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Research Article

Acyanotic Congenital Heart Disease among Nigerian Children: A Ten Year Experience at the Lagos State University Teaching Hospital

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Abstract

Background and Objective: The diagnosis of an acyanotic congenital heart disease is often made in-utero in the developed countries but delayed in the developed countries due to social issues such as poverty, poor health seeking behavior, illiteracy, reliance on orthodox treatment and weak health systems.

Methods: Prospective and cross-sectional study involving all consecutive cases of acyanotic congenital heart diseases diagnosed with echocardiography at the Lagos State University Teaching Hospital between January 2007 and June 2016. Data were analyzed using Statistical Package for Social Sciences version 20. Level of significance set at $p < 0.05$.

Results: A total of 716 subjects were diagnosed during the study period with a male to female ratio of 1:1. The children were aged 2 days to 14years with a mean ± of 23.92 ± 36.47 months. There were 474 subjects with isolated defects and 242 subjects with more than one defects. The most common type (both isolated and multiple defects) was Ventricular septal defect followed ASD and PDA. The most common indication for echocardiography was a suspicion of an ACHD from history and physical examination.

Conclusion: In all the subjects, the most prevalent were Ventricular septal defect, atrial septal defect and patent ductus arteriosus in descending order of frequency.

Keywords: *Acyanotic; Congenital; Heart; Disease; Children; Nigeria*

Introduction

The onset and clinical manifestations of children with acyanotic congenital heart diseases (ACHDs) are protean and it depends on the type/s, degree of defects and associated co-morbidities. In resource poor countries with poor health seeking behaviour, poor resources and unavailable in or inaccessible health care, the onset of presentation may be further delayed [1-3]. Common presentations include features of heart failure, growth impairment and recurrent chest infections [1,4-7]. Furthermore, the systemic examination of children with dysmorphologies or other congenital anomaly may reveal an ACHD [8-10]. In others, an incidental finding of a murmur during a routine physical examination may identify an asymptomatic ACHD [11].

A combination of clinical presentation and echocardiographic reviews are required to make a diagnosis of ACHD. Compared to developed countries, cases of ACHD are identified in-utero with fetal echocardiography and for those who are symptomatic, or in whom a high index of suspicion is present, diagnosis is made soon after birth [3]. The reverse is the case in developing countries in sub-Saharan Africa [12]. This is not unrelated to social issues which borders on poverty, poor health seeking behaviour, illiteracy, reliance on orthodox

treatment and weak health systems. Thus diagnosis may be delayed even for those who are symptomatic earlier in life [1,3]. However, in the last decade, more centres in sub-Saharan Africa have facility for echocardiography [13] and thus the cases that may have been missed or labelled with other diagnosis have been identified as ACHD.

The management of children with ACHD is multidisciplinary [14]. Medical treatment may be combined with surgical correction of the defects. The cost implication is enormous and is shared by the family, hospital and the government. The cost of procuring medication and surgical treatment is usually more than anticipated [15-17]. For most cases, surgical corrections will be sort at facilities in developed countries and this is unaffordable for the average family. Aside from the financial stress, family members may lose work days in taking care of their children when they are sick [18-19]. The diagnosis of an ACHD alone also comes with psychological issues such as depression, anxiety, sleep disorders, guilt despair to mention a few [20,21]. The extreme involvement of family often drains the family of energy, financial resources and leisure time. For the government, resources are lost daily to medical tourism and the development of medical manpower in the care of such patient is reduced because of the medical tourism [22].

It is thus important to describe the pattern of the ACHD in the sub-region. This will be useful in planning health development in the region. The present study will document the pattern and clinical profile of children with ACHD in a tertiary hospital in Nigeria over a nine and the half year period. The findings will be compared with reports from previous studies within and outside the region.

Material and Methods

Study design: This was a prospective, observational, and cross sectional hospital based study involving consecutive subjects with acyanotic congenital heart disease.

Study setting: The study was carried out at the Lagos State University Teaching Hospital, a tertiary hospital in South Western Nigeria. The study participant was recruited as part of a large study from January 2007 to June 2016.

Participant: All the participants were children aged 0-14years of age who had a diagnosis of an acyanotic congenital heart disease by echocardiography during the study period.

Study instrument: A structured questionnaire was used. Echocardiography was performed using a 2-D echocardiography machine with facility for coloured Doppler and M-mode is available [4].

