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PSYCHOSOCIAL ADJUSTMENT AND PRONENESS TO PSYCHOPATOLOGY IN ADOLESCENTS AND YOUNG ADULTS WITH CONGENITAL HEART DISEASE

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PSYCHOSOCIAL ADJUSTMENT AND PRONENESS TO PSYCHOPATOLOGY IN ADOLESCENTS AND YOUNG ADULTS WITH CONGENITAL HEART DISEASE

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ABSTRACT

Objectives: Our purpose was to study psychosocial adjustment and psychiatric morbidity of adolescents and young adults with congenital heart disease (CHD).
Design: All assessment measures were obtained on a single occasion in patients with a diagnosis of Congenital Heart Disease coming to consultation in a tertiary hospital.
Participants: We evaluated 110 CHD patients (62 male) aged from 12 to 26 years old (mean=18.00 ± 3.617), 58 cyanotic.
Primary and secondary outcome measures: All assessment measures were obtained on a single occasion. Demographic information and clinical history were collected. Questionnaires regarded topics as social support, family educational style, self-image and physical limitations, a standardized psychiatric interview SADS-L, and a self-report questionnaire on psychosocial adjustment, YSR or ASR. One of the relatives completed an observational version of the same questionnaires (CBCL or ABCL).
Results: We found a 21.8% lifetime prevalence of psychopathology, 31.3%, in females, 14.5% in males, showing a somewhat increased proneness in CHD patients. Females also showed worse psychosocial adjustment, with more somatic complaints (u=260.000; p=0.011), anxiety/ depression (u=984.000; p=0.002), aggressive behavior (u=920.500; p=0.001), attention problems (u=1123.500; p=0.027), thought problems (u=1069.500; p=0.010), internalization (u=869.000; p=0.000) and externalization (u=1163.000; p=0.050). Patients with severe CHD (u=939.000; p=0.030) and surgical repaired (u=719.000; p=0.037) showed worse psychosocial adjustment. Those with poor social support showed more withdrawn (u=557.500; p=0.000) and social problems (u=748.500; p=0.023), and patients with unsatisfactory school performance revealed more anxiety/depression (u=916.000; p=0.020) and attention problems (u=861.500; p=0.007).
Conclusions: CHD males with good social support and good academic performance have a better psychosocial adjustment.

Key Words: Congenital Heart Disease, Psychosocial Adjustment, Psychopathology

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Abbreviations: CHD - congenital heart disease; SADS-L - Schedule for Affective Disorders and Schizophrenia – Lifetime version; YSR - Youth Self Report; CBCL - Child Behavior Checklist; ASR - Adult Self Report; ABCL - Adult Behavior Checklist

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ARTICLE SUMMARY

The study focuses in:
1. studying psychiatric morbidity and psychosocial adjustment in adolescents and young adults with congenital heart disease, comparing them with healthy population.
2. investigating which demographic, psychosocial and clinical variables contribute to foster resilience and adaptation and which ones have detrimental effects.
Our research is an important contribution to the state-of-the-art on this scientific issue because it systematically tests the contribution of a wide range of demographic, psychosocial and clinical variables in psychosocial and emotional adjustment and psychiatric morbidity.
Strengths: 110 patients were evaluated; A wide sellection of demographic, psychosocial and clinical variables were assessed; we used a very strict method for evaluating psychiatric morbidity and lifetime prevalence of psychpathology, a structured psychiatric interview.
Limitations: The fact that there are no nationwide normative studies of lifetime prevalence of psychopathology in Portugal limits the possibility to compare the prevalence rate in our patients with the general population, and to make clear inferences about it.

INTRODUCTION

The survival rate in the 1950's for children born with moderate CHD was about 20% whereas today about 90% of these children achieve adulthood.[1]
There has been a decrease in child mortality thanks to advances over the last four decades in diagnostic, surgical and catheter interventional techniques.[2] As these children survive, the interest in issues such as psychosocial outcomes have increased also.[3]
Most children with CHD were diagnosed in uterus or in infancy, and are expected to undergo surgical procedures either to correct or palliate their defect.[4, 5] These children need to be seen regularly by a cardiologist.[4]
Many studies have been conducted assessing the impact of CHD on children's or adolescents' psychosocial and cognitive functioning. Although, a consensus among these studies have not yet been reached, some report higher rates of behaviour problems in children and adolescents with CHD, while others have not found any differences between patients with CHD and norms.[4]
It is believed that children with CHD have a higher risk of developing behavioural and emotional problems, when compared to healthy children. Several studies have reported that these children have increased feelings of anxiety and inferiority, higher degrees of impulsiveness, higher levels of emotional and behavioural problems.[6]
On the other hand, in European studies have showed a good psychological functioning in adults with CHD.[7]
Not much is known about this topic, as some studies say that, in a 25-year follow-up, more psychosocial distress was found in adults with CHD in comparison with a normative group. The differences found were limited to somatic complaints and thought problems and behaviours.[6]
As for psychopathology, studies have also disagreed in some aspects, many authors

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believe that CHD patients have a higher probability of having psychopathological symptoms while others have found similar numbers between these patients and healthy children and adolescents.[8]
Several characteristics can be described as facilitators of positive perspectives of stressful life situations and reduced psychological distress such as self-esteem and similar conduct like self-concept and self-perception. Some studies found that usually CHD patients have lower self-esteem although, after surgery, patients reported better self-esteem or self-concept.[9]
Cognitive perceptions are believed to have an influence on a CHD patient's life. The more negative these perceptions are, the higher psychological distress was found. The negative perception can be associated, more than the severity of the disease itself, to higher distress and worse psychological adjustment.[9]
Some studies have shown that patients with cyanotic heart disease have a higher risk of presenting behavioral problems compared to patients with non-cyanotic heart defects, but other studies did not show this association.[5, 10]
Patients with CHD who underwent surgical procedures, had more behavioral problems when compared with those who did not require surgery, and more likely to develop psychiatric problems.[5]
As far as the physical condition is concerned, most patients with CHD have limitations, that leading to more behavioral and emotional problems.[5, 8]
In this study, we aimed to evaluate psychosocial adjustment and proneness to psychopathology in adolescents and young adults with CHD. The importance of our investigation is that it systematically addressed the question of how the several demographic and clinical variables relate to psychiatric morbidity and to psychosocial adjustment, using very strict methods of psychiatric diagnosis.

METHODS

Participants

The study enrolled 110 CHD patients, 62 male and 48 female, with a mean age of 8.00 ± 3.62 years (range:12-26 years old). The participants who had not achieved an educational level that enabled them to understand and complete the written questionnaires were excluded from the study.
At the time of the interview, two participants were married, one was divorced, two were living in a marital union. All the others (105) were single.
53 patients had completed their secondary education (12th grade), 40 the 3rd cycle (9th grade), 11 the 2nd cycle (6th grade) and 6 had graduated from college. Of these patients, 55 had at least repeated one year at school (mean=1,49 ± 0,50 year).
Of the 110 participants, 20 were employed full- or part-time, 7 were unemployed and all the others 83 were students.
Complete medical records were available for all the patients, who had been followed in the pediatric cardiology or cardiology departments of a tertiary hospital.
For 58 individuals the CHD was cyanotic and for 52 it was acyanotic; 34 of these patients had a severe form of CHD, 18 a moderate and 58 a mild one; 41 patients had some physical limitations while 69 did not. 4 patients had severe residual lesions, 21 moderate and 85 mild lesions. 23 patients were never submitted to any kind of surgical procedure, while 42 had 1 surgery, 25 had two, 11 had three, 5 had four, 3

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had five and 1 had 9 surgeries. 47 patients were on pharmacological therapy while 63 were not.

In many of our participants, the main CHD was combined with other heart diseases. Patients with associated cardiac malformations or chromossomopathies were excluded from the study. The participants had the following distribution of pathologies: transposition of the great arteries (9; two of them had also ventricular septal defect and aortic stenosis, and one had ventricular septal defect and pulmonary stenosis), tetralogy of Fallot (30), coarctation of the aorta (11; one had also ventricular septal defect and one aortic stenosis) ventricular septal defect (24; one had also interruption of the aortic arch and one had mitral insuficiency), atrial septal defect (6; one had also mitral atresia and pulmonary hypertension, and one had Ebstein disease), atrioventricular septal defect (4), aortic stenosis (6), pulmonary stenosis (6), single ventricle (2; one had also pulmonary atresia and one had pulmonary stenosis), patent ductus arteriosus (2), double-outlet right ventricle (1), pulmonary atresia (3), Ebstein disease (3), mitral valve prolapse (1), bicuspid aortic valve (1) and tricuspid valve regurgitation (1).

The diagnosis was determined during the neonatal period for 61, before the first birthday for 28, 5 were diagnosed between the ages of 1 to 3 years, 6 were diagnosed between the ages of 3 to 6 years and between the ages of 6 and 12 for 12 participants.

The first surgery was performed for 5 of the participants during the neonatal period, before the first birthday for 30, between the ages of 1 and 3 for 19, and between the ages 3 and 6 for 20 participants, between the ages 6 to 12 for 8 and between the ages of 12 to 18 for 28.

We also invited one relative of each patient to participate in this study and 100 accepted to take part in it.

Assessment Instruments

In this study, we used different surveys to collect the necessary information: an identification form, a semi-structured interview, a standardized psychiatric interview SADS-L, the self-report and observational questionnaires of the ASEBA system for psychosocial adjustment, ASR, YSR, ABCL and CBCL (for patients > 18 and < 18). Additional questionnaires used in this research are described in detail in another report.

