The Impact of Echocardiographic Screening for Rheumatic Heart Disease on Patient Quality of Life

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Objective To assess the impact of rheumatic heart disease (RHD) on child-reported health-related quality of life (HRQOL) in the context of a Ugandan school-based echocardiographic screening program. Echocardiography-based screening has emerged as a tool for the early detection of RHD, but little is known about its impact on those screened.

Study design Participants included 358 children from Gulu and 28 children from Kampala Uganda. The Pediatric Quality of Life Inventory Version 4.0 was used to assess HRQOL in 4 groups of children: Gulu prescreen, Gulu postscreen, Gulu previously linked to care, and Kampala previously linked to care. Children in the pre- and postscreen groups were selected from a single school before and after screening occurred and matched by age and sex. Children previously linked to care were recruited from previous screening studies.

Results When the echocardiogram was normal, there was no difference in HRQOL in the prescreen and postscreen groups. In the postscreen group, identification of latent RHD resulted in lower physical (75.3 vs 68.3, $P = .03$) and emotional (71.7 vs 63.4, $P < .01$) HRQOL, despite a lack of symptoms. The Kampala group had longer linkage to care (42 months vs 6 months, $P < .01$) and demonstrated greater HRQOL scores compared with the Gulu-linked group (70.7 vs 77.8, $P < .01$) and the combined Gulu cohort (77.8 vs 69.4, $P = .02$).

Conclusions Echocardiography-based screening for RHD does not diminish HRQOL in Ugandan children; rather, a diminished HRQOL score may be associated with being identified as RHD positive. Further investigation is needed to understand if longer linkage to care may ultimately normalize or improve HRQOL. *(J Pediatr 2016;175:123-9)*.

Rheumatic heart disease (RHD) is the result of an abnormal immune response to untreated group A streptococcal infections.[1] Although RHD is rare in developed nations, it remains the leading cause of cardiovascular disability and death in young people worldwide.[2] Data from the 2010 Global Burden of Disease study indicate that there are more than 34 million current cases of RHD, resulting in at least 345 000 deaths each year.[3] Many of these deaths would be preventable with early detection and initiation of secondary prophylaxis.[2,3]

Screening for RHD is appealing, because the disease typically is cumulative, resulting in a latent period during which valvular damage is clinically silent.[4] Auscultatory screening has repeated shown both poor sensitivity and specificity.[5] In contrast, screening with echocardiography uncovers 3-10 cases of latent RHD for every 1 clinical diagnosis.[5,6] Accordingly, the World Health Organization recommends echocardiographic screening, when feasible, in RHD-endemic areas.[7]

There are now efforts to expand echocardiographic screening for RHD; however, there remains little investigation on the impact of screening on asymptomatic children and communities. Caregiver data indicated that an abnormal screening echocardiogram decreased the quality of life (QOL) of both the child screened and the caregiver.[8] This study raises concerns regarding the potential negative impact of echocardiography-based RHD screening. The goal of the present study was to use child-reported health-related QOL (HRQOL) to determine the effect of echocardiography-based RHD screening on school children in Uganda.

Methods

The study was conducted over 4 weeks in July and August of 2014 in the cities of Gulu and Kampala, Uganda. Four distinct groups of children (aged 5-17 years) were selected for HRQOL survey, including children before school-based echocardiographic screening (prescreen group), children after school-

HRQOL	Health-related quality of life
PedsQL	Pediatric Quality of Life
PedsQL$_{TM}$4.0	Pediatric Quality of Life Inventory Version 4.0
QOL	Quality of life
RHD	Rheumatic heart disease

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based echocardiographic screening (postscreen group), children who were RHD-positive detected through previous school-based screening and already linked to care at the Gulu Regional Referral Hospital in Gulu, Uganda (Gulu-linked), and children who were RHD-positive detected through school-based screening and already linked to care at the Joint Clinical Research Centre in Kampala, Uganda (Kampala-linked). The prescreen and postscreen groups consisted of different children but were matched by school, age, and sex. A shared school environment implies similar socioeconomic status, location of primary residence, and average distance to a health clinic. The postscreen group was aware of their preliminary diagnosis but had not presented for follow-up evaluation at Gulu Regional referral hospital. The Gulu-linked and Kampala-linked groups consisted of children found to be RHD positive during past school-based screenings who had been linked to clinical follow-up since 2013 (Gulu)[9] and 2010 (Kampala).[6] In the school children who were RHD-positive, latent diagnoses of borderline vs definite RHD were given in accordance to the criteria of the World Heart Federation.[10]

