

Research Article

Psychological Distress in Children with Surgically Corrected Congenital Heart Disease

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Received: 21 May 2020; **Accepted:** 05 June 2020; **Published:** 12 June 2020

Citation: Amalia Făgărășan, Flavia-Cristina Al-Akel, Andreea Cerghit-Paler, Brad Laura, Liliana Gozar. Psychological Distress in Children with Surgically Corrected Congenital Heart Disease. Journal of Pediatrics, Perinatology and Child Health 4 (2020): 042-051.

Abstract

Purpose/Aim/Objective: The purpose of this study was to investigate the neuropsychological modifications and intellectual functioning parameters (such as predictors of school performance – Intelligence quotient - IQ) of children with surgically corrected congenital heart defects. By investigating these changes, we might be able to develop a screening tool which facilitates the early detection of psychological distress, optimizes psychological therapy.

Methods: The study was conducted in the Paediatric Cardiology Clinic of Tîrgu Mureș, Romania, between 2016-2018, on 86 children with congenital heart disease (CHD), aged 6 to 17 years old, who have undergone surgical repair - compared to 102 healthy controls, matched by age. The study tested the hypothesis that children with CHD have lower intellectual function than the control group and analysed behavioural outcomes for these patients. Exclusion criteria: children aged less than 6 years old, confirmed genetic syndromes and a family history of psychiatric disorders.

Results: 59.3% of children in the eligible group were male, with a median age of 11.1 years \pm 3.2 SD. The average IQ of the patients with cyanotic CHD was 84.09, while the control group had an average IQ of 91.10. Analysis of the psychological profile indicated that 33.7% of children with surgically corrected CHD had anxiety, and 11.6% of them had attention deficit disorder.

Conclusions: Children with surgically corrected cyanotic CHD had a lower IQ than the control group. Anxiety and attention deficit disorder were more frequent in children with CHD.

Keywords: Intellectual Functioning; Congenital Heart Disease; Children; Anxiety

Abbreviations: IQ-Intelligence Quotient; CHD-Congenital Heart Disease; NYHA class-New York Heart Association class; DORV-Double Outlet Right Ventricle; MAPCA-Major Aorto-Pulmonary Collateral Artery; SD-Standard Deviation; CI-Confidence Interval; OR-Odds Ratio; PD-Psychological Distress

1. Introduction

The prevalence at birth of congenital heart diseases (CHD) has a varied geographical distribution, ranging from 9,3/1000 live births in Asia to 1,5/1000 in Africa and the high impact of morbidity and mortality at all stages of life is a major public health problem [1, 2]. Advances in prenatal and postnatal diagnosis, improved surgical techniques and the introduction of hybrid approaches have resulted in important decreased of the operative mortality in the past few decades [2-5]. Many children with complex CHD have neurodevelopmental and neuropsychological impairments and the impact on their quality of life is very important. Studies on the correlation between CHD and the neuropsychological behaviour of patients are currently being conducted

throughout the world in order to obtain the most accurate results that are useful for developing strategies, planning early and multilevel approach for the best social integration [6-8].

2. Material and Methods

Participants were recruited from a tertiary paediatric heart centre in Tîrgu Mures, Romania over a period of three years (2016-2018). 86 children, aged between 6 and 18 years old, who underwent surgical repair for CHD during this period were compared to 102 healthy controls. To obtain the study group, a sampling formula depending on the prevalence of CHD in Europe was used. The study group and the control group were matched by age. The primary outcome of the study was to test the hypothesis that children with CHD have reduced intellectual functioning (for example, the best predictor of school performance: IQ) when compared to a healthy population, and that the severity of CHD correlates with the degree of intellectual impairment. The secondary outcome was to analyse the behavioural patterns of children with surgically corrected CHD. Eligibility criteria: patients aged between 6 and 18 years-old with surgically corrected CHD, without any previous psychological evaluation. Exclusion criteria: patients younger than 6 years old, with confirmed genetic syndromes and/or psychiatric disorders in personal or family history.