We used an identification form to collect personal and demographic data from each patient (e.g., marital status, educational level and occupation), as well as all relevant aspects from their medical history (diagnosis, severity and category of heart disease, surgical interventions, pharmacological therapy, and presence of residual lesions, among others).

The semi-structured interview included 38 multiple-choice or short-answer questions that focused on different topics such as social support, family upbringing, self-image, functional limitations and emotional adjustment.

A standardized psychiatric interview, SADS-L (Schedule for Affective Disorders and Schizophrenia – Lifetime version) [11], was administered to obtain a clinical diagnosis of any psychopathological disorders that may have existed before the interview in these patients.

The YSR and ASR are self-report questionnaires, designed to collect a description of a child or adult's functioning; they assess individuals in scales of withdrawn behavior, somatic complaints, anxiety/depression, thought problems, social problems, attention

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problems, delinquent behavior, aggressive behavior, internalization and externalization.[12] The CBCL and ABCL are observational versions of the same questionnaires, to be completed by the patients' parents or caregivers, having as a requirement being knowledgeable about the patient, as they report their perception on the behavior and possible problems occurring in the patient. For their similarities, and to have a better representative sample, the results of the YSR and ASR were pooled, as well as the results of the CBCL and the ABCL, and for statistical purposes the overall results were counted for each scale.

Procedure

Prospective participants were contacted while waiting for their appointment in cardiology or pediatric cardiology department. Our first concern was to get their or their parents' consent (when they were under 18 years old). At this time, they were informed about all aspects of the research, and when they accepted to participate, they completed an informed consent form approved by the hospital's ethical committee, which followed international conventions guaranteeing the rights of the patients. The interview happened on the spot. The parents or caregivers accompanying the patient were asked to fill out a questionnaire, and 10 caregivers refused to participate or were not present for the application of the protocol, and subsequently expressed their intention not to participate.

Design

All the assessment measures were obtained on a single occasion. Clinical data were collected retrospectively using each patient's clinical record, with assistance from hospital medical staff.

Data Analysis

Statistical analysis of the data was processed using the software IBM SPSS (Statistical Package for the Social Sciences, Chicago, IL, USA), version 19. The distribution of all the variables was tested. Differences for parametric variables were established using Student's *t*-tests, while differences for non-parametric variables (the majority) were established using Mann-Whitney *U* test and Chi-square tests of association.

RESULTS

We found that 21.8% of our participants had a psychiatric disorder and that there was a statistical difference between the two genders, with females almost doubling males' rate (31.3% in females and 14.5% in males; p=0.035). One or more of the following psychiatric disorders had been diagnosed for our participants in all their lifetime prior to interview: Minor or Major Depressive Syndrome (13), Panic Disorder (3), Anxiety Disorder (4), or Manic Syndrome (3), Cyclothymic Personality (1).

For the sake of data analysis, we grouped the results on psychosocial adjustment in either self-reported or observational.

The self-report measures on psychosocial adjustment revealed statistical differences

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between genders, as females showed higher levels of somatic complaints (u=260.000; p=0.011), anxiety/ depression (u=984.000; p=0.002), aggressive behavior (u=920.500;
p=0.001), attention problems (u=1123.500; p=0.027), thought problems (u=1069.500; p=0.010), internalization (u=869.000; p=0.000) and externalization (u=1163.000; p=0.050) and overall worse psychosocial adjustment than males.

On the other hand, teens and young adults with a severe CHD showed worse psychosocial adjustment, with more somatic complaints (u=264.000; p=0.022) and higher levels of internalization (u=917.000; p=0.015) in self-report measures, when compared with patients with moderate-to-mild CHD.

When analyzing the impact of the kind of CHD in psychosocial adjustment, no statistical differences were found.

Patients who underwent surgical interventions revealed worse psychosocial adjustment than patients with no surgical repairs, showing higher levels of withdrawn behavior (u=719.500; p=0.037) in self-assessment.

Patients with poor social support reported worse psychosocial adjustment, with higher levels of withdrawn (u=557.500; p=0.000) and social problems (u=748.500; p=0.023) when compared with patients with good social support.

Patients with limited physical competence showed more withdrawn behavior (u=1023.000; p=0.015) when compared to patients with satisfactory physical competence, thus presenting worse adjustment.

Patients with worse academic performance showed higher levels of anxiety/depression (u=916.000; p=0.020) and attention problems (u=861.500; p=0.007) in self-report, when compared to those who feel their academic performance was satisfactory.

Patients with severe-to-moderate residual lesions revealed worse psychosocial adjustment in self-report than those with mild residual lesions, showing higher levels of internalization (u=782.500; p=0.046).

No differences were found in self-report between patients with or without need for pharmacological therapy.

According to the assessment of patients' caregivers, no differences were found between patients who underwent surgical procedures and the ones who did not had surgical procedures done.

In the caregivers' assessment, male patients are perceived as having worse psychosocial adjustment than females, as they were assessed as having higher levels of withdrawn (u=911.500; p=0.020) and aggressive behaviors (u=945.500; p=0.038).

Again on the relatives' standpoint, the cyanotic patients were assessed as having higher levels of attention problems (u=981.500; p=0.045) than the acyanotic, thus showing worse psychosocial adjustment.

When compared with patients with good social support, those with poor support showed, also on the stand point of the caregivers, higher levels of withdrawn (u= 517.000; p=0.001) and internalization (u=608.000; p=0.007) and thus, worse psychosocial adjustment. According to their relatives' assessment, patients with severe to moderate forms of residual lesions showed higher levels of social problems (u=205.500; p=0.008), attention problems (u=649.500; p=0.028) and internalization (u=567.500; p=0.004), than the ones with moderate to mild residual lesions, and thus a worse psychosocial adjustment.

DISCUSSION

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This study is important because it systematically tested the effects of different demographic, clinical and psychosocial variables in psychosocial adjustment and psychiatric morbidity. In a growing population of adults with CHD, this information is rather important in unveiling strategies that can be used to assist and care for patients, leading to better emotional adjustment and better outcomes in different life challenges.

The assessment instruments used for psychosocial adjustment enabled us to compare sub-groups of patients, regarding demographic, clinical and psychosocial variables. One main finding of our study was that females with CHD reported higher levels of somatic complaints, anxiety/depression, thought problems, attention problems, aggressive behavior, internalization and externalization, than males, thus showing a worse psychosocial adjustment. Contrariwise, relatives find male patients more withdrawn than girls.

In our study, female patients had almost the double of the lifetime prevalence of psychopathology than males. These findings on psychiatric morbidity and on the different scales of psychosocial adjustment are consistent with other studies on the general population that report differences between genders, with females showing higher rates of emotional problems. Studies show also that females have greater likelihood of displaying higher levels of anxiety/depression and somatic complaints when facing negative obstacles that interfere with the interpersonal level, resulting in higher levels of internalization.[6,8,13]

This may be due to the presence of a scar, situated on the chest, being a source of uncertainties or discomfort. In addition to affecting sexual relationships CHD can also interfere with pregnancy and delivery, leading to a sense of anxiety about their physical condition.[6,8]

Some studies show that females are more likely to develop depressive symptoms when facing negative life events than males.[6,10]

In this study, adolescents or young adults with severe type of CHD reported having higher levels of social problems and, thus, worse psychosocial adjustment, compared with those with moderate or mild form of CHD.

These results may be related to the fact that they need further medical care throughout their life, while patients with mild or moderate CHD may have a daily life similar to healthy adolescents and young adults.[8] Patients with severe forms of CHD show higher level of internalization and somatic complaints and that may be associated to the fact that these patients are more vigilant about their health, being more anxious about any complications. This may explain the results, since anxiety is a component of internalization scale.[14]

The type of CHD did not show any impact with statistical relevance in patients' self-report measures of psychosocial adjustment. However, the caregivers' standpoint seems to be more sensitive regarding this feature, as they perceive the cyanotic patients as having more attention problems and worse psychosocial adjustment.

Other published studies also showed that the cyanosis is not a stable indicator that patients will have behavioral and emotional problems.[8,10,15]

Patients who underwent surgical procedures revealed higher levels of withdrawn behavior. This may be related with the fact that admissions are long as well as the recovery, thus providing a prolonged absence from education and from contact with the peer groups, which could lead to difficulties of reintegration and therefore to the isolation of patients.[5,7,16]

Patients with worse social support had higher levels of withdrawn behavior and social

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problems, and thus, a worse psychosocial adjustment. The assessment of the caregivers also reported higher levels of withdrawn behavior and internalization in patients with poor social support, showing worse psychosocial adjustment.

According to several studies, parents and siblings of adolescents or young adults with CHD are more prone to face a different number of psychosocial stresses putting the whole family in need of psychosocial support. Many studies reveal a higher need for intervention on family problems in families with children with chronic medical diseases. When the complexity of the disease is low, parents seem to be more fitted to provide support.[17] These families are reported to experience more stress, that can have an impact on the child's adjustment.[15]

Parents of children with CHD can be overprotective and hyper vigilant about their child's health, making it hard for their children to be more independent. Many studies show that these patients are more likely to have "dependent lifestyles" than healthy adolescents or young adults.[7] The participation in leisure time activities can be a contributor to a better social outcome.[13]

Limited physical competence translated into more withdrawn, feeling more isolated, when compared with patients with satisfactory physical competence. Self-report showed that patients with physical limitations have worse psychosocial adjustment. A low exercise capacity can be translated into more internalizing problems. For older heart patients, limited physical competence lead to concerns and anxiety about their health.

