



# Psychosocial Problems In Congenital Adrenal Hyperplasia Children During Pandemic Covid-19

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## Abstract

Congenital Adrenal Hyperplasia (CAH) is a chronic condition that need longtime medical treatment. Chronic disease is a psychosocial stressor that related to mental, emotional and behavior problems in children and adolescent. Pandemic Covid-19 will increase risk of psychosocial problems. This study aims to identify psychosocial problems in children with CAH during pandemic Covid-19. This study took place at the Pediatric Endocrinology Outpatient Clinic in Dr Soetomo Academic General Hospital, Surabaya, Indonesia, assigning children aged 3 to 17 as sample. The sample size of this study was 28 subjects. Data were accumulated through interviews, questionnaires and medical records. Strength and Difficulty Questionnaire (SDQ) was used in this study, distributed via google form and filled by the parents. The statistical analysis was undertaken using SPSS 21.0. Results showed that out of 28 children with CAH, 28.6% have emotional problem, 25% with behavioural problem, 46.4% have peer problem, 82.1% did not have hyperactivity/inattention problem and 85.7% with normal prosocial behaviour. It was concluded that children with CAH prone to have psychosocial problems.

**Keywords:** Congenital Adrenal Hyperplasia, psychosocial problems, Strength and Difficulty Questionnaire, pandemic Covid-19

## Introduction

Congenital Adrenal Hyperplasia (CAH) is an autosomal recessive genetic condition, consist of a family of defects in the synthesis of steroid hormones in the adrenal cortex. More than 90% of cases are caused by 21-hydroxylase deficiency, resulting in impaired synthesis of cortisol and aldosterone and steroid precursors being converted to adrenal androgens [1]. Congenital Adrenal Hyperplasia is one of the chronic condition that requires lifelong medical treatment and frequent hospital visits [2]. Chronic physical illness will increase risk of mental, emotional and behavioural problems in children and adolescent [3]. Psychopathological risk will increase 2.5 times in children with chronic condition compared to normal children [4]. In the pandemic situation, when there are massive restriction of social activities, online school, limited acces of health services and disruption of drug distribution, causing many patients with chronic diseases not be able to visit hospital and took medication regularly. All of these are additional stressor that can lead to psychosocial problems in children with chronis illness like CAH [5]. Psychosocial problems in children and adolescent is a serious condition because its impact on developmental, productivity and quality of life [6].

Psychosocial disorder is a mental illness caused or influenced by life experiences, as well as maladjusted cognitive and behavioral processes [7]. Strength and Difficulty Questionnaire is one of the instrument to detect psychosocial problems in children and adolescent. Its consist of five subscale, emotional, behavioural, hyperactivity/inattention, peer and prosocial problems [8]. Study about psychosocial problems in CAH children already done along the world, but in Indonesia, this study especially during pandemic condition not yet exist. Until now, therapy in CAH children is more focused on medical aspect, while the psychological aspects are still often neglected. This prompted researchers to analyze the psychosocial problems in CAH children, especially during pandemic Covid-19.

## Methods

This research is a cross-sectional study, that was conducted at Pediatric Endocrinology Outpatient Clinic in Dr. Soetomo Academic General Hospital, Surabaya, Indonesia. Participants from this study were 3-17 years old children who had previously been diagnosed with CAH and underwent outpatient treatment. The study was carried out between March to April 2021. Subjects were recruited with a simple random sampling method that eligible for the inclusion and exclusion criteria. The inclusion criteria in this study were children and adolescents aged 3-17 years with CAH, parent agreed to participate in this study and filled the questionnaires. Children and adolescents with a critical condition, family history with psychologic problems and taken psychotropic drugs were excluded. During the two-month study period, 28 children with CAH were found and were included as research subjects. The informed consent was waived and signed by the participant's parents and/or legal guardians. This study was approved by the Clinical Research Unit of Dr. Soetomo Academic General Hospital, Surabaya, Indonesia number 0159/KEPK/III/2021.

The participants who met the inclusion criteria and had signed the informed consent form will be given SDQ questionnaire through google form. Anamnesis was taken for the demographic data. Background clinical data were collected from medical records including age at diagnosis, clinical and biochemical presentation at diagnosis, sex assignment and history of genitoplasty. The diagnosis of CAH is based on the result of 17-hydroxyprogesterone (17-OHP). Assessment of psychosocial problems was done using the parent-reported SDQ. Emotional problems was defined when SDQ score 7-10, while behavioural problems when SDQ score 5-10. Hiperactivity/inattention problems when SDQ score 7-10. Peer problem when the score 6-10. Prosocial problem was defined when SDQ score was 0-4. The SDQ score can be used as continuous variables or by classifying them as normal, borderline and abnormal [8].