The sample was split into three categories based on the severity of CHD, according to the classification of Task Force 1 of the 32nd Bethesda Conference and current functional status (NYHA class-New York Heart Association) [9]. First group - mild non-cyanotic models: corrected atrial septal defects, isolated ventricular septal defects. Second group - moderate non-cyanotic models: bicuspid aortic valve with stenosis, aortic regurgitation, subaortic stenosis, atrioventricular canal defects, pulmonary stenosis, aortic stenosis. The

third group - cyanotic models: tetralogy of Fallot, DORV (double outlet right ventricle)-type Fallot and MAPCA (major aortopulmonary collateral artery), Ebstein anomaly, anomalous pulmonary venous drainage, transposition of the great arteries. Several instruments have also been used in order to accomplish the psychological assessment. First, we evaluated the intellectual level by Raven's coloured progressive matrices for children aged 5 to 8 years-old and Raven's standard progressive matrices for children older than 9 years. Secondly, the clinical interview and observation were used to assess anxiety in children older than 6 years. Hamilton Depression Scale and Children's Depression Inventory test by Kovacs Maria PhD was used. The same psychologist assessed the children, after obtaining the written informed consent from parents. The University's Medical Ethics Committee approved this study. For statistical calculations, we used Graph Pad 3.6 State Software, San Diego, California, USA. Data were considered as nominal or quantitative variables. Nominal variables were characterized using frequencies. Quantitative variables were tested for normality of distribution using Kolmogorov-Smirnov test and were characterized by median and percentiles (25-75%) or by mean and standard deviation (SD), when appropriate. A chi-square test was used in order to compare the frequencies of nominal variables. Quantitative variables were compared using t test, or ANOVA test. We used the Bonferroni correction in order to account for multiple comparisons. The level of statistical significance was set at $p < 0.05$.

The odds ratio were computed to identify if children with operated CHD have different behaviour patterns compared to children belonging to the control group. The analysis was conducted based on age, sex and severity of CHD. The results were estimated considering a 95% confidence level. When situations with lack of significance and lack of computation

appeared, the conclusion was extracted based on 95% statistical significant confidence level (CI does not include 1) if found relevant. Three categories were considered: 1) the externalizing behaviour, comprising attention deficit, behaviour disorders; 2) the anxiety and 3) the depression.

3. Results

The study group included 86 subjects, of which 59.3% male, divided as follows: 24 into the mild non-cyanotic category, 18 into the moderate non-cyanotic category and 44 into the cyanotic category. Median age was 11.1 years \pm 3.2 SD. The Anova and the Bonferroni Correction tests were applied to analyse the IQ of the children in the control group and the IQ of the three risk models. One of the hypotheses of this study asserts that children with cyanotic CHD would have a lower IQ, and consequently lower school yield, compared to others (control group or the mild and moderate non-cyanotic). The results indicated that the mean IQ value in children with cyanotic CHD was 84.09, compared to the control group, with a mean IQ score of 91.10 ($p = 0.045$) (Figure 1), also considering the confidence intervals for the mean IQ scores (Table 1). When compared the IQ scores of children corresponding to the NYHA classes, no statistically significant difference was observed ($p = 0.95$) (Figure 2). A chi-square test was used to analyse the psychological profile for the study group. Data suggested that 33.7% of children with CHD had anxiety and 11.6% had attention deficit disorders, compared to the control group ($p = 0.0001$) (Figure 3). Analysing the three risk models in the study group appears that anxiety was similar in children with mild non-cyanotic defects (37.5%) and cyanotic CHD (35.6%) (Table 2). One of the hypotheses that could not be confirmed is the assumption that the female gender is more prone to neuropsychological disorders than male subjects ($p = 0.17$ and $p = 0.84$) (Figure 4 and 5).

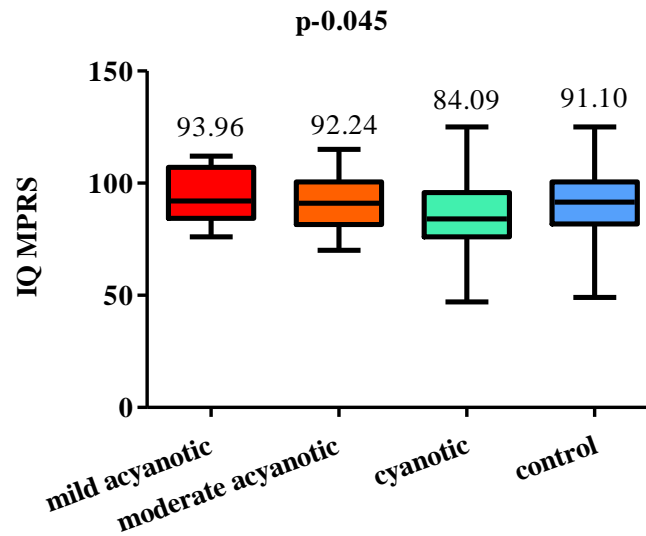


Figure 1: The mean IQ scores for control group and the risk models.