According to some authors, patients submitted to physical training interventions, showed a decrease in internalizing problems.[8]

Physical limitations and school absences prevent full participation in different activities, leading to isolation and social awkwardness. This can be translated into restricted employment opportunities.[7]

In our study, an unsatisfactory academic performance led to worse psychosocial adjustment, as patients report having higher levels of anxiety/depression, attention problems and externalization than those with good academic performance. Several previous published studies show that CHD has an impact on school careers, for the many hospitalizations and restrictions, being the main reason for the attendance of special education by these patients. When compared to healthy adolescents or young adults, the CHD patients are more unlikely to complete a lower educational level.[13]

Sometimes, children with CHD have neurodevelopment deficits. These often will not show until school age, when the academic demands start having an impact on their lives. Many families rationalize their child's developmental delay to the disease and the several hospitalizations.[18]

Some studies show that unsatisfactory educational background can be translated into lower educational and occupational achievement.[7]

This study showed a 21.8% prevalence of psychiatric disorder in our patients. Females showed a higher percentage of psychiatric disorder with 31%, and males only had 14%.

When compared to the reference value of the World Health Organization (WHO), 10% of the world population, it seems that adolescents and young adults with CHD have an increased proneness for psychiatric diagnosis.[19] However, a study of six different European countries showed a prevalence of 25% in the general population, which is closer but higher than the results for CHD patients in our study.[20] Another study estimated that the life time prevalence of psychopathology is 19.4% in Spain, 18.1% in Italy (countries that can be considered culturally close to Portugal), and 25.2% in Germany, but in striking contrast, 37.9% in France and 47.4% in the United

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States of America.[21]

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COMPETING INTERESTS
There are no competing interests.

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DATA SHARING STATEMENT

I am available to share any still unpublished data from the study. Any people interested may contact me to metega@sapo.pt
Maria Emília Areias (corresponding author)

CONTRIBUTORSHIP STATEMENT
Authors 1, 2 and 3 are last year students of the masters in clinical and health psychology; they performed the field work, interviewing participants and their relatives, they encoded data and performed the statisctical analysis; author number 1 revised the state-of-the-art about this subject matter and wrote the article.
Author 4 is pediatric cardiologist and advised in collecting clinical data of participants from files and interpreting them.
Author 5 is clinical psychologist; he advised on the methods and strategies for statistical analysis.
Author 6 is pediatric cardiologist and he contributed in planning the research, in all the clinical aspects and procedures in the department.
Author 7 is clinical psychologist and the supervisor of the master students (authos 1, 2 and 3); she planned the design of the study, the methods, chose the assessment instruments to be used, she planned the steps of the statistical analysis, she reviwed the state-of-the-art of this subject matter, and revised this manuscript.

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STROBE 2007 (v4) Statement—Checklist of items that should be included in reports of *cross-sectional studies*

Section/Topic	Item #	Recommendation	Reported on page #
Title and abstract	1	(*a*) Indicate the study's design with a commonly used term in the title or the abstract	Yes (Abstract/ Methods)
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found	Yes (Abstract/ Methods/ Results)
Introduction			
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	Yes (Introduction)
Objectives	3	State specific objectives, including any prespecified hypotheses	Yes (Abstract/ Introduction/ Methods)
Methods			
Study design	4	Present key elements of study design early in the paper	Yes (Abstract/ Methods)
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	Yes (Abstract/ Methods)
Participants	6	(*a*) Give the eligibility criteria, and the sources and methods of selection of participants	Yes (Methods, Participants)
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	Yes (Methods/ Results)
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	Yes (Methods/ Results)
Bias	9	Describe any efforts to address potential sources of bias	*Yes (Methods)*
Study size	10	Explain how the study size was arrived at	Yes (Methods)
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why	Yes (Methods)
Statistical methods	12	(*a*) Describe all statistical methods, including those used to control for confounding	Yes (Methods; Methods of

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			Statistical Analysis)
		(*b*) Describe any methods used to examine subgroups and interactions	Yes (Methods)
		(*c*) Explain how missing data were addressed	….
		(*d*) If applicable, describe analytical methods taking account of sampling strategy	….
		(*e*) Describe any sensitivity analyses	….
Results			
Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	There is only one stage
		(b) Give reasons for non-participation at each stage	Yes (Methods)
		(c) Consider use of a flow diagram	No
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	Yes (Methods/ Results)
		(b) Indicate number of participants with missing data for each variable of interest	The only missing are some relatives of patients) who did not accept to participate in the study
Outcome data	15*	Report numbers of outcome events or summary measures	Yes (Methods/ Results)
Main results	16	(*a*) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	…..
		(*b*) Report category boundaries when continuous variables were categorized	Yes (Methods/ Results)
		(*c*) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	……
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	Yes (Methods/ Results)
Discussion			
Key results	18	Summarise key results with reference to study objectives	Yes (Results/ Discussion)

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Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias	Yes (Discussion)
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	Yes (Discussion)
Generalisability	21	Discuss the generalisability (external validity) of the study results	Yes (Discussion)
Other information			
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	Yes (Funding/ Aknowledgements)

*Give information separately for cases and controls in case-control studies and, if applicable, for exposed and unexposed groups in cohort and cross-sectional studies.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at http://www.plosmedicine.org/, Annals of Internal Medicine at http://www.annals.org/, and Epidemiology at http://www.epidem.com/). Information on the STROBE Initiative is available at www.strobe-statement.org.

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PSYCHOSOCIAL ADJUSTMENT AND PSYCHOPATHOLOGY IN ADOLESCENTS AND YOUNG ADULTS WITH CONGENITAL HEART DISEASE

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PSYCHOSOCIAL ADJUSTMENT AND ~~(PRONENESS TO)~~ PSYCHOPATOLOGY IN ADOLESCENTS AND YOUNG ADULTS WITH CONGENITAL HEART DISEASE

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Key Words: Congenital Heart Disease, Psychosocial Adjustment, Psychopathology

Word Count: 4027 with references and abstract; 3177 without references and Abstract

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PSYCHOSOCIAL ADJUSTMENT AND ~~(PRONENESS TO)~~ PSYCHOPATOLOGY IN ADOLESCENTS AND YOUNG ADULTS WITH CONGENITAL HEART DISEASE

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ABSTRACT

Objectives: Our purpose was to study psychosocial adjustment and psychiatric morbidity of adolescents and young adults with congenital heart disease (CHD).

Methods: We evaluated 110 CHD patients (62 male) aged from 12 to 26 years old (mean=18.00 ± 3.617), 58 cyanotic. All assessment measures were obtained on a single occasion in a tertiary hospital. Demographic information and clinical history were collected. Questionnaires regarded topics as social support, family educational style, self-image and physical limitations, a standardized psychiatric interview SADS-L, and a self-report questionnaire on psychosocial adjustment, YSR or ASR. One of the relatives completed an observational version of the same questionnaires (CBCL or ABCL).

Results: We found a 21.8% lifetime prevalence of psychopathology, 31.3%, in females, 14.5% in males, showing a somewhat increased proneness in CHD patients. Females also showed worse psychosocial adjustment, with more somatic complaints (u=260.000; p=0.011), anxiety/ depression (u=984.000; p=0.002), aggressive behavior (u=920.500; p=0.001), attention problems (u=1123.500; p=0.027), thought problems (u=1069.500; p=0.010), internalization (u=869.000; p=0.000) and externalization (u=1163.000; p=0.050). Patients with severe CHD (u=939.000; p=0.030) and surgical repaired (u=719.000; p=0.037) showed worse psychosocial adjustment. Those with poor social support showed more withdrawn (u=557.500; p=0.000) and social problems (u=748.500; p=0.023), and patients with unsatisfactory school performance revealed more anxiety/depression (u=916.000; p=0.020) and attention problems (u=861.500; p=0.007).

Conclusions: Our study emphasizes the need for evaluating the psychosocial adjustment of adolescents and young adults with CHD.

Comment [U3]: Reviewer 2: we deleted "CHD males with good social support and good academic performance have a better psychosocial adjustment."

Comment [U4]: Reviewer 2: new sentence for conclusions

Key Words: Congenital Heart Disease, Psychosocial Adjustment, Psychopathology

Word Count: 4027 with references and abstract; 3177 without references and abstract

Abbreviations: CHD - congenital heart disease; SADS-L - Schedule for Affective Disorders and Schizophrenia – Lifetime version; YSR - Youth Self Report; CBCL - Child Behavior Checklist; ASR - Adult Self Report; ABCL - Adult Behavior Checklist

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INTRODUCTION

The survival rate in the 1950's for children born with moderate CHD was about 20% whereas today about 90% of these children achieve adulthood.[1]

There has been a decrease in child mortality thanks to advances over the last four decades in diagnostic, surgical and catheter interventional techniques.[2] As these children survive, the interest in issues such as psychosocial outcomes have increased also.[3]

Most children with CHD are diagnosed in uterus or in infancy, and are expected to undergo surgical procedures either to correct or palliate their defect.[4, 5] These children need to be seen regularly by a cardiologist.[4]

Many studies have been conducted assessing the impact of CHD on children's or adolescents' psychosocial and cognitive functioning. Although, a consensus among these studies have not yet been reached, some report higher rates of behaviour problems in children and adolescents with CHD, while others have not found any differences between patients with CHD and norms.[4]

It is believed that children with CHD have a higher risk of developing behavioural and emotional problems, when compared to healthy children. Several studies have reported that these children have increased feelings of anxiety and inferiority, higher degrees of impulsiveness, higher levels of emotional and behavioural problems.[6] On the other hand, some European studies have showed a good psychological functioning in adults with CHD.[7]

