, in our hospital (Table 1). The cost of cardiac interventions in India ranged between $6,000 and $15,000 USD (approximately 2.1-5.5million naira equivalent), excluding travel and other logistic expenses.

3.2 Pattern of Cardiac Interventions

Congenital heart diseases (CHDs) were the predominant lesions 62 (98.4%) for which interventions were carried out. Only one (1.6%) patient with AHD in form of rheumatic valvular heart disease (MR + severe AR) had cardiac intervention (Aortic valve replacement). Table 2 shows the distributions of the structural lesions (CHDs and AHD) for which interventions were performed, with PDA and TOF being the most common.

Open heart surgery (OHS) was performed in 61 (96.8%) patients while catheter-based procedures were performed in 2 (3.2%) patients (ASD device closure-1 PDA device closure -1). All the interventions performed in our centre (14 cases) were for simple left to right shunts [PDA - 10 independently by the local team, VSD -2 and ASD-2 performed in collaboration with a visiting cardiac team (Table 3).

3.3 Outcome of Cardiac Interventions

Three patients died (two in-hospital deaths, one post discharge) within the first six months of intervention, giving a mortality rate of 4.8%. The remaining 60 (95.2%) were followed up for the first six months after the intervention. The mean duration of Paediatric ICU (PICU) stay was 79.8 + 76.9hours (range =24-384 hours) while the mean duration of hospital stay was 15.1 + 7.1 days (range = 7-38days). A higher proportion of children with cyanotic CHD 45.6% (10/22) had prolonged PICU stay (>72hrs) than those with acyanotic CHD 30.0% (12/40), though the difference was not statistically significant (X^2 = 1.01, P = 0.39). In the same vein, 78.6% (11 of 14) of children with CHD and associated facial dysmorphism had prolonged PICU stay (> 72hrs) compared with 22.9% (11 of 40) of those who had no facial dysmorphism. The difference was statistically significant (X^2 = 5.32, p = 0.02).

Table 1. Socio-demographic characteristics of subject who had cardiac interventions

Variable	N (%)
Age group (years)	
0-5.0	40 (63.5)
>5-10.0	17 (26.9)
>10 -15.0	6 (9.5)
Gender	
Male	35 (59.5)
Female	28 (40.5)
Social class	
Upper	22 (34.9)
Middle	29 (46.0)
Lower	12 (19.1)
Source of sponsorship	
Self-sponsored	35 (55.6)
Solely by State government	21 (33.3)
Some support from government	6 (9.5)
Non-governmental organization(NGO)	1 (1.6)
Country of intervention	
India	47 (74.6)
Nigeria	14(22.2)
Ghana	1 (1.6)
Sudan	1 (1.6)
Types of SHD	
Acyanotic CHD	40 (63.5)
Cyanotic CHD	22 (34.9)
AHD (RHD)	1 (1.6)

Table 2. Distribution of structural cardiac lesions among the subjects

Type of lesion	N (%)
Patent Ductus Arteriosus (PDA)	14 (22.2)
Tetralogy of Fallot (TOF)	14 (22.2)
Ventricular Septal Defect (VSD)	13 (20.6)
Isolated Atrial Septal Defect (ASD)	6 (9.5)
Double outlet right ventricle	4 (6.3)
Atrioventricular septal defect (AVSD)	4 (6.3)
Truncus arteriosus	2 (3.2)
VSD+ PDA	1 (1.6)
VSD + ASD	1 (1.6)
Common atrium with left atrial isomerism	1 (1.6)
Partial anomalous pulmonary venous connection	1 (1.6)
Severe Congenital Aortic valve stenosis	1 (1.6)
Rheumatic Valvalur Heart Disease (*MR+AR)	1 (1.6)
Total	63 (100.0)

**MR=mitral regurgitation, AR-Aortic regurgitation*

Observed complications in the immediate postoperative period included pulmonary hypertensive crises, low cardiac output state, pericardial effusion, pleural effusion, Arrythmias (complete heart block, supraventricular tachycardia and junctional ectopic tachycardia) and left pulmonary artery branch stenosis warranting re-intervention (Table 4). Pleural and pericardial effusions were managed by drainage, antibiotics and anti-inflammatory (Dexamethasone and ASA) medications. Patient with low cardiac output state were managed with inotropes (dobutamine, dopamine, epinephrine and norepinephrine) while pulmonary artery hypertensive crises was managed with inhaled nitric oxide (NO), Bosentan and/or sildenafil. SVT was treated with adenosine while JET was treated with intravenous lidocaine.

