# **ORIGINAL ARTICLE**

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# Increasing the health-related quality of life in children with congenital Rubella syndrome: A mixed-methods study



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### ABSTRACT

**Introduction:** Rubella infection during the first trimester of pregnancy is a significant cause of fetal malformation syndrome, or Congenital Rubella Syndrome (CRS), which affects the children's quality of life (QoL). A Community Engagement Program was arranged collaboratively to increase the QoL of children with CRS and enhance parents' skills and knowledge to better care for their children with CRS.

**Methods:** A mixed-method descriptive study was conducted from April to October 2022 at Dr. Sardjito General Hospital. Sample recruitment used the purposive sampling method, which had been clinically and serologically diagnosed with CRS. Children's QoL data were analyzed with a cross-sectional study design. The tools used were the Pediatric Quality of Life Inventory (PedsQL)TM 4.0 Generic Core Scales and SPSS version 26 to analyze paired T-tests. A focus group discussion (FGD) was used for qualitative thematic analysis.

**Results:** We involved ten members willing to participate voluntarily in the PedsQL assessment and FGD. The result was an increase in patients' QoL in the Physical Function domain that was statistically significant: pre-test and post-test physical health summary [490 ( $\pm$ 261) vs. 510 ( $\pm$ 251); p=0.022]. While social, emotional, and school functions increased were not statistically significant. From the FGD, the parents had diverse experiences and understanding of CRS-supporting examinations and treatments needed. Parents felt the children's growth and development, as social and school functions were the common burdens. In order to help CRS prevention, the parents suggested a more comprehensive premarital program and antenatal care.

**Conclusions:** The Community Engagement Program helps to increase the patient's QoL in the Physical Function domain. Comprehensive multidiscipline management is needed to support children's growth, social and educational functions. Periodic evaluations are required to monitor the QoL of children with CRS.

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of cases originating from Southeast Asia, particularly in nations without rubella immunization programs.<sup>3,4</sup> A retrospective study performed between July 2008 and June 2013 at Dr. Sardjito Hospital, Yogyakarta, Indonesia, found that CRS incidence was 0.05- 0.0.25 per 1000 live births.<sup>5</sup> Meanwhile, after the measlesrubella vaccination campaign in 2017, the incidence of CRS decreased to 0.08 per 1000 live births, according to a study conducted in Yogyakarta, Indonesia.<sup>6</sup>

The Government of Indonesia is firmly committed to eliminating measles, rubella, and Congenital Rubella Syndrome through the MR vaccination campaign. The Ministry of Health arranged a surveillance system for CRS to analyze the disease burdens of CRS and the long-term impact of MR vaccination. The surveillance was first introduced in 2018 using a sentinel system in 13 sentinel hospitals and was gradually expanded to 22 sentinel hospitals in 2022.<sup>7,8</sup>

The term, health-related quality of life (HRQoL), refers to the effect of a specific illness, medical therapy, or health care policy on the capacity of patients to operate in and derive personal pleasure from diverse physical, psychological, and social life situations.<sup>9</sup> Measuring children's HRQoL might help in medical care

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# INTRODUCTION

Rubella infection is a highly contagious virus-caused illness with typically mild symptoms.<sup>1</sup> However, a fetal malformation syndrome known as Congenital Rubella Syndrome (CRS) can be caused by maternal infection during the first trimester of pregnancy.<sup>2</sup> The most prevalent abnormalities associated with CRS are hearing loss, vision impairment (primarily congenital cataracts), congenital heart disease, and global developmental delay.<sup>1,2</sup> Nerve deafness was CRS patient's most prevalent finding.<sup>1</sup>

Annually, there are more than 100,000 cases of CRS reported globally, with 46%

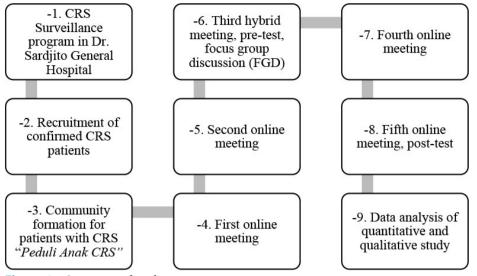
and enhance communication between doctors, parents, and children.<sup>9</sup> Previous studies showed lower HRQoL results in children with congenital abnormalities, including congenital heart disease (CHD), congenital hearing loss, low language, and congenital cataracts.<sup>9-11</sup>