Descriptive statistical were used in this study. Categorical data were reported as frequency or percentage. Numeric data were reported as mean and standard deviation (SD). The statistical analysis was undertaken using Statistical Package for the Social Sciences (SPSS) 21.0.

## Results and Discussion

The number of samples that meet the inclusion criteria were 28 children, 10 male children and 18 female children. There were psychosocial problems in CAH children, consisting of emotional, behavioural, hyperactivity/inattention, peer and prosocial problems.

## 1. Basic Characteristics of Research Subject

The research subjects were presented based on basic characteristics, namely gender, age, race, family history and initial symptoms of CAH. The basic characteristics of research subjects are presented in Table 1

**Table 1.** Basic Characteristics of children with CAH

Characteristics	Children with CAH (N=28)
Sex	
Male	10 (35.7)
Female	18 (64.3)
Age (months)*	94.86±47.85
3-10 yr	21 (75)
11-17 yr	7 (25)
Race	
Javanese	26 (92.8)
Non Javanese	2 (7.2)
Initial symptoms	
Ambiguous genitalia	14 (50)
Vomiting	12 (42.8)
Without symptom	2 (7.2)
Family history with CAH	
Yes	4 (14.3)
No	24 (85.7)
Family history with psychosocial problems	
Yes	0 (0)
No	28 (100)
History consumption of psychotropic drugs	
Yes	0 (0)
No	28 (100)

\*mean±SD

The number of patients with CAH in the Pediatric Endocrinology Outpatient Clinic Dr. Soetomo Academic General Hospital, Surabaya, Indonesia during the study period of this research, namely from March to April 2021 were 30 patients. One subject was below 3 years old and the other was refused to fill up the questionnaires. In this study, 28 children with CAH who met the inclusion criteria were included as samples. All the CAH children were diagnosed based on increased in blood concentrations of 17-OHP. The results showed serum 17-OHP with minimum value of 178 ng/dL and maximum value of 1771 ng/dL. In CAH, there were deficiency enzyme 21-OH, causing accumulation of 17-OHP because of aldosterone and cortisol formation pathways were blocked. Congenital Adrenal Hyperplasia can be diagnosed definitively by finding an elevated level of 17-OHP [1].

Globally, data from neonatal screening involving 6.5 million newborns show an incidence of 1 in 15,000 babies experiencing CAH, making CAH as a rare disease [9]. Data from endocrinology unit Indonesian Pediatric Society registered 400 children with CAH [10].

In this study, 10 (35.7%) children were male and 18 (64.3%) children were female with the youngest age 3 years old and the oldest was 17 years old. Female with CAH are at higher risk of behavioural problems (emotional, conduct, hyperactivity/inattention, peer relation problems, and prosocial behaviour) due to excessive androgenization [11].

Fifty percents presented with ambiguous genitalia, while 42.8% presented with vomiting as initial symptoms. Girls who born with CAH are typically identified at birth or during early infancy and usually presented with varied degrees of virilization. The appearance of external genitalia can vary from mild, isolated clitoromegaly to ambiguous genitalia, while there is no genital ambiguity in male with CAH [12]. In this study 4/18 female children with CAH who presented with ambiguous genitalia already done genitoplasty. Age of patients at the time of surgery ranged from 3 to 9 years. The remaining have not done genitoplasty because parents opted to wait until the children were older before considering surgery.

Twelve children with CAH in this study have done karyotyping examination with the results 11 children with 46 XX and 1 child with 46 XY. Due to financial problems, the remaining of the subjects can not do karyotyping examination. As many as 85.7% subjects from this study did not have the same family history. This is similar to study conducted by Hamed, 2020 in Egypt, showed that only 3/55 children with CAH (5.45%) have family history of CAH [11].

In recent years, increasing attention has been drawn to psychosocial adjustment and mental health of individuals with CAH and their need for psychological support. It has also been suggested that atypical early hormone environment associated with classic CAH like cortisol insufficiency or overproduction of androgen, may contribute to mental health problems [13].