Table 3. Pattern of cardiac interventions according to country performed

Type of Cardiac intervention	Country performed			
	India n (%)	Nigeria n (%)	Sudan n (%)	Ghana n (%)
Open Heart Surgery				
PDA ligation	3	10		-
TOF ICR repair (VSD closure + RVOT reconstruction)	14	-	-	
VSD patch closure	10	2	-	
ASD patch closure	3	2	-	-
DORV -Intraventricular repair (with use of Baffle)	4			
AVSD (intracardiac repair)	3			
Truncus repair	2			
VSD patch closure+ PDA ligation	1			
VSD patch closure + ASD patch closure	1			
Atrial septation for common atrium	-		1	
*Others	3	-	-	-
Palliative procedures:				
PA banding for large VSD	1		-	-
**Right modified BT shunt for TOF	-		-	1
Glenn shunt (for unbalanced AVSD)	1		-	-
Catheter-based interventions:				
Transcatheter ASD closure (Amplatzer septal occluder)	1		-	-
PDA device closure (Amplatzer duct occluder device)	1		-	-
Total	**48**	**14**	**1**	**1**

**Others: Interventions for other CHDs (partial anomalous pulmonary venous connection, severe congenital aortic valve stenosis) and rheumatic heart disease (aortic valve replacement).*
***The patient also had intracardiac repair for TOF later*

Table 4. Frequency of complications in children after cardiac interventions

*Observed complication	N (%)
Pericardial effusion	12 (19.5)
Low cardiac output state	11 (17.5)
Pleural effusion	7 (11.1)
Pulmonary artery hypertensive crises	7 (11.1)
Arrythmias	4 (6.3)
Infections	3(4.8)
Lung collapse	2(3.2)
Chylothorax	2 (3.2)
Left pulmonary artery branch stenosis	1 (1.9)

**Some patients have more than one complication at a time*

The three patients that died were children with Down syndrome with AV Canal defect, Truncus arteriosus and TOF. They died at 4 weeks, 6weeks and 8 weeks post-operative period respectively.

The patient with downs syndrome with AV canal defect developed complete heart block postoperatively. Though initially stable after discharge, he deteriorated during follow up and died at home, before pacemaker could be inserted.

The patient with Truncus arteriosus died at six-week post operatively in India following persistent pulmonary hypertensive crisis, low cardiac output state with inability to be weaned off ventilator; while the case of TOF was a an 8month old infant who initially had palliation with Blalock-Taussig (BT) shunt during neonatal period and then definitive intracardiac repair at six-months of life. He was discharged after prolonged ICU stay (>2weeks) at India, but developed intractable heart failure and systemic oxygen desaturation. Re-evaluation by cardiac catheterization and CT pulmonary angiogram revealed severe left pulmonary artery branch stenosis. He had reintervention at India, but died during the procedure (i.e. 8weeks after the definitive TOF repair).

During the follow up period, patients who survived showed statistically significant improvement in their weight from a mean weight of 10.9+6.8Kg pre-operatively to a mean weight of 13.8 +7.0Kg at six months after cardiac interventions (p=.0.001).

4. DISCUSSION

The present study shows that only small proportion (8.1%) of patients within the study area had cardiac interventions. This is a common finding in many developing countries, where access to cardiac surgery remains challenging [3,6,8,9]. In a study by Falase *et al* [7] in Lagos-Nigeria, only 3.4% of patients including adults with structural heart diseases were able to undergo cardiac surgeries. A more recent data from the National Paediatric Cardiac Registry revealed that less than 20% of children who required cardiac interventions actually received the necessary surgery [9]. The limited capacity for carrying out these interventions locally and the prohibitive cost of surgery abroad are some of the reasons responsible for this. In our setting, where healthcare financing is still largely dependent on out-of-pocket system of payment [11,16], it is even more difficult for many patients to undergo definitive treatment within or outside the shores of the country. Previous studies have noted these challenges as posing significant obstacles to the management of structural heart diseases in sub-Saharan Africa [1,4,6,11,17].

Not surprisingly, most of our patients who had interventions belonged to either the middle or high socioeconomic class (SEC). These were the affluent and the gainfully employed categories, who were able to source for funds, either from state governments or through self-sponsorship, in order to undergo treatment abroad. Except for one patient, there was no any support from NGO for our patients. This is in contrast to report from a study in south-south Nigeria [17] where full sponsorship for cardiac surgery abroad was provided by NGO, thus accounting for the higher proportion of their patients (37.5%) that accessed cardiac surgery. Ekure *et al* [9], in an earlier nation-wide survey, have shown that patients in the southern part of Nigeria are generally more likely to receive required cardiac intervention than their counterparts in the north. Thus, many children with structural heart diseases in our settings are managed conservatively pending the availability of surgery; or they resort to traditional/herbal treatment alternatives.