Not much is known about this topic, as some studies say that, in a 25-year follow-up, more psychosocial distress was found in adults with CHD in comparison with a normative group. The differences found were limited to somatic complaints and thought problems and behaviours.[6]

As for psychopathology, studies have also disagreed in some aspects, many authors believe that CHD patients have a higher probability of having psychopathological symptoms while others have found similar numbers between these patients and healthy children and adolescents.[8]

Several characteristics can be described as facilitators of positive perspectives of stressful life situations and reduced psychological distress such as self-esteem and similar conduct like self-concept and self-perception. Some studies found that usually CHD patients have lower self-esteem although, after surgery, patients reported better self-esteem or self-concept.[9]

Cognitive perceptions are believed to have an influence on a CHD patient's life. The more negative these perceptions are, the higher psychological distress was found. The negative perception can be associated, more than the severity of the disease itself, to higher distress and worse psychological adjustment.[9]

Some studies have shown that patients with cyanotic heart disease have a higher risk of presenting behavioral problems compared to patients with non-cyanotic heart defects, but other studies did not show this association.[5, 10]

Patients with CHD who underwent surgical procedures, had more behavioral problems when compared with those who did not require surgery, and more likely to develop psychiatric problems.[5]

As far as the physical condition is concerned, most patients with CHD have limitations, ~~that leading to~~which can lead to more behavioral and emotional problems.[5, 8]

In this study, we aimed to evaluate psychosocial adjustment and ~~proneness to~~prevalence of psychopathology in adolescents and young adults with CHD. The importance of our investigation is that it systematically addressed the question of how the several demographic and clinical variables relate to psychiatric morbidity and to psychosocial adjustment, using very strict methods of psychiatric diagnosis.

Comment [I5]: Reviewer 1 "were" changed to "are"

Comment [I6]: Reviewer 1 "in European studies" changed to "some European studies"

Comment [I7]: Reviewer 1 "that leading to" changed to "which can lead to"

Comment [I8]: Reviewer 1 "Proneness to" changed to "prevalence of"

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METHODS

Participants

The study enrolled 110 CHD patients, 62 male and 48 female, with a mean age of 8.00 ± 3.62 years (range:12-26 years old). The participants who had not achieved an educational level that enabled them to understand and complete the written questionnaires were excluded from the study.

At the time of the interview, two participants were married, one was divorced, two were living in a marital union. All the others (105) were single.

53 patients had completed their secondary education (12th grade), 40 the 3rd cycle (9th grade), 11 the 2nd cycle (6th grade) and 6 had graduated from college. Of these patients, 55 had at least repeated one year at school (mean=1,49 ± 0,50 year).

Of the 110 participants, 20 were employed full- or part-time, 7 were unemployed and all the others 83 were students.

Complete medical records were available for all the patients, who had been followed in the pediatric cardiology or cardiology departments of a tertiary hospital.

For 58 individuals the CHD was cyanotic and for 52 it was acyanotic; 34 of these patients had a severe form of CHD, 18 a moderate and 58 a mild one; 41 patients had some physical limitations while 69 did not, these limitations were given by their doctors, stating that the most severe cases could not have any kind of physical activity while the moderate or mild cases were allowed controlled physical activity. 4 patients had severe residual lesions, 21 moderate and 85 mild lesions. 23 patients were never submitted to any kind of surgical procedure, while 42 had 1 surgery, 25 had two, 11 had three, 5 had four, 3 had five and 1 had 9 surgeries. 47 patients were on pharmacological therapy while 63 were not.

In many of our participants, the main CHD was combined with other heart diseases. Patients with associated extracardiac malformations or genetic disorders were excluded from the study. The participants had the following distribution of pathologies:

Table 1. Patients and cardiac diseases: ~~transposition of the great arteries (9; two of them had also ventricular septal defect and aortic stenosis, and one had ventricular septal defect and pulmonary stenosis), tetralogy of Fallot (30), coarctation of the aorta (11; one had also ventricular septal defect and one aortic stenosis) ventricular septal defect (24; one had also interruption of the aortic arch and one had mitral insuficiency), atrial septal defect (6; one had also mitral atresia and pulmonary hypertension, and one had Ebstein disease), atrioventricular septal defect (4), aortic stenosis (6), pulmonary stenosis (6), single ventricle (2; one had also pulmonary atresia and one had pulmonary stenosis), patent ductus arteriosus (2), double outlet right ventricle (1), pulmonary atresia (3), Ebstein disease (3), mitral valve prolapse (1), bicuspid aortic valve (1) and tricuspid valve regurgitation (1).~~

9 TGA: from those, 2 also had VSD and AS, 1 also had VSD and PS
30 TF
11 CA: from those, 1 also had VSD and 1 also had AS
24 VSD: from those, 1 also had IAA, 1 also had MI
6 ASD: from those, 1 also had MA and PH, 1 also had ED

Comment [I9]: Reviewer 2
Explanation of the patients physical limitations

Comment [I10]: Reviewer 1
"Cardiac malformations" changed to "extracardiac malformations"

Comment [I11]: Reviewer 1
"chromosomopathies" changed to "genetic disorders"

Comment [I12]: Reviewer 1 and 2

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4 AVSD
6 AS
2 SV: from those, 1 also had PA, 1 also had PS
2 PDA
1 DORV
3 PA
3 ED
1 MVP
1 BAV
1 TVR

TGA = Transposition of the Great Arteries; TF = Tetralogy of Fallot; CA = Coarctation of the Aorta; VSD = Ventricular Septal Defect; ASD = Atrial Septal Defect; AVSD = Atrioventricular Septal Defect; AS = Aortic Stenosis; PS = Pulmonary Stenosis; MI = Mitral Insuficiency; ED = Ebstein Disease; SV = Single Ventricle; PA = Pulmonary Atresia; MA = Mitral Atresia; PDA = Patent Ductus Arteriosus; DORV = Double Outlet Right Ventricle; MVP = Mitral Valve Prolapse; BAV = Bicuspid Aortic Valve; TVR = Tricuspid Valve Regurgitation; MVP = Mitral Valve Prolapse; PH = Pulmonary Hypertension; IAA = Interruption of the Aortic Arch;

The diagnosis was determined during the neonatal period for 61, before the first birthday for 28, 5 were diagnosed between the ages of 1 to 3 years, 6 were diagnosed between the ages of 3 to 6 years and between the ages of 6 and 12 for 12 participants.
The first surgery was performed for 5 of the participants during the neonatal period, before the first birthday for 30, between the ages of 1 and 3 for 19, and between the ages 3 and 6 for 20 participants, between the ages 6 to 12 for 8 and between the ages of 12 to 18 for 28.
We also invited one relative of each patient to participate in this study and 100 accepted to take part in it.

Assessment Instruments

In this study, we used different surveys to collect the necessary information: an identification form, a semi-structured interview, a standardized psychiatric interview SADS-L, the self-report and observational questionnaires of the ASEBA system for psychosocial adjustment, ASR, YSR, ABCL and CBCL (for patients $\geq$ 18 years and < 18 years). Additional questionnaires used in this research are described in detail in another report. [11]

Comment [U13]: Reviewer 2: New citation

We used an identification form to collect personal and demographic data from each patient (e.g., marital status, educational level and occupation), as well as all relevant aspects from their medical history (diagnosis, severity and category of heart disease, surgical interventions, pharmacological therapy, and presence of residual lesions, among others).
The semi-structured interview included 38 multiple-choice or short-answer questions that focused on different topics such as social support, family upbringing, self-image, functional limitations and emotional adjustment.
A standardized psychiatric interview, SADS-L (Schedule for Affective Disorders and Schizophrenia – Lifetime version) [12], was administered to obtain a clinical diagnosis of any psychopathological disorders that may have existed before the interview in these patients.
The YSR and ASR are self-report questionnaires, designed to collect a description of a child or

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adult's functioning; they assess individuals in scales of withdrawn behavior, somatic complaints, anxiety/depression, thought problems, social problems, attention problems, delinquent behavior, aggressive behavior, internalization and externalization.[12] The CBCL and ABCL are observational versions of the same questionnaires, to be completed by the patients' parents or caregivers, having as a requirement being knowledgeable about the patient, as they report their perception on the behavior and possible problems occurring in the patient. For their similarities, and to have a better representative sample, the results of the YSR and ASR were pooled, as well as the results of the CBCL and the ABCL, and for statistical purposes the overall results were counted for each scale.

Procedure

Prospective participants were contacted while waiting for their appointment in cardiology or pediatric cardiology department. Our first concern was to get their or their parents' consent (when they were under 18 years old). At this time, they were informed about all aspects of the research, and when they accepted to participate, they completed an informed consent form approved by the hospital's ethical committee, which followed international conventions guaranteeing the rights of the patients. The interview happened on the spot. The parents or caregivers accompanying the patient were asked to fill out a questionnaire, and 10 caregivers refused to participate or were not present for the application of the protocol, and subsequently expressed their intention not to participate. The whole investigation (plan, assessment instruments used, procedures) was previously submitted to the appreciation of the hospital's ethical committee and was approved.

Comment [U14]: Reviewer 1: The whole investigation (plan, assessment instruments used, procedures) was previously submitted to the appreciation of the hospital's ethical committee and was approved.

Design

All the assessment measures were obtained on a single occasion. Clinical data were collected retrospectively using each patient's clinical record, with assistance from hospital medical staff.