Study protocol: All children who were referred for cardiac evaluation within and or outside the hospital were reviewed by the members of the Paediatric Cardiology unit. History and physical examinations were obtained as necessary. Plain chest radiograph, electrocardiography and other ancillary investigations were done as required. Echocardiography was subsequently carried out on all the participant. Treatment was commenced as required based on standard protocol within the cardiology unit. Patients who required surgical treatment were either managed at the study center or referred for surgical correction outside Nigeria. All the subjects were followed up.

Variables: The outcome variables were:

1. Type of Acyanotic congenital heart disease.
2. Mean age of the children.
3. Indication for echocardiography and cardiac evaluation.
4. Clinical presentation of the children with ACH.

Ethical consideration

The patients were managed in accordance with standard protocol in the department. No patient personal information was used in the study. No patient samples were also used for any experiment.

Data Management: The source of the data were echocardiography reports and case notes of the patients. Details concerning the patient's biodata, clinical presentation, diagnosis, treatment, follow-up and other relevant information were recorded prospectively. The data were analyzed using Statistical Package for Social Sciences (SPSS) version 20.

Descriptive statistics was presented as percentages or means and standard deviation. Means of normally distributed variables were compared using the Student T test and proportions using Chi-square test. Skewed distribution were analyzed using appropriate non-parametric tests. Level of significance set at $p < 0.05$.

Results

Participants recruited

Between January 2007 and June 2016 a total of 1,767 echocardiographs were performed. Of all the patients who had echocardiography, 1193 children had structural heart disease; of which 1068 and 125 had congenital and acquired heart disease respectively. Of the 1068 who had congenital heart disease, 716 and 352 had ACHD and CCHD respectively. Only the patients with ACHD were analyzed here. Figure 1 depicts the study flow diagram.

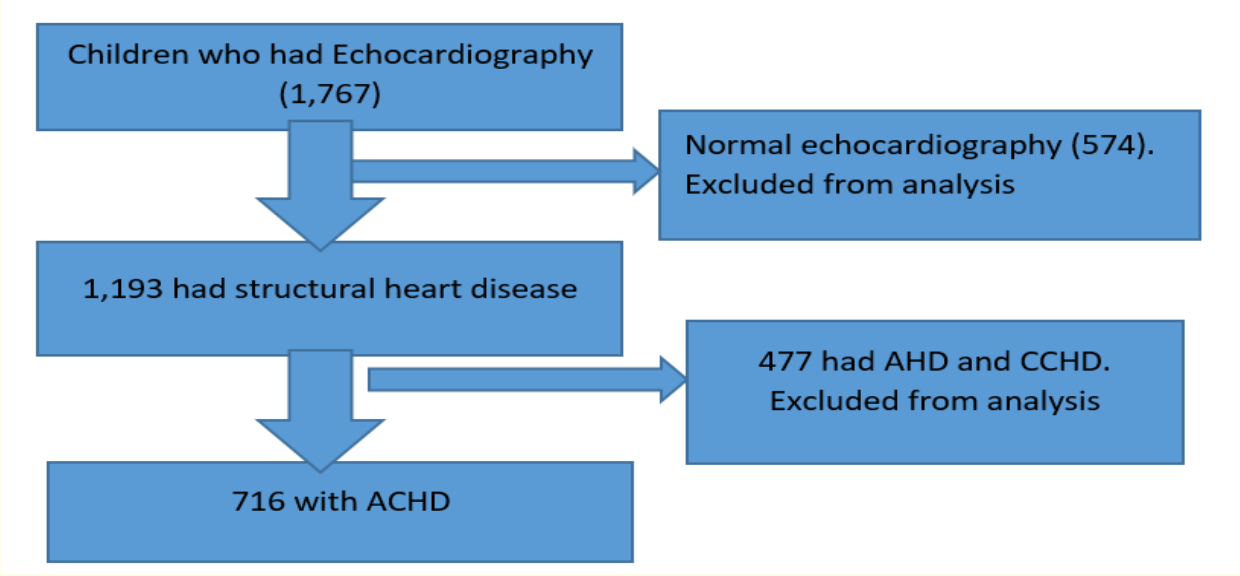

Figure 1: *Study flow diagram.*

Demographic characteristics of the study participants

Table 1 shows the demographic characteristics of the study participants. A total of 716 children had echo diagnosis of ACHD with a male to female ratio of 1:1. The children were aged 2 days to 14years with a mean of 23.92 ± 36.47 months and median of 8 years. The males and females were aged 20.71 ± 32.31 and 27.32 ± 40.15 months respectively. The age of the males was significantly lower than those of the females ($p = 0.021$). The ages of the children were ranked into subgroups as depicted on table 1. Most of the children were less than one year of age at diagnosis. As the age increased the number of children decreased. There was no significant difference in the gender distribution across the different age subgroups, ($p = 0.067$).