	Noncyanotic simple	Noncyanotic moderate	Cyanotic	Control
Number of values	24	17	44	102
Minimum	76,00	70,00	47,00	49,00
25% Percentile	84,25	81,50	76,00	81,75
Median	92,00	91,00	84,00	91,50
75% Percentile	107,0	100,5	95,75	100,5
Maximum	112,0	115,0	125,0	125,0
Mean	93,96	92,24	84,09	91,10
Std. Deviation	11,90	14,06	16,90	15,08
Std. Error of Mean	2,430	3,409	2,547	1,493
Lower 95% CI	88,93	85,01	78,95	88,14
Upper 95% CI	98,98	99,46	89,23	94,06

Table 1: The confidence intervals of the mean IQ for the studied groups.

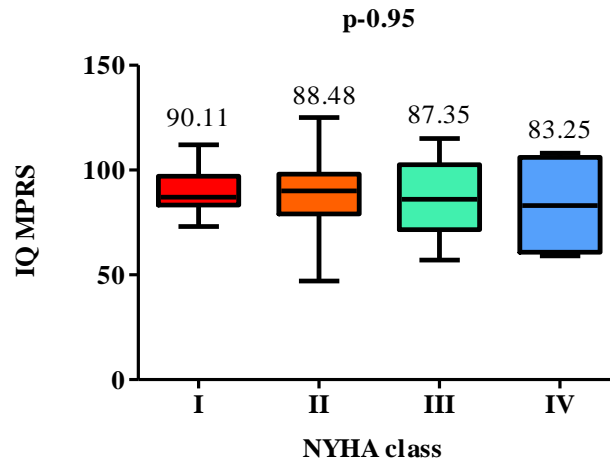


Figure 2: Statistical correlation between NYHA class and IQ values.

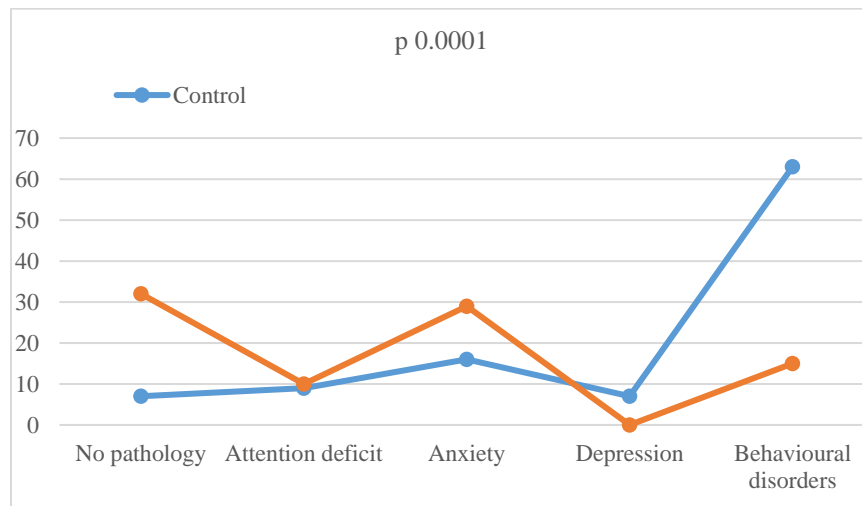


Figure 3: Comparison between the control group and the study group, according to psychological profile.

p-0.0001 chi-square test			CHD				Total
			cyanotic	control	moderate non-cyanotic	mild non-cyanotic	
Psychological distress (PD)	Anxiety	Number	16	16	4	9	45
		% PD	35.6%	35.6%	8.9%	20.0%	100.0%
		% CHD	35.6%	15.7%	23.5%	37.5%	23.9%
	Attention deficit	Number	6	9	1	3	19
		% PD	31.6%	47.4%	5.3%	15.8%	100.0%
		% CHD	13.3%	8.8%	5.9%	12.5%	10.1%
	Depression	Number	0	7	0	0	7
		% PD	0.0%	100.0%	0.0%	0.0%	100.0%

		% CHD	0.0%	6.9%	0.0%	0.0%	3.7%
No pathology	Number	20	7	9	3	39	
	% PD	51.3%	17.9%	23.1%	7.7%	100.0%	
	% CHD	44.4%	6.9%	52.9%	12.5%	20.7%	
Behavioral disorders	Number	3	63	3	9	78	
	% PD	3.8%	80.8%	3.8%	11.5%	100.0%	
	% CHD	6.7%	61.8%	17.6%	37.5%	41.5%	
Total	Number	45	102	17	24	188	
	% PD	23.9%	54.3%	9.0%	12.8%	100.0%	
	% CHD	100.0%	100.0%	100.0%	100.0%	100.0%	

Table 2: The psychological profile for the control group and the three subgroups of study batch.