Data Analysis

Statistical analysis of the data was processed using the software IBM SPSS (Statistical Package for the Social Sciences, Chicago, IL, USA), version 19. The distribution of all the variables was tested. Differences for parametric variables were established using Student's *t*-tests, while differences for non-parametric variables (the majority) were established using Mann-Whitney *U* test and Chi-square tests of association.

RESULTS

We found that 21.8% of our participants had a psychiatric disorder and that there was a statistical difference between the two genders, with females almost doubling males' rate (31.3% in females and 14.5% in males; p=0.035); The gender differences in the rates of psychiatric disorder still maintain (p=0.049) when we consider separately the age-range 19 years old and above, but they are not significant in the other age groups. One or more of the following psychiatric disorders had been diagnosed for our participants in all their lifetime prior to interview: Minor or Major Depressive Syndrome (13), Panic Disorder (3), Anxiety Disorder (4), or Manic Syndrome (3), Cyclothymic Personality (1).

Comment [U15]: Reviewer 2: new explanation

For the sake of data analysis, we grouped the results on psychosocial adjustment in either self-reported or observational.

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In table 2 and 3, we summarize the main results of this study regarding self-report measures.

The self-report measures on psychosocial adjustment revealed statistical differences between genders, as females showed higher levels of somatic complaints (u=260.000; p=0.011), anxiety/ depression (u=984.000; p=0.002), aggressive behavior (u=920.500; p=0.001), attention problems (u=1123.500; p=0.027), thought problems (u=1069.500;p=0.010), internalization (u=869.000; p=0.000) and externalization (u=1163.000; p=0.050) and overall worse psychosocial adjustment than males.

On the other hand, teens and young adults with a severe CHD showed worse psychosocial adjustment, with more somatic complaints (u=264.000; p=0.022) and higher levels of internalization (u=917.000; p=0.015) in self-report measures, when compared with patients with moderate-to-mild CHD.

When analyzing the impact of the kind of CHD in psychosocial adjustment, no statistical differences were found.

Patients who underwent surgical interventions revealed worse psychosocial adjustment than patients with no surgical repairs, showing higher levels of withdrawn behavior (u=719.500; p=0.037) in self-assessment.

Patients with poor social support reported worse psychosocial adjustment, with higher levels of withdrawn (u=557.500; p=0.000) and social problems (u=748.500; p=0.023) when compared with patients with good social support.

Patients with limited physical competence showed more withdrawn behavior (u=1023.000; p=0.015) when compared to patients with satisfactory physical competence, thus presenting worse adjustment.

Patients with worse academic performance showed higher levels of anxiety/depression (u=916.000; p=0.020) and attention problems (u=861.500; p=0.007) in self-report, when compared to those who feel their academic performance was satisfactory.

Patients with severe-to-moderate residual lesions revealed worse psychosocial adjustment in self-report than those with mild residual lesions, showing higher levels of internalization (u=782.500; p=0.046).

No differences were found in self-report between patients with or without need for pharmacological therapy, for cardiac disease.

Comment [I16]: Reviewer 2
Explanation of pharmacological therapy: this is for cardiac disease and not for psychiatric disorder

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Table 2: Comparing psychosocial adjustment measures (self-report) in male versus female participants, in severe versus moderate to mild CHD, in participants with versus without surgical intervention and with severe/ moderate versus mild residual lesions

	Male (N=62)	Female (N=48)			Severe CHD (N=34)	Moderate/ mild CHD (N=76)			With Surgical intervention (N= 87)	No Surgical intervention (N=23)			Severe/ moderate residual lesions (N=25)	Mild Residual lesions (N=85)		
ASR/YSR (Self-report)	***Mean***	***Mean***	***U***	***P***	***Mean***	***Mean***	**U**	***P***	***Mean***	***Mean***	**U**	***P***	***Mean***	***Mean***	**U**	***P***
Withdrawn	54,02	57,42	1396,000	0,58	62,63	52,31	1049,000	0,11	58,74	43,26	**719,000**	**0,04**	65,14	52,56	821,500	0,08
Somatic complaints	26,03	37,70	**260,000**	**0,01**	37,43	26,77	**264,000**	**0,02**	32,80	23,60	234,000	0,07	37,7	28,14	231,500	0,07
Anxiety/ Depression	47,37	66,00	**984,000**	**0,00**	56,53	55,04	1257,000	0,82	58,09	45,70	775,000	0,10	59,46	54,34	963,500	0,48
Social problems	51,85	60,22	1261,500	0,17	62,63	52,31	1049,500	0,11	56,82	50,50	885,500	0,39	64,96	52,72	826,000	0,09
Thought problems	48,75	64,22	**1069,500**	**0,01**	63,06	52,12	1035,000	0,10	56,47	51,83	916,000	0,53	57,52	54,91	1012,000	0,71
Attention problems	49,62	63,09	**1123,500**	**0,03**	54,40	55,99	1254,500	0,81	58,48	44,24	741,500	0,06	57,62	54,88	1009,500	0,70
Agressive behaviour	55,71	55,23	**920,500**	**0,00**	61,87	52,65	1120,000	0,26	55,23	56,52	890,500	0,42	63,38	53,18	968,000	0,51
Internalization	46,35	67,32	**869,000**	**0,00**	60,56	53,24	**917,000**	**0,02**	56,76	50,72	880,500	0,38	59,28	54,39	**782,500**	**0,05**
Externalization	45,52	68,40	**1163,000**	**0,05**	66,53	50,57	1214,500	0,62	56,88	50,28	817,500	0,18	**66,70**	**52,21**	1045,500	0,90

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Table 3: Comparing psychosocial adjustment measures (self-report) in participants with good versus poor social support, in those with versus without physical limitations, in those with poor versus good academic performance

	Good Social support (N=85)	Poor social support (N=25)			With Psysical limitations (N=41)	No Psysical limitations (N=69)			Poor academic performance (N=77)	Good academic performance (N=33)		
ASR/YSR (Self-report)	***Mean***	***Mean***	***U***	***P***	***Mean***	***Mean***	**U**	***P***	***Mean***	***Mean***	**U**	***P***
Withdrawn	**49,56**	**75,70**	**557,500**	**0,00**	**65,05**	**49,83**	**1023,000**	**0,02**	53,71	59,68	1132,500	0,36
Somatic complaints	**31,02**	**29,06**	329,000	0,70	**34,41**	**28,24**	332,000	0,18	31,20	28,40	306,000	0,59
Anxiety/ Depression	**54,98**	**57,28**	1018,000	0,75	**61,93**	**51,68**	1151,000	0,10	**50,90**	**66,24**	**916,000**	**0,02**
Social problems	**51,81**	**68,06**	**748,500**	**0,02**	**57,80**	**54,13**	1320,000	0,55	53,32	60,59	1102,500	0,27
Thought problems	**56,11**	**53,42**	1010,500	0,71	**59,78**	**52,96**	1239,000	0,27	53,44	60,32	1011,500	0,29
Attention problems	**53,96**	**60,74**	931,500	0,35	**61,12**	**52,75**	1225,000	0,24	**50,19**	**67,89**	**861,500**	**0,01**
Agressive behaviour	**53,49**	**62,32**	911,000	0,28	**62,99**	**51,05**	1254,500	0,32	54,44	57,97	1116,000	0,31
Internalization	**53,72**	**61,56**	892,500	0,23	**59,40**	**53,08**	1121,500	0,07	53,49	60,18	1056,500	0,16
Externalization	**53,50**	**62,30**	1032,500	0,83	**62,65**	**51,25**	1353,500	0,71	**52,72**	**61,98**	**973,500**	**0,05**

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According to the assessment of patients' caregivers, no differences were found between patients who underwent surgical procedures and the ones who did not had surgical procedures done.

In the caregivers' assessment (table 4), male patients are perceived as having worse psychosocial adjustment than females, as they were assessed as having higher levels of withdrawn (u=911.500; p=0.020) and aggressive behaviors (u=945.500; p=0.038).

Again on the relatives' standpoint, the cyanotic patients were assessed as having higher levels of attention problems (u=981.500; p=0.045) than the acyanotic, thus showing worse psychosocial adjustment.

When compared with patients with good social support, those with poor support showed, also on the stand point of the caregivers, higher levels of withdrawn behavior (u= 517.000; p=0.001) and internalization (u=608.000; p=0.007) and thus, worse psychosocial adjustment. According to their relatives' assessment, patients with severe to moderate forms of residual lesions showed higher levels of social problems (u=205.500; p=0.008), attention problems (u=649.500; p=0.028) and internalization (u=567.500; p=0.004), than the ones with mild residual lesions, and thus a worse psychosocial adjustment.

