



Echocardiographic screening in a resource poor setting: Borderline rheumatic heart disease could be a normal variant



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ABSTRACT

Objective: To estimate the echocardiography confirmed prevalence of rheumatic heart disease (RHD) in school children in Fiji.

Design: Cross-sectional observational study.

Setting: Ten primary schools in Fiji.

Patients: School children aged 5–14 years.

Interventions: Each child had an echocardiogram performed by an echocardiographic technician subsequently read by a paediatric cardiologist not involved with field screening, and auscultation performed by a paediatrician. **Main outcome measures:** Echocardiographic criteria for RHD diagnosis were based on those previously published by the National Institutes of Health (NIH) and World Health Organization (WHO), and data were also analyzed using the new World Heart Federation (WHF) criteria. Prevalence figures were calculated with binomial 95% confidence intervals.

Results: Using the modified NIH/WHO criteria the prevalence of definite RHD prevalence was 7.2 cases per 1000 (95% CI 3.7–12.5), and the prevalence of probable RHD 28.2 cases per 1000 (95% CI 20.8–37.3). By applying the WHF criteria the prevalence of definite and borderline RHD was 8.4 cases per 1000 (95% CI 4.6–14.1) and 10.8 cases per 1000 (95% CI 6.4–17.0) respectively. Definite RHD was more common in females (OR 5.1, 95% CI 1.1–48.3) and in children who attended school in a rural location (OR 2.3, 95% CI 0.6–13.50). Auscultation was poorly sensitive compared to echocardiography (30%).

Conclusion: There is a high burden of undiagnosed RHD in Fiji. Auscultation is poorly sensitive when compared to echocardiography in the detection of asymptomatic RHD. The results of this study highlight the importance of the use of highly sensitive and specific diagnostic criteria for echocardiography diagnosis of RHD.

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1. Introduction and background

Pacific Island countries have among the highest prevalence of rheumatic heart disease (RHD) and incidences of acute rheumatic fever (ARF) documented in the world [1–4]. The Global Burden of Disease study estimated that RHD caused 4126 deaths in 2010 in the Oceania and Australasia regions, with 37,789 disability-adjusted life years lost [5,6]. The true figures are likely to be higher as death reporting in

many Pacific countries is poor and autopsies to confirm the cause of death are not commonly performed.

Since 2005 Fiji has had a RHD control and prevention programme, structured according to World Health Organization (WHO) recommendations [7]. The WHO guidelines recommend screening for new cases to identify RHD before it becomes symptomatic so that secondary prevention can be commenced. However no guidelines for undertaking screening as part of a RHD prevention programme are available.

In 2006 a study undertaken by our group utilised a two-step process of auscultation followed by echocardiography of children with suspicious murmurs, using diagnostic criteria on echocardiography defined by National Institutes of Health (NIH)/WHO Working Group [8]. In that study, 3462 primary school children aged 5–15 years were screened, and the prevalence of definite RHD was 4.1 per 1000 (95%

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CI 2.2–6.8) while the prevalence of definite plus probable RHD was 8.4 cases per 1000 (95% CI 5.6–12)

These data support the contention that there is a high prevalence of previously undiagnosed RHD in school children in Fiji. However, other studies have found auscultation to have low sensitivity for detecting RHD, [9] suggesting that the two-step process of screening may underestimate the true disease burden. Therefore, in the present study, we aimed to determine the prevalence of RHD in iTaukei (indigenous Fijians) school children using echocardiography, and to compare echocardiographic screening of all participants with auscultatory screening.

2. Materials and methods

2.1. Setting

Fiji is a nation of approximately 300 islands located in the Western Pacific. It has a population of 837,271 people comprised of 2 main racial groups: iTaukei (Melanesian) (56.8%) and Indo-Fijians (37.5%) with the remaining 5.7% of the population consisting of people of other racial backgrounds (other Pacific Islanders, Chinese, Europeans and mixed race ethnicities) [10]. This project was undertaken in the Central Medical Division of Fiji on the Island of Viti Levu. In Fiji, there is a very high rate of school enrolment (98%), however school attendance diminishes throughout primary school, particularly in rural areas where it is estimated that 15% of children do not complete primary school, due to poverty [11].

2.2. Study design

This was an observational cross-sectional study of school children aged 5–14 years. Each child enrolled had a screening echocardiogram for RHD as well as auscultation performed by a paediatrician.

We used stratified sampling to account for population distribution by ethnicity and by rural/urban location. Ten schools in the Central Medical Division of Fiji were randomly selected to participate in the study, with a total number of 2297 students enrolled in 2008. Recruitment and enrolment took place between February 2009 and October 2010.

We used the prevalence of RHD found in our previous study in Fiji (0.84%) to guide our sample size calculation. Reasoning that echocardiographic screening of all children should result in a higher prevalence, we calculated that a sample size of 1900 children would allow us to detect a prevalence of RHD of 1% with 95% CI of 0.6–1.5%.

Children were allocated to urban or rural status depending on the location of the school they attend, not their place of residence as this is difficult to ascertain accurately in this setting. School aged children were enrolled after written informed consent was obtained; assent was also obtained for all children aged more than ten years.

2.3. Echocardiography

All children underwent a screening echocardiogram. Echocardiography was performed by a trained technician using a Siemens Cypress Accuson portable echocardiography machine using either a 7–3 or 3–2 MHz phased array transducer as appropriate. The screening protocol aimed to assess the morphology of the mitral and aortic valves using two dimensional (2-D) modes in three views, and an assessment of regurgitant flow by colour Doppler across the mitral and aortic valves in three views. When regurgitation was apparent, color wave Doppler was used to quantify velocity and duration of the regurgitant jet. These views also enabled a screening examination for common congenital abnormalities including ventricular and atrial septal defects.