The capacity for carrying out complex interventions in our locale is still limited [14]. Less than a quarter of the interventions were performed at our institution, which was consistent with previous study showing that only 20.7% of all corrective cardiac interventions in Nigerian children were done in the country [9]. It is instructive that all the interventions undertaken at our centre were for simple lesions: PDA ligation independently performed by the local team and VSD/ ASD closure undertaken in collaboration with visiting cardiac team. While periodic cardiac mission does provide intermittent relief for a few patients and creates opportunity for some skill transfer [7], it may not adequately cater for the large volume of patients on the waiting list. Comprehensive strategy that will ensure availability of such services locally, and at regular and sustainable basis, is therefore needed. Such strategy would entail setting up of well-equipped cardiac catheterization laboratories and cardiothoracic surgery centres across the country; and providing incentives for training and retraining of specialists in this regard [14]. This will reduce the inconveniences being faced by patients and minimize the negative impact on our nation's economy, by limiting unnecessary capital flight [18].

Most of the cardiac interventions (97.3%) were performed for children with CHDs, with only one intervention (aortic valve replacement) for a patient with acquired (Rheumatic) heart disease. This wide disparity could be attributed to the fact that CHDs are generally more prevalent than AHDs in children [19]. Unlike CHDs that cuts across all socioeconomic groups, RHD (the most common surgically-amenable AHD) tends to be confined to children of low socioeconomic class [4,5,20], whose parents/guardians are less likely to afford the high cost of surgery. PDA, VSD and TOF were the most common CHDs for which the interventions were performed in our subjects, which was consistent with other previous studies [17,21]. This pattern is a further reflection to the fact that these lesions are the most prevalent CHDs in children within the study area, as earlier highlighted by Sani et al. [20]. The small number of patients with complex CHDs in our series would suggest that many of the children might have died earlier due to the severity of their lesions [1].

The spectrum of post-operative complications observed in the study was similar to what was reported in the literature [21]. Pericardial and pleural effusion, low cardiac out-put state, and

pulmonary artery hypertensive crises constituted more than half of the complications in our patients. Pericardial effusion is known to be a common problem following cardiac surgery [21-23] and has been reported in up to 64% of patients [23]. Its occurrence has been attributed to a number of factors including surgical and/or microvascular bleeding, epicardial tear with bleeding during pacing wire removal, post-operative anticoagulation, hypoalbuminemia and surgery-induced inflammatory reaction of the pericardium among others [24]. Complete resolution of the effusion following conventional treatment is the norm; but occasionally it may become persistent and/or recurrent, or even progressed to life-threatening cardiac tamponade, requiring invasive therapy [25].

Other less common post-operative complications in our series included sternal wound infection and arrythmias. The latter comprising complete heart block, which occurred in a child with Down syndrome after AV canal repair. This is consistent with earlier observation that heart block can occur after cardiac surgery for some lesions including AV canal defect, TOF and VSD [21-29]. A plausible explanation is that these defects tend to be closely related to the conduction system, which easily gets damaged during cardiac surgery [21,26,29].

Though our patients had interventions at a relatively older age (mean age of six years), their outcome was favourable. Our post-operative mortality of 4.1% was lower than the range of 13.6% to 17.4% reported by other studies [7,17,21,30]. However, the favourable post-operative outcome may not be surprising since majority of the interventions were undertaken at an Indian hospital, which was well known for its longstanding experience and advanced level of care in cardiac surgery and cardiovascular medicine. In addition, the preponderance of simple lesions in our study cohorts, careful patient's selection before referral and continuation of the postoperative care by the local cardiac team, upon patients' return and during subsequent follow up, might also have contributed significantly to the good outcome.

5. CONCLUSION

Majority of the cardiac interventions were surgical, predominantly performed in older children with Congenital Heart Diseases and associated with good outcome. Most of the interventions were undertaken outside the country, which underscores the need to make these services locally available and accessible to the teeming children in dire need.

CONSENT AND ETHICAL CONSIDERATION

Ethical consideration: The study was performed after obtaining informed consent from the parents plus assent for children above 7 years. Ethical approval was obtained from the Hospital Ethics and Research Committee of UDUTH, Sokoto.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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