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Table 4: Comparing psychosocial adjustment measures (caregivers'report) in male versus female participants, in cyanotic versus acyanotic CHD, in participants with good versus poor social support and with severe/ moderate versus mild residual lesions

	Male (N=58)	Female (N=43)			Cyanotic (N=51)	Acyanotic (N=50)			Good Social support (N=76)	Poor social support (N=25)			Severe or moderate residual lesions (N=24)	Mild residual lesions (N=77)		
ABCL/CBCL (report from caregivers)	***Mean***	***Mean***	***U***	***P***	***Mean***	***Mean***	**U**	***P***	***Mean***	***Mean***	**U**	***P***	***Mean***	***Mean***	**U**	***P***
Withdrawn	**56,78**	**43,20**	**911,500**	**0,02**	54,15	47,79	1114,500	0,27	**45,30**	**68,32**	**517,000**	**0,00**	**56,46**	**49,30**	793,000	0,29
Somatic complaints	47,19	56,14	1026,000	0,12	54,44	47,49	1099,500	0,23	49,92	54,28	868,000	0,51	**55,73**	**49,53**	810,500	0,36
Anxiety/ Depression	49,66	52,81	1169,000	0,59	55,19	46,73	1061,500	0,15	48,65	58,14	771,500	0,16	**57,29**	**49,04**	773,000	0,23
Social problems	31,08	29,57	404,000	0,74	33,88	27,34	351,500	0,14	29,01	34,59	286,500	0,27	**39,91**	**26,78**	**205,500**	**0,01**
Thought problems	48,64	54,19	1110,000	0,32	54,27	47,66	1108,000	0,23	50,91	51,26	943,500	0,96	**57,58**	**48,95**	766,000	0,18
Attention problems	52,34	49,20	1169,500	0,59	**56,75**	**45,13**	**981,500**	**0,05**	48,82	57,62	784,500	0,19	**62,44**	**47,44**	**649,500**	**0,03**
Agressive behaviour	50,77	51,31	1233,500	0,92	51,21	50,79	1264,500	0,94	50,52	52,46	913,500	0,77	**53,92**	**50,09**	854,000	0,57
Internalization	**45,80**	**58,01**	**945,500**	**0,04**	54,07	47,87	1118,500	0,29	49,79	54,68	858,000	0,47	**59,15**	**48,46**	728,500	0,12
Externalization	49,55	52,95	1063,000	0,56	55,86	46,04	1027,000	0,09	**46,50**	**64,68**	**608,000**	**0,01**	**65,85**	**46,37**	**567,500**	**0,00**

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DISCUSSION

This study ~~is important because it~~ systematically tested the effects of different demographic, clinical and psychosocial variables in psychosocial adjustment and psychiatric morbidity. In a growing population of adults with CHD, this information is rather important in unveiling strategies that can be used to assist and care for patients, leading to better emotional adjustment and better outcomes in different life challenges.

The assessment instruments used for psychosocial adjustment enabled us to compare sub-groups of patients, regarding demographic, clinical and psychosocial variables. One main finding of our study was that females with CHD reported higher levels of somatic complaints, anxiety/depression, thought problems, attention problems, aggressive behavior, internalization and externalization, than males, thus showing a worse psychosocial adjustment. Conversely, relatives find male patients more withdrawn than girls.

In our study, female patients had almost the double of the lifetime prevalence of psychopathology than males. These findings on psychiatric morbidity and on the different scales of psychosocial adjustment are consistent with other studies on the general population that report differences between genders, with females showing higher rates of emotional problems. Studies show also that females have greater likelihood of displaying higher levels of anxiety/depression and somatic complaints when facing negative obstacles that interfere with the interpersonal level, resulting in higher levels of internalization.[6,8,14]

This may be due to the presence of a scar, situated on the chest, being a source of uncertainties or discomfort. In addition to affecting sexual relationships CHD can also interfere with pregnancy and delivery, leading to a sense of anxiety about their physical condition.[6,8]

Some studies show that females are more likely to develop depressive symptoms when facing negative life events than males.[6,10]

In this study, adolescents or young adults with severe type of CHD reported having higher levels of social problems and, thus, worse psychosocial adjustment, compared with those with moderate or mild form of CHD.

These results may be related to the fact that they need further medical care throughout their life, while patients with mild or moderate CHD may have a daily life similar to healthy adolescents and young adults.[8] Patients with severe forms of CHD show higher level of internalization and somatic complaints and that may be associated to the fact that these patients are more vigilant about their health, being more anxious about any complications. This may explain the results, since anxiety is a component of internalization scale.[15]

The type of CHD did not show any impact with statistical relevance in patients' self-report measures of psychosocial adjustment. However, the caregivers' standpoint seems to be more sensitive regarding this feature, as they perceive the cyanotic patients as having more attention problems and worse psychosocial adjustment.

Other published studies also showed that the cyanosis is not a stable indicator that patients will have behavioral and emotional problems.[8,10,16]

Patients who underwent surgical procedures revealed higher levels of withdrawn behavior. This may be related with the fact that admissions are long as well as the recovery, thus providing a prolonged absence from education and from contact with the peer groups, which could lead to difficulties of reintegration and therefore to the isolation of patients.[5,7,17]

Patients with worse social support had higher levels of withdrawn behavior and social problems, and thus, a worse psychosocial adjustment. The assessment of the caregivers also reported higher levels of withdrawn behavior and internalization in patients with poor social support, showing worse psychosocial adjustment.

Comment [I17]: Reviewer 2
As asked, I'll leave to the reader's opinion to state if this study is important or not

Comment [I18]: Reviewer 1
"contrarywise" changed to "conversely"

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According to several studies, parents and siblings of adolescents or young adults with CHD are more prone to face a different number of psychosocial stresses putting the whole family in need of psychosocial support. Many studies reveal a higher need for intervention on family problems in families with children with chronic medical diseases. When the complexity of the disease is low, parents seem to be more fitted to provide support.[18] These families are reported to experience more stress, that can have an impact on the child's adjustment.[16]

Parents of children with CHD can be overprotective and hyper vigilant about their child's health, making it hard for their children to be more independent. Many studies show that these patients are more unlikely to have "independent lifestyles" than healthy adolescents or young adults.[7]

Comment [I19]: Reviewer 2 "more likely" changed to "more unlikely"

Comment [U20]: independent

The participation in leisure time activities can be a contributor to a better social outcome.[14]

Limited physical competence translated into more withdrawn, feeling more isolated, when compared with patients with satisfactory physical competence. Self-report showed that patients with physical limitations have worse psychosocial adjustment. A low exercise capacity can be translated into more internalizing problems. For older heart patients, limited physical competence lead to concerns and anxiety about their health.

According to some authors, patients submitted to physical training interventions, showed a decrease in internalizing problems.[8]

Physical limitations and school absences prevent full participation in different activities, leading to isolation and social awkwardness. This can be translated into restricted employment opportunities.[7]

In our study, an unsatisfactory academic performance led to worse psychosocial adjustment, as patients report having higher levels of anxiety/depression, attention problems and externalization than those with good academic performance. Several previous published studies show that CHD has an impact on school careers, for the many hospitalizations and restrictions, being the main reason for the attendance of special education by these patients. When compared to healthy adolescents or young adults, the CHD patients are more unlikely to complete a lower educational level.[14]

Sometimes, children with CHD have neurodevelopment deficits. These often will not show until school age, when the academic demands start having an impact on their lives. Many families rationalize their child's developmental delay to the disease and the several hospitalizations.[19]

Some studies show that unsatisfactory educational background can be translated into lower educational and occupational achievement.[7]

This study showed a 21.8% prevalence of psychiatric disorder in our patients. Females showed a higher percentage of psychiatric disorder with 31%, and males only had 14%.

When compared to the reference value of the World Health Organization (WHO), 10% of the world population, it seems that adolescents and young adults with CHD have an increased proneness for psychiatric diagnosis.[20] However, a study of six different European countries showed a prevalence of 25% in the general population, which is closer but higher than the results for CHD patients in our study.[21] Another study (Table 5) estimated that the life time prevalence of psychopathology is 19.4% in Spain, 18.1% in Italy (countries that can be considered culturally close to Portugal), and 25.2% in Germany, but in striking contrast, 37.9% in France and 47.4% in the United States of America.[22]

In conclusion, our study emphasizes the need for evaluating and helping adolescents and young adults with CHD. Results from the current investigation confirm and extend those from previous published reports.

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Table 5. Comparing the prevalence of psychiatric disorders in our participants with data from studies in other countries [22]

Author/Year /Country	Number of Participants	Kind of population	Lifetime Prevalence of Psychiatric Disorders (%)	Comments
Portugal	**110**	CHD patients	**21,8**	These numbers were obtained between 2010 and 2012.
Kessler R C, Angermeyer M, Anthony J C, et al / 2007/ Spain (22)	**842**	Population in general	**19,4**	These results were done in the WMH data using discrete-time survival analysis to predict onset of disorders across age groups 18-34, 35-49, 50-64, and 65+ (between 2002 and 2005). No meaningful difference exists between less developed and developed countries.
Kessler R C, Angermeyer M, Anthony J C, et al / 2007/ Italy (22)	**612**	Population in general	**37,9**	
Kessler R C, Angermeyer M, Anthony J C, et al / 2007/ France (22)	**847**	Population in general	**18,1**	
Kessler R C, Angermeyer M, Anthony J C, et al / 2007/ Germany (22)	**573**	Population in general	**25,2**	
Kessler R C, Angermeyer M, Anthony J C, et al / 2007/ United States of America (22)	**3929**	Population in general	**47,4**	

Comment [U21]: Reviewer 1: new table to synthesize the comparisons on lifetime prevalence of psychiatric disorders in different countries

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COMPETING INTERESTS

There are no competing interests.