Currently, there are generic and specific instruments for measuring QoL. The generic instrument is designed to assess this indicator in both the healthy population and patients with various pathologies, considering the shared interests of these groups. In comparison, specific instruments are used to collect data on specific items, such as typical disease symptoms or adverse effects of treatments.12 The most commonly used generic questionnaire to measure pediatric QoL is the Pediatric Quality of Life Inventory (PedsQL) TM 4.0 Generic Core Scales. In the previous study about the QoL in children with hearing loss in Australia, the main cause of the decreased QoL was learning difficulty and impairment in physical and social function.<sup>11</sup>

CRS accounts for severe morbidity that requires comprehensive treatment from a multidiscipline. A Community Engagement Program was arranged collaboratively by the Department of Child Health; the Department of Ear, Nose, and Throat (ENT); and the Department of Ophthalmology Faculty of Medicine, Public Health and Nursing Universitas Gadjah Mada in Yogyakarta, Indonesia. The multidisciplinary approaches of this program were to increase the QoL of children with CRS and to enhance parents' skills and knowledge to better care for and manage their children with chronic disease.

# METHODS

The Community Engagement Program team conducted this activity from April to October 2022. Participants were parents of children at Dr. Sardjito Hospital who had been clinically and serologically diagnosed with CRS. Dr. Sardjito Hospital is a teaching hospital of the medical faculty of the Universitas Gadjah Mada, a tertiarycare referral hospital of the Yogyakarta and Central Java Provinces, and assigned as one of the centers for CRS surveillance by the Indonesian Ministry of Health. This







**Figure 2.** Routine meeting of *Peduli Anak CRS*. (a) Routine online meeting, and (b) Hybrid meeting of *Peduli Anak CRS*.

program was initiated with community formation for parents of children with CRS, and it was called "*Peduli Anak CRS.*" There were 28 children participating in this community and were gathered in the WhatsApp group. The activities of this program consisted of five routine meetings between the parents and the doctors, CRS educational video-making, publishing a monitoring book for children with CRS, and publishing two educational leaflets about CRS to help parents take care of their children with CRS (Figure 1).

The first and second meetings were held online, the third was hybrid, and the last two were online (Figure 2). There were presentations from the specialists about the problems in CRS patients: hearing problems topic in the first meeting; growth and development and medical rehabilitation topics in the second meeting; metabolic disorder topic in the third meeting; visual impairment and cerebral palsy topics in the fourth meeting, and congenital heart disease topic in the fifth meeting.

The media for this program were developed by a community engagement team consisting of a pediatrician, ophthalmologist, and otolaryngologist. The video is 8 minutes 51 seconds long and contains educational material about CRS natural course and management from pediatric neurologists, cardiologists, ophthalmologists, and otolaryngologists. The team develops two leaflets. The first leaflet contains general information about CRS, including the cause, transmission, epidemiology, and manifestation criteria. The second contains management and care for CRS patients, forming multidisciplinary а approach. The monitoring book was developed to help the parents monitor the growth and development based on the anthropometric measurements and milestones, help parents to provide stimulation guidance for the children, and help to maintain the schedule of routine control.

A mixed-method descriptive study was conducted in this program to assess the QoL improvement in children with CRS and to determine parents' knowledge about CRS. The assessment of children's QoL was conducted at the third meeting for the pretest and after the fifth meeting for the post-tests. Parents' knowledge survey and FGD were arranged to gather parents' knowledge, perception, and experience. The Medical and Health Research Ethics Committee of the Faculty of Medicine, Public Health and Nursing, Universitas Gadjah Mada approved all recruitment procedures and the research protocol with KE/0574/05/2022.