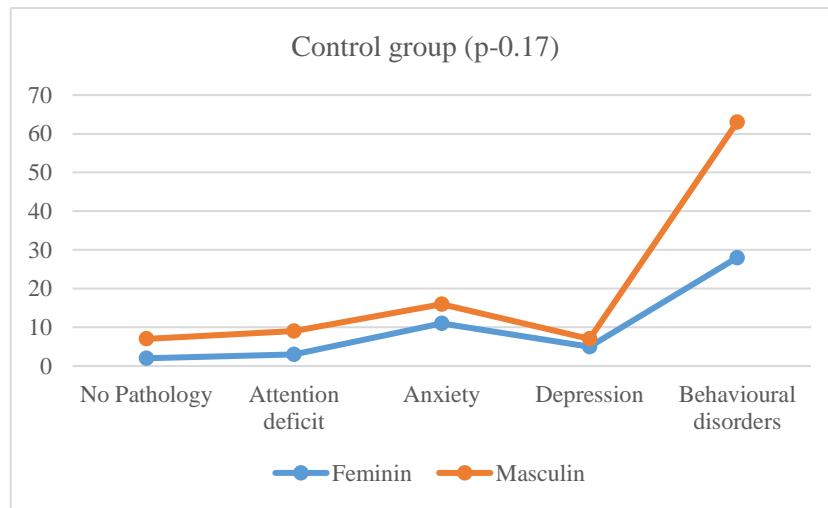


Figure 4: Correlation between sex and psychological profile in the control group (p=0.17).

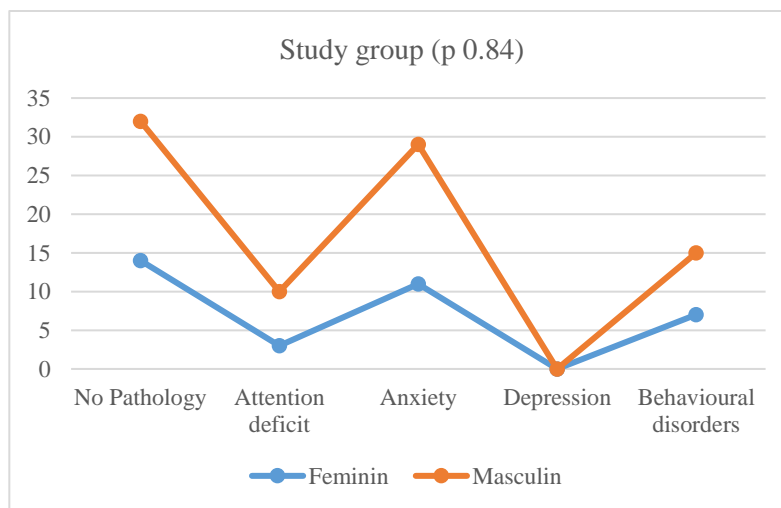


Figure 5: Correlation between sex and psychological profile in the study group (p=0.84).

Analysing the incidence of neuropsychological pathology by age groups, one can notice that anxiety and attention deficit are more frequently encountered in the age group of 6-10 years ($p=0.042$). In terms of odds ratio, when taking into account only the age category, children from the control group aged between 6 and 10 years old, and between 11 and 14 years old, are more likely to have externalizing behaviour (OR 10.28 with CI 3.47-30.40, respectively 5.18 with CI 1.81-14.77) than children with CHD, while children 6 to 10 years old from the study group are 8.28 times more likely to have anxiety behaviour (CI 1.74-39.39). When considering sex and age category, the results remained unchanged when comparing boys and girls between 6 and 10 years of age with surgically corrected CHD with similar age and sex children from the control group, as they are less likely to present externalizing behaviour. The odds ratio for the control group is 9 (CI 2.39-33.86) for 6-10 years old boys and 13.5 (CI 2.00-90.69) for 6-10 years old girls in terms of externalizing behaviour. The externalizing behaviour was more frequent in boys from the control group, 11 to 14 years old, with odds ratio 5.5 (CI 1.39-21.68). When considering sex, age category and severity of CHD, some changes were observed, while other results remain unchanged. Moderate non-cyanotic boys 6 to 10 years old are 13.5 times more likely (CI 1.34-35.98) to have anxiety behaviour compared to the control group. Analysing cyanotic group, both boys and girls 6 to 10 years old, and 11 to 14 years old children from the control group are more likely to have externalizing behaviour than other risk models matched by sex and age. The odds ratios are 10.5 (CI 2.33-47.20) for boys 6-10 years old, 9.16 (CI 1.41-59.58) for boys 11-14 years old, 27 (CI 1.97-368.4) for girls 6-10 years old and 10.28 (CI 1.01-103.95) for girls 11-14 years old.