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PSYCHOSOCIAL ADJUSTMENT AND PRONENESS TO PSYCHOPATOLOGY IN ADOLESCENTS AND YOUNG ADULTS WITH CONGENITAL HEART DISEASE

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Key Words: Congenital Heart Disease, Psychosocial Adjustment, Psychopathology

Word Count: 4027 with references and abstract; 3177 without references and abstract

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ABSTRACT

Objectives: Our purpose was to study psychosocial adjustment and psychiatric morbidity of adolescents and young adults with congenital heart disease (CHD).
Design: All assessment measures were obtained on a single occasion in patients with a diagnosis of Congenital Heart Disease coming to consultation in a tertiary hospital.
Participants: We evaluated 110 CHD patients (62 male) aged from 12 to 26 years old (mean=18.00 ± 3.617), 58 cyanotic.
Primary and secondary outcome measures: All assessment measures were obtained on a single occasion. Demographic information and clinical history were collected. Questionnaires regarded topics as social support, family educational style, self-image and physical limitations, a standardized psychiatric interview SADS-L, and a self-report questionnaire on psychosocial adjustment, YSR or ASR. One of the relatives completed an observational version of the same questionnaires (CBCL or ABCL).
Results: We found a 21.8% lifetime prevalence of psychopathology, 31.3%, in females, 14.5% in males, showing a somewhat increased proneness in CHD patients. Females also showed worse psychosocial adjustment, with more somatic complaints (u=260.000; p=0.011), anxiety/ depression (u=984.000; p=0.002), aggressive behavior (u=920.500; p=0.001), attention problems (u=1123.500; p=0.027), thought problems (u=1069.500; p=0.010), internalization (u=869.000; p=0.000) and externalization (u=1163.000; p=0.050). Patients with severe CHD (u=939.000; p=0.030) and surgical repaired (u=719.000; p=0.037) showed worse psychosocial adjustment. Those with poor social support showed more withdrawn (u=557.500; p=0.000) and social problems (u=748.500; p=0.023), and patients with unsatisfactory school performance revealed more anxiety/depression (u=916.000; p=0.020) and attention problems (u=861.500; p=0.007).
Conclusions: CHD males with good social support and good academic performance have a better psychosocial adjustment.

Key Words: Congenital Heart Disease, Psychosocial Adjustment, Psychopathology

Word Count: 4027 with references and abstract; 3177 without references and abstract

Abbreviations: CHD - congenital heart disease; SADS-L - Schedule for Affective Disorders and Schizophrenia – Lifetime version; YSR - Youth Self Report; CBCL - Child Behavior Checklist; ASR - Adult Self Report; ABCL - Adult Behavior Checklist

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ARTICLE SUMMARY

The study focuses in:
1. studying psychiatric morbidity and psychosocial adjustment in adolescents and young adults with congenital heart disease, comparing them with healthy population.
2. investigating which demographic, psychosocial and clinical variables contribute to foster resilience and adaptation and which ones have detrimental effects.
Our research is an important contribution to the state-of-the-art on this scientific issue because it systematically tests the contribution of a wide range of demographic, psychosocial and clinical variables in psychosocial and emotional adjustment and psychiatric morbidity.
Strengths: 110 patients were evaluated; A wide sellection of demographic, psychosocial and clinical variables were assessed; we used a very strict method for evaluating psychiatric morbidity and lifetime prevalence of psychpathology, a structured psychiatric interview.
Limitations: The fact that there are no nationwide normative studies of lifetime prevalence of psychopathology in Portugal limits the possibility to compare the prevalence rate in our patients with the general population, and to make clear inferences about it.

INTRODUCTION

The survival rate in the 1950's for children born with moderate CHD was about 20% whereas today about 90% of these children achieve adulthood.[1]
There has been a decrease in child mortality thanks to advances over the last four decades in diagnostic, surgical and catheter interventional techniques.[2] As these children survive, the interest in issues such as psychosocial outcomes have increased also.[3]
Most children with CHD were diagnosed in uterus or in infancy, and are expected to undergo surgical procedures either to correct or palliate their defect.[4, 5] These children need to be seen regularly by a cardiologist.[4]
Many studies have been conducted assessing the impact of CHD on children's or adolescents' psychosocial and cognitive functioning. Although, a consensus among these studies have not yet been reached, some report higher rates of behaviour problems in children and adolescents with CHD, while others have not found any differences between patients with CHD and norms.[4]
It is believed that children with CHD have a higher risk of developing behavioural and emotional problems, when compared to healthy children. Several studies have reported that these children have increased feelings of anxiety and inferiority, higher degrees of impulsiveness, higher levels of emotional and behavioural problems.[6]
On the other hand, in European studies have showed a good psychological functioning in adults with CHD.[7]
Not much is known about this topic, as some studies say that, in a 25-year follow-up, more psychosocial distress was found in adults with CHD in comparison with a normative group. The differences found were limited to somatic complaints and thought problems and behaviours.[6]
As for psychopathology, studies have also disagreed in some aspects, many authors

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believe that CHD patients have a higher probability of having psychopathological symptoms while others have found similar numbers between these patients and healthy children and adolescents.[8]

Several characteristics can be described as facilitators of positive perspectives of stressful life situations and reduced psychological distress such as self-esteem and similar conduct like self-concept and self-perception. Some studies found that usually CHD patients have lower self-esteem although, after surgery, patients reported better self-esteem or self-concept.[9]

Cognitive perceptions are believed to have an influence on a CHD patient's life. The more negative these perceptions are, the higher psychological distress was found. The negative perception can be associated, more than the severity of the disease itself, to higher distress and worse psychological adjustment.[9]

Some studies have shown that patients with cyanotic heart disease have a higher risk of presenting behavioral problems compared to patients with non-cyanotic heart defects, but other studies did not show this association.[5, 10]

Patients with CHD who underwent surgical procedures, had more behavioral problems when compared with those who did not require surgery, and more likely to develop psychiatric problems.[5]

As far as the physical condition is concerned, most patients with CHD have limitations, that leading to more behavioral and emotional problems.[5, 8]

In this study, we aimed to evaluate psychosocial adjustment and proneness to psychopathology in adolescents and young adults with CHD. The importance of our investigation is that it systematically addressed the question of how the several demographic and clinical variables relate to psychiatric morbidity and to psychosocial adjustment, using very strict methods of psychiatric diagnosis.

METHODS

Participants

The study enrolled 110 CHD patients, 62 male and 48 female, with a mean age of 8.00 ± 3.62 years (range:12-26 years old). The participants who had not achieved an educational level that enabled them to understand and complete the written questionnaires were excluded from the study.

At the time of the interview, two participants were married, one was divorced, two were living in a marital union. All the others (105) were single.

53 patients had completed their secondary education (12th grade), 40 the 3rd cycle (9th grade), 11 the 2nd cycle (6th grade) and 6 had graduated from college. Of these patients, 55 had at least repeated one year at school (mean=1,49 ± 0,50 year).

Of the 110 participants, 20 were employed full- or part-time, 7 were unemployed and all the others 83 were students.

Complete medical records were available for all the patients, who had been followed in the pediatric cardiology or cardiology departments of a tertiary hospital.

For 58 individuals the CHD was cyanotic and for 52 it was acyanotic; 34 of these patients had a severe form of CHD, 18 a moderate and 58 a mild one; 41 patients had some physical limitations while 69 did not. 4 patients had severe residual lesions, 21 moderate and 85 mild lesions. 23 patients were never submitted to any kind of surgical procedure, while 42 had 1 surgery, 25 had two, 11 had three, 5 had four, 3

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had five and 1 had 9 surgeries. 47 patients were on pharmacological therapy while 63 were not.

In many of our participants, the main CHD was combined with other heart diseases. Patients with associated cardiac malformations or chromossomopathies were excluded from the study. The participants had the following distribution of pathologies: transposition of the great arteries (9; two of them had also ventricular septal defect and aortic stenosis, and one had ventricular septal defect and pulmonary stenosis), tetralogy of Fallot (30), coarctation of the aorta (11; one had also ventricular septal defect and one aortic stenosis) ventricular septal defect (24; one had also interruption of the aortic arch and one had mitral insuficiency), atrial septal defect (6; one had also mitral atresia and pulmonary hypertension, and one had Ebstein disease), atrioventricular septal defect (4), aortic stenosis (6), pulmonary stenosis (6), single ventricle (2; one had also pulmonary atresia and one had pulmonary stenosis), patent ductus arteriosus (2), double-outlet right ventricle (1), pulmonary atresia (3), Ebstein disease (3), mitral valve prolapse (1), bicuspid aortic valve (1) and tricuspid valve regurgitation (1).

The diagnosis was determined during the neonatal period for 61, before the first birthday for 28, 5 were diagnosed between the ages of 1 to 3 years, 6 were diagnosed between the ages of 3 to 6 years and between the ages of 6 and 12 for 12 participants.

The first surgery was performed for 5 of the participants during the neonatal period, before the first birthday for 30, between the ages of 1 and 3 for 19, and between the ages 3 and 6 for 20 participants, between the ages 6 to 12 for 8 and between the ages of 12 to 18 for 28.

We also invited one relative of each patient to participate in this study and 100 accepted to take part in it.

Assessment Instruments

In this study, we used different surveys to collect the necessary information: an identification form, a semi-structured interview, a standardized psychiatric interview SADS-L, the self-report and observational questionnaires of the ASEBA system for psychosocial adjustment, ASR, YSR, ABCL and CBCL (for patients > 18 and < 18). Additional questionnaires used in this research are described in detail in another report.

We used an identification form to collect personal and demographic data from each patient (e.g., marital status, educational level and occupation), as well as all relevant aspects from their medical history (diagnosis, severity and category of heart disease, surgical interventions, pharmacological therapy, and presence of residual lesions, among others).

The semi-structured interview included 38 multiple-choice or short-answer questions that focused on different topics such as social support, family upbringing, self-image, functional limitations and emotional adjustment.

A standardized psychiatric interview, SADS-L (Schedule for Affective Disorders and Schizophrenia – Lifetime version) [11], was administered to obtain a clinical diagnosis of any psychopathological disorders that may have existed before the interview in these patients.