The presence of hearing impairment, congenital cataracts, CHD, and developmental delay was determined by an ENT specialist using brainstemresponse audiometry, evoked an ophthalmologist, a pediatric cardiologist using echocardiography, and a pediatric neurologist, respectively. According to the World Health Organization (WHO) definition, a pediatric neurologist decides the diagnosis of CRS. The parents' educational level was divided into the following classification: high (senior high school or more) and not-high (junior high school or less).

The tool that was used to assess children's QoL was the Pediatric Quality of Life Inventory (PedsQL)TM 4.0 Generic Core Scales, which was developed by Varni et al.<sup>12</sup> This questionnaire was already considered valid and reliable for the evaluation of QoL of children in various populations using different languages such as Chinese, Thai, and also Indonesian.<sup>13-16</sup> The PedsQL 4.0 GC consists of a 23-item questionnaire and encompasses four scales: physical functioning (8 items), emotional functioning (5 items), social functioning (5 items), and academic functioning (5 items). Parents only fill out this questionnaire because of the incapability of children with CRS.

Data were processed and analyzed by SPSS version 26 (IBM Corp, Armonk, NY, USA). Patients' characteristics were presented using frequencies and percentages. The data normality test used the Shapiro-Wilk test because the total samples were less than 50. The increasing QoL of the children pre and post-test with numerical scale was analyzed with paired T-tests.

In order to gather parents' knowledge

#### Table 1.FGD lists of topics

Focus Group Discussion (FGD) Topics:

- 1. Disease course and examinations
- 2. Disease etiology and transmission
- 3. Parents' burden in caring for a child with CRS
- 4. Parents' solutions and problem-solving to prevent CRS

\*CRS: Congenital Rubella Syndrome

Table 2.	Demographics and	l medical c	haracteristics of	the participants
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Characteristics	n (10)	%
Gender		
Female	4	40
Male	6	60
Age (years)		
<1	1	10
1-5	5	50
>5	4	40
Hearing Impairment		
Yes	9	90
No	1	10
Congenital Heart Disease		
Yes	5	50
No	5	50
Visual Impairment		
Yes	6	60
No	4	40
Developmental Delay		
Yes	9	90
No	1	10
Fathers' education level		
Not High (Junior High School or less)	2	20
High (Senior High School or more)	8	80
Mothers' education level		
Not High (Junior High School or less)	2	20
High (Senior High School or more)	8	80

\*CRS: Congenital Rubella Syndrome

Та	ble 3	<b>3.</b>	Pre-test a	and	post-test	analy	sis
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PedsQL 4.0 GC Domain	n	Pre-test Total Score ± SD	Post-test Total Score ± SD	P-value
Physical	10	490±261	510±251	0.022
Social	10	322±126	337±113	0.217
Emotional	10	370±105	397±105	0.065
School	10	342±94	355±97	0.586

\*P-value <0.05 was considered significant. GC: Generic Core; SD: Standard Deviation.

about CRS, a survey and a FGD were conducted. The survey consisted of multiple-choice questions concerning the cause of CRS, the disease's transmission, the affected organ systems, the supporting examinations needed, and multidiscipline management. The survey was distributed in the first meeting to provide an overview of parents' knowledge and perceptions. The survey results were compared and combined with the parents' experiences shared during the FGD.

Focus Group Discussion (FGD) is a technique for gathering in-depth qualitative data on a particular social issue or topic to create more authentic settings in which participants may influence and be impacted by one another.<sup>17,18</sup> A pediatric neurologist facilitated the discussion as the facilitator. The facilitator

#### Table 4. "Peduli Anak CRS" parents' questionnaire

Topics	Understand n (%)	Did not understand n (%)
CRS etiology and transmission	10 (100%)	0 (0%)
CRS complication in organs (brain, eyes, heart, ears)	10 (100%)	0 (0%)
Supporting examinations needed		
Blood examination	8 (80%)	2 (20%)
Heart examination	8 (80%)	2 (20%)
Hearing test	8 (80%)	2 (20%)
Eye examination	8 (80%)	2 (20%)
USG/CT scan of the head	6 (60%)	4 (40%)
Prevention and managements needed		
Prevention	6 (60%)	4 (40%)
Routine health control	8 (80%)	2 (20%)
Nutritional status monitoring	8 (80%)	2 (20%)
Growth and development monitoring	10 (100%)	0 (0%)
Children stimulation	8 (80%)	2 (20%)
Physiotherapy	8 (80%)	2 (20%)
Cataract surgery	7 (70%)	3 (30%)
Hearing aid device	7 (70%)	3 (30%)
Heart catheterization/ surgery	6 (60%)	4 (40%)

\*Total Participants = 10, CRS: Congenital Rubella Syndrome; CT: Computerized Tomography; USG: Ultrasonography.