All echocardiograms were reviewed by a cardiologist independent of the initial screening. If the cardiologist viewed a screening echocardiogram and there was uncertainty, a second cardiologist was asked to review the echocardiogram, blinded to the review of the first cardiologist.

2.4. Diagnostic criteria

Echocardiographic criteria for RHD were based on those previously published by the NIH and WHO, [8] modified following recent RHD screening studies in Tonga [12] and Fiji [13]. These criteria require auscultatory findings consistent with RHD (i.e. a murmur of mitral and/or aortic regurgitation) in addition to echocardiographic findings for the diagnosis of “definite” and “probable” RHD, but not “possible” RHD. For the present study we removed the requirement for auscultatory findings and focused on echocardiography alone. These criteria are subsequently referred to as the “modified NIH/WHO criteria”.

New standardized guidelines for echocardiographic diagnosis of RHD were published by the World Heart Federation (WHF) in 2012 (referred to as the “WHF criteria”) after the commencement of this study [14]. Although our study was not designed specifically for the WHF criteria, we made inferences from our echocardiographic findings to make a diagnosis of RHD using the WHF criteria in order to compare them to the modified NIH/WHO criteria in the following ways: 1) we assumed that the subjective recording of a valve as ‘thick’ using the modified NIH/WHO criteria corresponded to a thickness ≥ 3 mm for the WHF criteria; 2) multiple morphological variables were combined to categorize data for analysis using WHF criteria including thickening of anterior or

posterior mitral valve leaflets, presence of elbow deformity, tethering of mitral valve chordae (excessive leaflet tip motion); and 3) Doppler measurement variables were adapted by assuming that a subjective assessment of “a high velocity mitral regurgitant Doppler jet” was equivalent to a measured jet with a velocity > 3 m/s in cases where velocity specific measurement was not recorded (Table 1).

2.5. Auscultation

Auscultation by a paediatrician was undertaken using a standard approach. The paediatrician was asked to categorise the nature of the murmur if present as ‘systolic’, ‘diastolic’, ‘thrill’ or ‘other’ and provide a diagnosis of ‘pathological’ or ‘suspicious’. If the murmur was termed ‘pathological’ the paediatrician was asked to record whether it was ‘mitral regurgitation’, ‘mitral stenosis’, ‘aortic regurgitation’ ‘atrial septal defect’, ‘ventricular septal defect’ or ‘other’.

2.6. Statistical analysis

Univariate and multivariate regression analysis was undertaken to determine risk associations comparing gender, ethnicity, location of school and age. Sensitivity and specificity were calculated for the auscultation component of the study when compared with echocardiography. All data were entered into Epidata version 3.1 (Denmark) electronic databases and exported into STATA 12 (STATA Corp., Texas, USA, 2012) for analysis.

3. Results

3.1. Echocardiography

We screened 1666 children with echocardiography. The most common finding detected by echocardiography was mitral regurgitation with 616 (37%) of all participants having some degree of mitral regurgitation in at least one view. However, only 72 (4.3%) and 34 (2.0%) had a mitral regurgitant jet measurement ≥ 1.5 cm and ≥ 2 cm in one view respectively. There were 65 children (3.9%) in whom morphological changes of the mitral valve were seen, and of these, 25 children (0.3% of total cohort) had more than one abnormality of the mitral valve recorded. 152 children (9%) had an aortic regurgitant jet seen in at least one view and 45 (2.7%) had an aortic regurgitant jet measured ≥ 1 cm, however only three children (0.1%) had an aortic regurgitant jet ≥ 1 cm and a full envelope seen. Of note, five of these cases with aortic regurgitation of ≥ 1 cm were found to have bicuspid aortic valves and three had minor congenital defects.

We found 12 cases of definite RHD (prevalence 7.2 per 1000) and 47 cases of probable RHD (prevalence 28.2 per 1000) using the modified NIH/WHO criteria giving a total prevalence of definite plus probable RHD of 35.4 cases per 1000. The echocardiographic features of the 12 definite cases of RHD are presented in Table 2; of note, one child with definite RHD was already known to have RHD. Applying the WHF criteria resulted in detection of an additional two cases of definite RHD (prevalence 8.4 per 1000), but 29 fewer cases of probable RHD compared to the modified NIH/WHO criteria (prevalence 19.2 cases per 1000).

Indigenous Fijian (iTaukei) children made up 66.5% of the cohort and 56.4% attended a school in a rural setting (Table 3). All cases identified with definite RHD using the modified NIH/WHO criteria were of iTaukei ethnicity (10.8 cases per 1000). Definite RHD was more common in females (OR 5.1, 95% CI 1.1–48.3) and in children who attended a school in a rural location (OR 2.3, 95% CI 0.6–13.50). These associations were not observed for cases of probable RHD, in which the prevalence was higher in males, in Indo-Fijians and in urban schools. No cases of other ethnicity were identified. The associations with risk factors were similar when we applied the WHF criteria (Table 3). A multivariate analysis, using a logistic regression model that included all four co-variables (ethnicity, gender, age and location of school) found a similarly strong association of definite RHD with female gender but no association could be tested with ethnicity as all cases were in iTaukei children (data not shown).

There were 31 confirmed cases of congenital heart disease, and of these there were 11 significant lesions (prevalence 6.6 per 1000, 95% CI 3.3–11.8). These cases included lesions of atrial septal defect

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