4. Discussions

The clinical relevance of the impact of operated CHD on neuropsychological development is helpful in everyday practice. IQ measurement is required for psychoeducational assessment, being an adequate predictor of difficulties in adapting to the school system. Focused studies have shown suboptimal values and the need of special educational programs for children with surgically corrected CHD [10-12]. Among factors that influence IQ score are biological parental IQ, parental social status, medical conditions and how each factor participates is debatable. Generally, patients with CHD are at risk for developmental disorders, this risk is multifactorial and probably synergistic [13-15]. In our study, the mean IQ score of children undergoing surgery due to cyanotic CHD was below average (84.09), lower than IQ scores in the control group ($p=0.045$). Statistical correlation between IQ score and NYHA class did not yield a significant result ($p=0.95$). Studies focused on possible factors involved in the impaired cognitive development of children with severe CHD have shown the role of reduced cerebral perfusion before and after surgery, prolonged hypoxia, exposure to anesthesia and repeated surgical interventions [15-18]. Pizarro C. et al. did not identify a clear correlation between the utilized surgical techniques and children's intellectual functioning [19]. In their study on 228 Swedish children with CHD, treated either by a surgical or an endovascular approach, Ryberg C. et al. had similar results on overall intellectual functioning [20]. Anxiety disorder is generally described in children with chronic conditions because of exposure to stress caused by frequent hospitalization, and the physical limitations imposed by the illness itself. The outcome/consequences of these disorders are the tendency for isolation and a sense of being rejected by their peers [21]. Johnson B, showed that children with CHD have more behavioural problems compared to their healthy counterparts [22]. Analysis of the psychological and

emotional profile of patients included in our study showed that 33.7% of children with operated CHD had anxiety, compared to the control group where anxiety was present in 15.7% of the cases ($p=0.0001$) and 11.6% of children in the study group had attention deficits. On the other hand, the odds ratios revealed that both boys and girls from the control group, aged 6 to 10 years old, and 11 to 14 years old respectively, are more likely to have an externalizing behaviour than children with cyanotic CHD, matched by age and sex. Subgroup analysis revealed that the frequency of anxiety was similar among children with mild non-cyanotic defects (37.5%) and cyanotic models (35.6%), meaning that the cardiac condition is a trigger for anxiety, especially for moderate non-cyanotic boys 6 to 10 years old. Correlation of NYHA class severity (as a distress trigger due to the inability to participate in intensive physical activities), of IQ and of psychological and emotional disorders did not appear statistically significant ($p=0.10$). Behavioural disorders were more frequent in the control group (61.8%). Adolescence has been described as a particularly vulnerable period for children with CHD, and has been associated with a higher risk of developing psychological and emotional disorders [23-26]. In our study anxiety and attention deficits are more frequently encountered in the age group of 6-10 years old ($p=0.042$) and the lack of specialized intervention for this age group will aggravate the disorder. It is accepted that there is a need for the adequate assessment and neuropsychological care of children with CHD based on risk categories, in order to develop an Individualized Education Program which improves the quality of life of these patients.

Study limitations: a relatively small cohort, but comprising all patients in the central part of Romania (exhaustive research). A limitation of the study is the lack of correlation of IQ score with parental social status or parental IQ score, or correlation of age at the

time of surgery and IQ score or psychological and emotional changes.

5. Conclusions

Children with surgically corrected cyanotic congenital heart disease have lower IQ scores than the control group. Anxiety and attention deficit disorders were more frequent in children with CHD.

Acknowledgements

The authors thank Anca Gherasim for her careful psychological assessment.

Funding

No funding was received.

Conflicts of Interests

The authors declare that they have no conflict of interest.

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