#### Table 5. Themes and sub-themes of the FGD

Themes	Sub-themes
CRS Manifestation	Heart Defect
	Hearing impairment
	Global developmental delay
	Cataract
CRS Examination	Ears: BERA, ASSR, OAE
	Heart: Echocardiography
	Head: CT scan and USG
	Serology testing for Rubella
CRS Management	Routine control
-	Heart: Heart surgery and catheterization
	Eyes: Cataract surgery, Eyeglasses
	Ears: Hearing aid, Cochlea implant
	Routine Physiotherapy
CRS Transmission	Did not know
Parents' burden	Growth and development
	Social function
Parents' proposed solution	Antenatal care
	Premarital program
	Government participation

\*ASSR: Auditory Steady-State Response; BERA: Brain Evoked Response Auditory; CRS: Congenital Rubella Syndrome; CT: Computerized Tomography; OAE: Oto Acoustic Emission; USG: Ultrasonography;

poses open-ended questions to elicit responses and generate the largest number of arguments and points of view within a given amount of time.<sup>18</sup> The FGD was conducted for 45 minutes during the third "*Peduli Anak* CRS meeting" (Table 1). The meeting was held offline at the Faculty of Medicine, Public Health and Nursing Universitas Gadjah Mada. Five participants lived in Yogyakarta Province and were able to attend. The qualitative study used the thematic analysis method and was analyzed by three pediatric neurologists, an ophthalmologist, and an otolaryngologist.

# RESULTS

In this program, we built a "*Peduli Anak* CRS" community with 28 members of CRS patients' parents. Ten members were willing to participate voluntarily and committed to the pre-and post-QoL assessment; they completed the parents' knowledge survey. Others were excluded due to only attending one QoL assessment; one participant only attended the pre-test assessment, three participants only attended the post-test assessment, and others could not be contacted. The response rate of the study was 35,71%.

The characteristics of the children in this program are listed in Table 2. Most children who participated in the assessment were male (60%), and their ages were mostly 1-5 years old (50%). The father's and mother's educational levels of the 10 children mostly ranked in the High level of education (80% respectively). The clinical manifestations most commonly found were hearing impairment in 9 (90%) subjects, CHD in 5 (50%) subjects, and developmental delay in 9 (90%) subjects.

#### Children's QoL assessment

Table 3 presents the results of the PedsQL4.0 GC questionnaire. The results weretested for the normality test with the

Shapiro-Wilk test before being analyzed with paired T-tests. The normality results showed normal distribution (*p*-value: 0.05) for the Physical Function pre-test and post-test (*p*-values: 0.316 and 0.309) and Psychosocial Function pre-test and post-test (*p*-values: 0.749 and 0.212). For the total 10 participants, pre-test and post-test comparisons for Physical Function were statistically significant, while the comparison results for Social, Emotional, and School Functions were not statistically significant.

# Parents' knowledge assessment and FGD

The parents' questionnaire assessment below was conducted by 10 members who voluntarily participated in the first CRS meeting (Table 4). All the participants were aware of the cause (100%), the disease transmission route (100%), and the organs' functions affected (100%). Most parents knew that CRS could be prevented (60%). In this assessment, the parents were allowed to choose multiple supporting examinations and management as described below.

The FGD was conducted during the third meeting of "*Peduli Anak CRS*." The meeting was held offline at the Faculty of Medicine, Public Health and Nursing Universitas Gadjah Mada. The discussion was effective, and the parents actively gave their opinions based on their experiences (Table 5).

i. CRS Manifestation

The discussion was initiated by sharing various manifestations of CRS of their children.

a) Heart Defect

"We had a consultation with a pediatric, and the doctor said my child had [ventricular septal defect] VSD." – Mother of AN.