Demographic data were collected by the use of the participant as the key informant. A study-specific questionnaire was used to capture age, school grade, and screening status (prescreen/postscreen). For participants who were postscreen, data on final diagnosis (normal, borderline RHD, definite RHD), date of diagnosis, cardiovascular symptoms, and prescription of regular penicillin prophylaxis were obtained from the medical record (**Table**). HRQOL surveys were administered in-person by volunteers in the language most comfortable for the participant, including English, Acholi (Gulu), or Luganda (Kampala).

The Pediatric Quality of Life Inventory Version 4.0 ($PedsQL_{TM}4.0$)[11] tool, a questionnaire for children aged 5-18 years, was used to assess HRQOL (http://www.pedsql.org). Although it has been validated in more than 90 country/language combinations, to our knowledge this was the first documented use of this tool in Uganda. The $PedsQL_{TM}4.0$ is a 23-item survey with 4 areas of focus: physical functioning (8 items), emotional functioning (5 items), social functioning (5 items), and school functioning (5 items). Each item had 5 possible responses measuring the extent to which the particular item was a problem during the past month. The possible responses were measured with a 5-point Likert scale from 0 = never a problem to 4 = almost always a problem. These reverse scaled scores were then transformed to a score of 0-100 to create the overall scaled score and the subcategory scores, with higher scores representing better QOL.

The study was approved by the Institutional Review Boards of Makerere University College of Health Sciences, Kampala, Uganda, the Uganda National Council of Science and Technology, and Children's National Health System, Washington, DC. Written permission for school participation was obtained from the headmaster or school advisory council. An informational letter (in the local language) was sent home with all students at least 1 week before school screening. According to local standards, written informed consent was obtained from participants 15 years of age or older; children between the ages of 8-15 years signed the written informed assent after parental permission for participation, and children younger than 8 years had only parental consent.

Statistical Analyses

Study data were collected and managed with the REDCap electronic data-capture system hosted at Children's National Health System.[12] Data analysis was undertaken with MedCalc for Windows version 12.2 (MedCalc Software, Ostend, Belgium). All tests were 2-sided, and *P*-value of <.05 was considered to be statistically significant. Continuous data were tested for normality and then evaluated with the Student *t* test. Results were expressed as a mean ± SD. Group comparisons of categorical data were conducted with the Pearson χ^2. Data from the 3 groups in Gulu were analysed as prescreen or postscreen and as RHD positive or RHD negative. Data from children linked to care in Kampala were evaluated in a subanalysis, because no matched control group (children who were RHD-negative or prescreen children) was available.

Results

In Gulu, 358 children participated. Of these children, 139 were from the prescreen group, 138 from the postscreen group, and 81 (37 borderline RHD and 44 definite RHD) from the Gulu-linked group. There were no differences in age between children who were in prescreen group and children in postscreen group (mean age 10.6 vs 10.6 years, $P = .84$). Of children in the postscreen and Gulu-linked group with a preliminary or final diagnosis of RHD, only 4

Table. Demographic data for schoolchildren undergoing study in Gulu and Kampala, Uganda

	Prescreen	Postscreen	Statistics	Gulu-linked	Kampala-linked	Statistics
n (total)	139	138		81	28	
Borderline	-	-		44 (54.3%)	17 (60.7%)	$\chi^2 = 1.9$, $P = .17$
Definite	-	-		37 (45.7%)	11 (39.3%)	
Age, y (mean, ±SD)	10.6 (7.7-13.5)	10.6 (7.8-13.4)	$P = .84$	12.1 (10.1-14.2)	13.1 (11.3-14.8)	$P = .04$*
Penicillin prophylaxis	-	-		52 (64%)	11 (40%)	$\chi^2 = 5.3$, $P = .02$*
Time from diagnosis, mean (range)	-	-		6 mo	42 mo	
	-	-		(2-8)	(38-44)	$P < .01$*

*$P < .05$.