## 2. Psychosocial Problems in CAH children

**Table 2.** Psychosocial problems in CAH children

SDQ	Frequency	Percentage
Emotional problems		
Normal	20	71.4
Abnormal	8	28.6
Behavioural problems		
Normal	21	75
Abnormal	7	25
Hiperactivity/inattention problems		
Normal	23	82.1
Abnormal	5	17.9
Peer problems		
Normal	15	53.6
Abnormal	13	46.4
Prosocial problems		
Normal	24	85.7
Abnormal	4	14.3

In this study, 8 children have emotional problems (28.6%). This is in line with study of Kung, 2018 at United Kingdom, showed that 7/42 children with CAH (16.7%) have emotional problems [14]. Study of Hamed, 2020 conducted in Egypt showed that 27% CAH children have emotional problems [11]. This is also supported with results of study conducted in 2020 by Medise et al. at RSCM Jakarta, Indonesia on 40 children with chronic illness, 6 children (15%) have emotional problems [15]. Mental emotional problems in children and adolescent were influenced by genetic, environment, friends and society [7]. Family parenting style will affects the emotional development of children. If the child grows in a family with positive emotions, the child's emotional development will be positive. In the opposite, if the parent expresses negative emotions, such as venting anger with an aggressive attitude, irritability, disappointment and pessimism in dealing with problems, then the child's emotional development will be negative [16].

In the behavioral disorder subscale, 7 children (25%) have behavioral disorders. This result is not much different from the results of Kung's study, 2018, which stated that as many as 23.8% CAH children have behavioral disorders [14]. Another study conducted by Idris, 2014 in Malaysia, it was found that 49 children with CAH had more emotional and behavioural problems than their unaffected relatives [17]. This behavioral disorder have a variety of adverse effects on children that can hinder them in the future. Behavioral problems that occur at a young age are thought to increase the risk of mental and behavioral disorders in middle age, so it is very important to detect and handle problems as early as possible [18].

From this study, most of the children (82.1%) do not have hyperactivity/inattention problems. In contrast with this, results of study conducted by Kung, 2018, in United Kingdom, where 19% of children have hyperactivity/inattention problems. Difference results in each country may be caused by cultural differences in health care also psychosocial support for children with chronic illness [14]. Another study in Turkey, conducted by Oner, 2009 mentioned that children and adolescent with CAH have higher aggressiveness scores [19]. Similar results was showed by Paterski, 2007 in Los Angeles. It mentioned that increase aggressivity and hyperactivity is only found in female children with CAH, while male CAH children

did not show significant result. The lack of behavioral changes in males with CAH is often attributed to feedback mechanisms that reduce testicular androgen production in response to the adrenal increase [20]. The form of hyperactivity in CAH children in this study were restlessness, overreacting, unable to remain silent for a long time and continuously moving restlessly and wriggling. However most children still have good attention and able to complete tasks or homework. According to Rahmadi, 2015, behavior that appears in hyperactive children is not able to sit quietly, always look restless, often leave the chair for no apparent reason, difficult to enjoy activities, always want to move actively and like to talk that is not in the context [21].

The results of the study on the subscale of relationship disorders with peers obtained as many as 13 children (46.4%) have peer problems. Form of peer problems in this study was tendency to be alone, playing alone and children were easier to make friends with adults than with peers. In line with this study, research conducted at United Kingdom by Kung, 2018 showed that 11.9% children with CAH have peer problems [14]. Peer relationship will affect emotion in children. If the child is well received by the peer group then pleasant emotions will become dominant, while if the child is rejected or ignored by the peer group then unpleasant emotions will become dominant to them [22].

This study showed that out of 28 children with CAH, most (85.7%) did not experience prosocial behavior issues. Prosocial behaviour is social behaviour that has the intent to benefit other people or society as a whole, such as helping, sharing, cooperating, obeying the rules and conforming to socially accepted behaviours. This results is in line with the results of research conducted by Shojaei, 2009, showed that children in Francis country, Ukraine and America have good prosocial abilities (80%), and only a small percentage of children who have prosocial behavior disorder [23].

## Conclusion

In summary, children with CAH have tendency to have psychosocial problems. Emotional, behaviour and peer problems were frequent happened in CAH children during pandemic Covid-19. Almost all CAH children do not have hyperactivity/inattention and prosocial behaviour problems.

There were some limitations in this study, such as the limitation of the number of samples studied. Future multicenter study is encouraged to ensure larger sample size to produce a rigorous study methodology. In addition, there were no data on psychosocial disorders in children with CAH before the pandemic, making it difficult to compare data on psychosocial disorders between before and during pandemic. However, this study can prove that there are psychosocial problems in CAH children especially during pandemic covid-19. In the future, clinicians need to screen all children with CAH for risk of psychosocial problems in order to early detection and intervention to minimize long-term adverse behavioural outcome that will affect children's growth and development.

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## Ethical Clearance

We sought approval of this research from the Clinical Research Unit of Dr. Soetomo Academic General Hospital Surabaya, Indonesia as our Ethical Committee Review Board. Before the subject recruitment, the explanation was done to the subjects and their parents.

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**Conflict of Interest – Nil**

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