"When he was born, a heart defect was detected" – Father of FR. "My child was born with a weight of 1.600 grams, the doctor said she had a heart defect" – Mother of KA.

b) Hearing Impairment "Our daughter had hearing impairment in both ears." – Mother of NL.

<sup>•</sup>*"She did not respond when we called her"- Mother of KA.* 

- c) Global developmental delay "She still could not sit, so we went to a pediatric and the result was she had global developmental delay" -Mother of NL.
- d) Cataract

"At three months old, he was first diagnosed with cataracts in his both eyes." – Father of FR.

*"Her eyes were covered with white membranes. The doctor said she had cataract." – Mother of KA.* 

ii. CRS Examination

The parents shared their experiences from the early course of the symptoms until the children were diagnosed with CRS.

a) Ears: BERA, ASSR, OAE

"At two months old, we had consultation with an otolaryngologist, and our daughter was checked with OAE. Then she was referred to have a BERA examination. At six months old, she had ASSR." – Mother of NL. "She had OAE and ASSR" – Mother of KA.

- b) Heart: Echocardiography
  "Her condition was getting worse, so she had echocardiography and the result was the VSD got bigger than before." – Mother of AN.
  "She already had an echocardiography examination with normal result." – Mother of NL.
- c) Head: CT scan and USG "He was referred to had CT scan." – Father of FR. "She was examined with

"She was examined with [ultrasonography] USG and CT scan with brain calcification" – Mother of AZ.

d) Serology testing for Rubella "She was tested for Rubella [serology testing] at nine months old." - Mother of AZ.

"She was tested with IgM Rubella; the result was positive." – Mother of KA. iii. CRS management

In this section, the parents explained the management given for the children. a) Routine control

"Until now, we have routine control to the pediatric neurologist, pediatric cardiologist, and nutritionist."-Mother of AN.

b) Heart: Heart surgery and catheterization

"He had heart catheterization at three months old." – Father of FR. "When she was two years old, she had heart surgery." – Mother of KA.

c) Ears: Hearing aid, cochlea implant "He used the hearing aids for two years but there was no improvement. In February 2021, he had cochlea implant surgery in one ear. Four months later, he could hear." – Father of FR.

"We bought the hearing aids when she was three years old" – Mother of KA.

d) Routine physiotherapy *"She also had a global developmental delay and got routine physiotherapy." Mother of NL.*

"He started to walk at two years and sixth months old with routine physiotherapy" – Father of FR.

iv. CRS Transmission

a) Did not know. The parents did not get sufficient information and education about CRS cause and transmission before the children were diagnosed.

"In my experience, I did not get premarital education about the diseases and congenital abnormalities of children, especially for the disease with mild symptoms in pregnant women that could affect the babies, such as Congenital Rubella Syndrome [CRS]. I just knew about the disease after my daughter was diagnosed with CRS." – Mother of KA.

"In early pregnancy, I got a fever, but I did not have any rashes. The fever was mild, so I did not go to the doctor" – Mother of FR.

v. Parents' burden

The next topic was the parents' burden of caring for a child with CRS. The parents were worried about school functions and their child's learning abilities. The parents are also concerned about their social life.

- a) Growth and development "I worry about my children's growth and development in the future due to her developmental delay" – Mother of AN.
- b) Social function

"I hope that our children can socialize, not feel inferior to others,

be brave, and would be able to take responsibility for themselves." – Mother of KA.

vi. Parents' proposed solution

a) Antenatal care "I hope that medical staff can improve antenatal care and health education comprehensively." – Mother of KA.

- b) Premarital program "In my opinion, the Rubella vaccine as CRS prevention should be integrated into the premarital program by the Religious Affairs Office." – Father of NL.
- c) Government participation "I hope that the government and doctors could actively participate and give attention to the forum or community for CRS patients." – Father of FR.