Variable	Total N = 716 (%)	Male N = 353 (%)	Females N = 344 (%)	P value
Age group				
< 1 year	389	208 (53.5)	181 (46.5)	0.067
1 - 5 years	181	93 (51.4)	88 (48.6)	
> 5 to 10 years	54	19 (35.2)	35 (64.8)	
> 10 years	26	11 (42.3)	15 (57.5)	
missing	66	22	25	
Mean ± SD (Months)	23.92 ± 36.47	20.71 ± 32.31	27.32 ± 40.15	
Median (Years)	8	6	9	

Table 1: *Demographic characteristics of the patients.*

Types of ACHD

Of the 716 patients with ACHD. The lesion in descending order of frequency were Ventricular Septal Defect (VSD), Atrial septal defect (ASD), Patent Ductus Arteriosus (PDA), Pulmonary Stenosis (PS), Atrioventricular canal defect (AVCD), Partial Anomalous Pulmonary Venous Drainage (PAPVC), Aortic Stenosis (AS), Coarctation of Aorta (Coart), and Common Atrium.

Out of the 716 patients with ACHDs, there were 474 subjects with isolated defects and 242 subjects with more than one defects. The most common type of ACHD (both isolated and multiple ACHD) was Ventricular septal defect followed ASD and PDA. Table 2 shows the types and frequency of all the ACHDs.

Type of ACHD	Frequency	%
VSD	314	31.8
ASD	246	24.9
PDA	216	21.9
PS	89	9.0
AVCD	64	6.5
PFO	24	2.4
AS	10	1
PAPVC	8	0.8
Coarctation of Aorta	7	0.7
Cotriatrium	6	0.6
Common Atrium	2	0.2
Mitral Stenosis	2	0.2
Total	988	100.0

Table 2: *Distribution of the types of ACHD.*

**Some had more than one defect.*

ACHD: Acyanotic Congenital Heart Defect; VSD: Ventricular Septal Defect; ASD: Atrial septal defect; PDA: Patent Ductus Arteriosus; AS: Aortic Stenosis; PFO- Patent Foramen Ovale; PAPVC: Partial Anomalous Pulmonary Venous Return; AVCD: Atrioventricular septal defect.

Among the subjects with ACHD, 242 (33.8%) had multiple ACHD, the most common combination was ASD + VSD followed by VSD + PDA and ASD + PDA. Table 3 shows the distribution of the cases with a combination of different ACHDs.

Type of ACHD	Frequency	%
ASD+VSD	63	28.7
VSD+PDA	39	17.8
ASD+PDA	28	12.8
ASD+PS	15	6.8
VSD+PS	15	6.8
ASD+VSD+PDA	14	6.4
ASD+VSD+PS	6	2.7
VSD+PA	6	2.7
ASD+PAPVC	5	2.3
AVCD+PDA	7	3.2
ASD+AVCD	4	1.8
Coarctation of Aorta + PDA	4	1.8
PDA+PS	3	1.4
ASD+VSD+PS	3	1.4
AVCD+PS	2	0.9
PDA+PFO	2	0.9
PDA+PA+VSD	2	0.9
Others* (24)	1	0.5
Total	219	100.0

Table 3: *Distribution of the cases with multiple ACHD.*

ACHD: Acyanotic Congenital Heart Defect; VSD: Ventricular Septal Defect; ASD: Atrial septal defect; PDA: Patent Ductus Arteriosus; AS: Aortic Stenosis; PFO- Patent Foramen Ovale; PAPVC: Partial Anomalous Pulmonary Venous Return; AVCD: Atrioventricular septal defect. Others includes one (1) each of the following combinations: ASD+PDA, AVCD+PDA, ASD+MA, ASD+AVCD+PDA, ASD+AVCD+PS, ASD+ Coarctation of Aorta + PDA, ASD+PA, ASD+PAPVC+VSD, AVCD+PDA+VSD, AS+ Coarctation of Aorta, AS+PDA, AS+MS, AS+PS, AS+VSD, Cotriatrium+PAPVC, Cotriatrium+VSD, MS+VSD, PA+PFO+VSD, PDA+PFO+VSD, PDA+PS+VSD, PDA+PS+PFO, PS+HPLH, PS+PFO, PS+PDA.

Almost two third of the patients, 474/716 (66.2%), had isolated ACHD. Ventricular Septal Defect was the most common isolated defect. It constituted one third of the subjects, it was followed by PDA and ASD. Table 4 shows the distribution of the isolated ACHDs.

Type of ACHD	Frequency	%
VSD	158	33.3
PDA	109	23.0
ASD	94	19.8
AVCD	47	10.0
PS	35	7.4
PFO	18	3.8
AS	5	1.1
Cotriatrium	4	0.8
Common atrium	2	0.4
PAPVC	1	0.2
Bicuspid Aortic valve	1	0.2
Total	474	100.0

Table 4: *Types and distribution of Isolated ACHD.*

ACHD: Acyanotic Congenital Heart Defect; VSD: Ventricular Septal Defect; ASD: Atrial septal defect; PDA: Patent Ductus Arteriosus; AS: Aortic Stenosis; PFO- Patent Foramen Ovale; PAPVC: Partial Anomalous Pulmonary Venous Return; AVCD: Atrioventricular septal defect.

Among the 716 children with ACHD, 171 (23.9) had an associated physical anomaly. Of these, 103 (60.2) had features of Downs syndrome. Other anomalies are shown on table 5.

Anomaly	Total (n = 171)	Frequency
Down Syndrome	103	59.2
Edward syndrome	4	2.3
Turners syndrome	2	1.2
Multiple congenital anomalies	27	15.6
Tracheoesophageal fistula	7	4.0
Cleft lip/palate	10	5.7
Spinal dysraphism	5	2.9
Omphalocele/Gastroschisis	10	5.7
Pierre Robin Syndrome	2	1.2
Ectopia cordis	1	0.6
Prune belly syndrome	1	0.6
Apert syndrome	1	0.6
Beckwith-Wiedemann syndrome	1	0.6
Total	174	100

Table 5: *Physical anomaly presents in the patients.*

Clinical presentation and associated anomaly

The most common indication for cardiac evaluation was a suspicion of an ACHD from history and physical examination. Other reasons are highlighted in table 6.

Indication for cardiac evaluation	Total (n = 700)	%
Suspicion of ACHD	280	40.0
CCF	22	3.1
Murmur	26	3.7
Breathlessness	58	8.3
Fast breathing	27	3.8
Recurrent LRTI	20	2.9
Cyanosis	17	2.4
Failure to thrive	36	5.1
Congenital/Chromosomal anomaly	171	24.4
Others*	43	6.1

Table 6: *Indications for cardiac evaluation.*

Others include – enlarged adenoid, feeding difficulties, haemangioma, chest pain, tachycardia, bradycardia, prematurity, cardiomegaly, Dextrocardia, congenital cataract, palpitation, cystic lung disease, Hirsch sprung disease, congenital rubella syndrome, stroke and prolonged jaundice with dysmorphologies.

ACHD: Acyanotic Congenital Heart Defect; CCF: Congestive Cardiac Failure; LRTI: Lower Respiratory Tract Infection.

Discussion

This is the largest series describing the profile of children with acyanotic congenital heart disease in Lagos and in Nigeria as a whole. This study highlights the clinical profile and prevalence of different types of ACHD in children in a tertiary hospital. We documented an equal prevalence of males and females in the present study. The finding in this regard is consistent with reports from previous studies [13,23,24]. The difference between the present study and the previous reports lies in the fact that those earlier studies included cyanotic and acyanotic congenital heart lesions. There are limited studies on acyanotic congenital heart disease. In the report of ACHD in children by Mundada and colleagues [6] from India, the gender of the children were reported for the individual ACHDs and not for the total group. Thus it was not feasible to compare between the present study and the report from India. The age distribution of the children in the present study is similar with reports from previous studies [13,23,24]. The implication of the present finding is that more cases of congenital heart diseases are diagnosed in children in the first year of life. This may be because of increased awareness and availability of skilled manpower and technologies necessary to make such diagnosis in the sub-region.