# DISCUSSION

In previous studies, patients with congenital abnormalities, including CHD, congenital hearing loss, low language, and congenital cataracts, had lower.<sup>9–11</sup> This Community Engagement Program aimed to increase CRS patients' QoL by organizing various activities, including arranging seminars and discussions with the experts, creating educational leaflets for the parents, making a monitoring book for patients' growth and development, and making an educational video.

The patients' QoL was assessed twice, before and after the implementation of the community engagement program. We used PedsQL TM 4.0 Generic Core Scales that consist of two domains, the Physical Function and Psychosocial Function. The result was that after the program implementation, there was a statistically significant increase in patients' QoL in the Physical Function domain. This community engagement program could help parents enhance their knowledge to care for their children with CRS. The social, emotional, and school domains results were not statistically significant, possibly because the parents assessed children's emotions and experiences. The parents might have more limited experience of the impact of a disorder in certain situations, such as in school or social life with peers. The parents also might not directly share their children's emotions, who experience

various limitations due to CRS. The QoL examinations were done briefly so that periodical assessments are needed to better monitor and evaluate the QoL development of the children with CRS.

According to the parents' assessment, the parents understood the Rubella virus as the etiology (100%) and transmission route of the virus in pregnancy (100%). From the discussion, some of the parents experienced fever in early pregnancy. However, the parents did not have information about CRS as a possible complication then. They knew about CRS after their children were diagnosed.

In the FGD session, the parents shared their experiences with their children's disease. Their diverse experiences affected their understanding of CRS-supporting examinations and treatments needed, as shown in the questionnaire result. For the supporting examinations, the blood examination (80%), heart examination (80%), ear examination (80%), and eve examination (80%) were chosen by many of the parents. At the same time, the USG/ CT scan of the head was the least chosen option. All parents chose growth and developmental monitoring (100%), while heart surgery/catheterization (60%) was the least chosen management option.

The parents were allowed to share their thoughts on managing CRS. A comprehensive premarital program and antenatal care are needed to prevent CRS. Continuous support from the Government and clinicians are necessary for the parents to care for their children with CRS.

In this program, each patient suffered from various manifestations of congenital malformations, including hearing impairment, congenital heart defect, visual impairment, seizure, and global developmental delay. These conditions affect the quality of life in physical, social, emotional, and school functions. The common burden felt by the parents also included children's growth and development and school function. The children's QoL assessment results showed a statistically significant increase in patients' QoL in the Physical Function domain. Multidisciplinary parties did this community engagement program in a short period. It could help the parents enhance their knowledge to care for their children and increase the quality of life of the children with CRS. Further comprehensive and sustainable multidiscipline management will be needed to help increase the children's QoL.

The community engagement program consists of online and hybrid activities. Some patients live outside the Yogyakarta Province (DIY); therefore, all the patients could attend the online activities in the community and online meetings. The focus group discussion was held hybrid, offline for patients who lived in DIY, and online for patients who lived outside DIY; unfortunately, the online participants were less active than the offline participants. Some circumstances could affect the effectiveness of online activities, including internet access, network stability, and situational conductivity. The methods of knowledge delivery in this program using various media consist of experts' presentations in online meetings, educational leaflets on CRS, CRS management leaflets, a monitoring book for patients, and an educational video that help to support parents' knowledge and understanding to care for the children.

# CONCLUSIONS

The purpose of the children's QoL assessment and the parents' FGD was to evaluate and obtain parents' perceptions to improve the QoL of children with CRS and develop parents' ability and knowledge to care for these children. The patients' QoL in the Physical Function domain increased in this Community Engagement Program. The overall concerns from the parents were their children's growth, social and educational functions, and they recommended that more proactive CRS prevention efforts through premarital and prenatal care are necessary. Periodic evaluations should be required to monitor and assess the improvement in the QoL of children with CRS. Our study had several limitations, including small sample size and the resulting low power, which might affect the detection of the significance of any predictors. Since there is no specific questionnaire for CRS, we used the generic scale for Health-related QOL. Further multicenter studies with a larger sample size are important to clarify and confirm our findings.

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# **CONFLICT OF INTERESTS**

The authors declare that they have no competing interest. The article has not been previously published and is their original work.

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