Concerning the distribution of the different isolated ACHDs, VSD was the most common followed by PDA and ASD. This ranking is a trend that has been reported in previous reports within and outside Nigeria [13,23,25]. However in a few studies, the ranking reported in order of frequency was VSD, ASD and PDA [24,26,27]. This is also the finding in the current study when all subjects including subjects with more than one defect. In both groups of ranking, a closer look at the figures showed that the difference between ASD and PDA was not much. Another important finding noted in the present study was that, the shunt defects were more common than the obstructive lesions. This is also not surprising given that the obstructive lesions are far less common compared to the shunt defects. In the present study, the distribution of the multiple ACHD featured mostly those combinations that had VSD, ASD and PDA. This was also not surprising given that those were the commonest ACHDs. It was difficult to compare the finding in the regard with other reports because the other studies did not comment on the distribution of the multiple ACHDs. The distribution of the different types of ACHD in the present study have shown that irrespective of the difference in race and geographic region, the prevalence of the different types of ACHD has remained unchanged.

The clinical presentation of children with ACHD are non-specific. In the present study the children presented with common findings such as breathlessness, fast breathing, CCF, failure to thrive and the incidental finding of a murmur. The findings in this regard is consistent with reports from previous studies [23,24]. The most common indication for cardiac evaluation was a suspicion of ACHD. Those children had clinical and physical findings and occasionally a radiological evidence of ACHD that necessitated cardiac evaluation and echocardiography. Almost a quarter of the children had an associated physical and or congenital anomaly necessitating cardiac evaluation. Some of the patients had more than one indication for a cardiac evaluation. The clinical features of ACHD are thus non-specific and a high index of suspicion is required to make a diagnosis.

The aetiology of ACHDs is largely unknown. However, some congenital and chromosomal anomalies have been linked with ACHD. In the present study, chromosomal anomalies such as Down syndrome, Edward and Turners syndrome were recorded in some patients. It has been proven that patients who present with those chromosomal anomalies may develop some ACHD [28]. In addition, some children with ACHD had multiple congenital anomalies. By multiple congenital anomalies, we mean children who presented with physical anomalies and dysmorphologies affecting more than one body structure or any other phenotypic presentation that was not specific for a known syndrome. Those patients could not afford karyotyping to identify a specific disorder or associated chromosomal anomaly. It has been established that children who have multiple congenital anomalies may also have some congenital heart diseases [28]. Furthermore, ACHDs were observed in some patients who presented with some physical disorders affecting a single organ such as cleft lip/palate, spinal dysraphism, omphalocele, Gastroschisis and Tracheoesophageal fistula. For example, congenital heart diseases such as ASD, VSD and PS were recorded in some children with cleft lip/palate, PDA and PS were noted in children with Omphalocele, the cases of spinal dysraphism had mainly ASD, VSD and PS and those with Tracheoesophageal had ASD, VSD and PDA. Our finding of congenital heart in those single organ defects is not out of place. This is because more than two to three decades ago some other researchers have earlier

reported cases of ACHD in disorders like cleft lip/palate [29]. Similarly, ACHDs such as ASD, PS and VSD have been reported in cases of omphalocele and Gastroschisis [30] while different congenital heart diseases have been reported in cases of isolated Tracheoesophageal fistulae [31]. Given that cases of ACHD were reported in children with chromosomal anomalies, multiple congenital anomalies and single organ defects it is imperative that all children with those kind of defects be screened for congenital cardiac anomalies that may worsen their prognosis. This will provide opportunities for early detection and treatment to improve the outcome.

Conclusion

This study has reported the clinical profile of children with ACHD. Ventricular septal defects, PDA and ASD were the most prevalent types in descending order of frequency in those with isolated defects. While in all, including subjects with more than one defect the most prevalent were VSD, ASD and PDA in descending order of frequency. A combination of multiple ACHD were noted and the most common were ASD+VSD, VSD + PDA and ASD + PDA combinations. Incidental findings of ACHD were noted in children with phenotypical syndromes and structural anomalies. It is recommended that children with physical anomalies be offered cardiac evaluation for early identification and treatment of the associated cardiac anomalies.

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Conflict of Interest

The authors declare no conflict of interest.

Authors Contribution

BA Animasahun was the project leader, she also conceived and designed the study. BA Animasahun, AD Madise-wobo and OY Kusimo were involved in the data collection, they also drafted the manuscript and were involved in the analysis and interpretation of data with critical review of the manuscript for important intellectual content. All the authors approved the final manuscript.

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