The YSR and ASR are self-report questionnaires, designed to collect a description of a child or adult's functioning; they assess individuals in scales of withdrawn behavior, somatic complaints, anxiety/depression, thought problems, social problems, attention

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problems, delinquent behavior, aggressive behavior, internalization and externalization.[12] The CBCL and ABCL are observational versions of the same questionnaires, to be completed by the patients' parents or caregivers, having as a requirement being knowledgeable about the patient, as they report their perception on the behavior and possible problems occurring in the patient. For their similarities, and to have a better representative sample, the results of the YSR and ASR were pooled, as well as the results of the CBCL and the ABCL, and for statistical purposes the overall results were counted for each scale.

Procedure

Prospective participants were contacted while waiting for their appointment in cardiology or pediatric cardiology department. Our first concern was to get their or their parents' consent (when they were under 18 years old). At this time, they were informed about all aspects of the research, and when they accepted to participate, they completed an informed consent form approved by the hospital's ethical committee, which followed international conventions guaranteeing the rights of the patients. The interview happened on the spot. The parents or caregivers accompanying the patient were asked to fill out a questionnaire, and 10 caregivers refused to participate or were not present for the application of the protocol, and subsequently expressed their intention not to participate.

Design

All the assessment measures were obtained on a single occasion. Clinical data were collected retrospectively using each patient's clinical record, with assistance from hospital medical staff.

Data Analysis

Statistical analysis of the data was processed using the software IBM SPSS (Statistical Package for the Social Sciences, Chicago, IL, USA), version 19. The distribution of all the variables was tested. Differences for parametric variables were established using Student's *t*-tests, while differences for non-parametric variables (the majority) were established using Mann-Whitney *U* test and Chi-square tests of association.

RESULTS

We found that 21.8% of our participants had a psychiatric disorder and that there was a statistical difference between the two genders, with females almost doubling males' rate (31.3% in females and 14.5% in males; p=0.035). One or more of the following psychiatric disorders had been diagnosed for our participants in all their lifetime prior to interview: Minor or Major Depressive Syndrome (13), Panic Disorder (3), Anxiety Disorder (4), or Manic Syndrome (3), Cyclothymic Personality (1).

For the sake of data analysis, we grouped the results on psychosocial adjustment in either self-reported or observational.

The self-report measures on psychosocial adjustment revealed statistical differences

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between genders, as females showed higher levels of somatic complaints (u=260.000; p=0.011), anxiety/ depression (u=984.000; p=0.002), aggressive behavior (u=920.500;
p=0.001), attention problems (u=1123.500; p=0.027), thought problems (u=1069.500; p=0.010), internalization (u=869.000; p=0.000) and externalization (u=1163.000; p=0.050) and overall worse psychosocial adjustment than males.

On the other hand, teens and young adults with a severe CHD showed worse psychosocial adjustment, with more somatic complaints (u=264.000; p=0.022) and higher levels of internalization (u=917.000; p=0.015) in self-report measures, when compared with patients with moderate-to-mild CHD.

When analyzing the impact of the kind of CHD in psychosocial adjustment, no statistical differences were found.

Patients who underwent surgical interventions revealed worse psychosocial adjustment than patients with no surgical repairs, showing higher levels of withdrawn behavior (u=719.500; p=0.037) in self-assessment.

Patients with poor social support reported worse psychosocial adjustment, with higher levels of withdrawn (u=557.500; p=0.000) and social problems (u=748.500; p=0.023) when compared with patients with good social support.

Patients with limited physical competence showed more withdrawn behavior (u=1023.000; p=0.015) when compared to patients with satisfactory physical competence, thus presenting worse adjustment.

Patients with worse academic performance showed higher levels of anxiety/depression (u=916.000; p=0.020) and attention problems (u=861.500; p=0.007) in self-report, when compared to those who feel their academic performance was satisfactory.

Patients with severe-to-moderate residual lesions revealed worse psychosocial adjustment in self-report than those with mild residual lesions, showing higher levels of internalization (u=782.500; p=0.046).

No differences were found in self-report between patients with or without need for pharmacological therapy.

According to the assessment of patients' caregivers, no differences were found between patients who underwent surgical procedures and the ones who did not had surgical procedures done.

In the caregivers' assessment, male patients are perceived as having worse psychosocial adjustment than females, as they were assessed as having higher levels of withdrawn (u=911.500; p=0.020) and aggressive behaviors (u=945.500; p=0.038).

Again on the relatives' standpoint, the cyanotic patients were assessed as having higher levels of attention problems (u=981.500; p=0.045) than the acyanotic, thus showing worse psychosocial adjustment.

When compared with patients with good social support, those with poor support showed, also on the stand point of the caregivers, higher levels of withdrawn (u= 517.000; p=0.001) and internalization (u=608.000; p=0.007) and thus, worse psychosocial adjustment. According to their relatives' assessment, patients with severe to moderate forms of residual lesions showed higher levels of social problems (u=205.500; p=0.008), attention problems (u=649.500; p=0.028) and internalization (u=567.500; p=0.004), than the ones with moderate to mild residual lesions, and thus a worse psychosocial adjustment.

DISCUSSION

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This study is important because it systematically tested the effects of different demographic, clinical and psychosocial variables in psychosocial adjustment and psychiatric morbidity. In a growing population of adults with CHD, this information is rather important in unveiling strategies that can be used to assist and care for patients, leading to better emotional adjustment and better outcomes in different life challenges.

The assessment instruments used for psychosocial adjustment enabled us to compare sub-groups of patients, regarding demographic, clinical and psychosocial variables. One main finding of our study was that females with CHD reported higher levels of somatic complaints, anxiety/depression, thought problems, attention problems, aggressive behavior, internalization and externalization, than males, thus showing a worse psychosocial adjustment. Contrariwise, relatives find male patients more withdrawn than girls.

In our study, female patients had almost the double of the lifetime prevalence of psychopathology than males. These findings on psychiatric morbidity and on the different scales of psychosocial adjustment are consistent with other studies on the general population that report differences between genders, with females showing higher rates of emotional problems. Studies show also that females have greater likelihood of displaying higher levels of anxiety/depression and somatic complaints when facing negative obstacles that interfere with the interpersonal level, resulting in higher levels of internalization.[6,8,13]

This may be due to the presence of a scar, situated on the chest, being a source of uncertainties or discomfort. In addition to affecting sexual relationships CHD can also interfere with pregnancy and delivery, leading to a sense of anxiety about their physical condition.[6,8]

Some studies show that females are more likely to develop depressive symptoms when facing negative life events than males.[6,10]

In this study, adolescents or young adults with severe type of CHD reported having higher levels of social problems and, thus, worse psychosocial adjustment, compared with those with moderate or mild form of CHD.

These results may be related to the fact that they need further medical care throughout their life, while patients with mild or moderate CHD may have a daily life similar to healthy adolescents and young adults.[8] Patients with severe forms of CHD show higher level of internalization and somatic complaints and that may be associated to the fact that these patients are more vigilant about their health, being more anxious about any complications. This may explain the results, since anxiety is a component of internalization scale.[14]

The type of CHD did not show any impact with statistical relevance in patients' self-report measures of psychosocial adjustment. However, the caregivers' standpoint seems to be more sensitive regarding this feature, as they perceive the cyanotic patients as having more attention problems and worse psychosocial adjustment.

Other published studies also showed that the cyanosis is not a stable indicator that patients will have behavioral and emotional problems.[8,10,15]

Patients who underwent surgical procedures revealed higher levels of withdrawn behavior. This may be related with the fact that admissions are long as well as the recovery, thus providing a prolonged absence from education and from contact with the peer groups, which could lead to difficulties of reintegration and therefore to the isolation of patients.[5,7,16]

Patients with worse social support had higher levels of withdrawn behavior and social

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problems, and thus, a worse psychosocial adjustment. The assessment of the caregivers also reported higher levels of withdrawn behavior and internalization in patients with poor social support, showing worse psychosocial adjustment.

According to several studies, parents and siblings of adolescents or young adults with CHD are more prone to face a different number of psychosocial stresses putting the whole family in need of psychosocial support. Many studies reveal a higher need for intervention on family problems in families with children with chronic medical diseases. When the complexity of the disease is low, parents seem to be more fitted to provide support.[17] These families are reported to experience more stress, that can have an impact on the child's adjustment.[15]

Parents of children with CHD can be overprotective and hyper vigilant about their child's health, making it hard for their children to be more independent. Many studies show that these patients are more likely to have "dependent lifestyles" than healthy adolescents or young adults.[7] The participation in leisure time activities can be a contributor to a better social outcome.[13]

Limited physical competence translated into more withdrawn, feeling more isolated, when compared with patients with satisfactory physical competence. Self-report showed that patients with physical limitations have worse psychosocial adjustment. A low exercise capacity can be translated into more internalizing problems. For older heart patients, limited physical competence lead to concerns and anxiety about their health.

According to some authors, patients submitted to physical training interventions, showed a decrease in internalizing problems.[8]

Physical limitations and school absences prevent full participation in different activities, leading to isolation and social awkwardness. This can be translated into restricted employment opportunities.[7]

In our study, an unsatisfactory academic performance led to worse psychosocial adjustment, as patients report having higher levels of anxiety/depression, attention problems and externalization than those with good academic performance. Several previous published studies show that CHD has an impact on school careers, for the many hospitalizations and restrictions, being the main reason for the attendance of special education by these patients. When compared to healthy adolescents or young adults, the CHD patients are more unlikely to complete a lower educational level.[13]

Sometimes, children with CHD have neurodevelopment deficits. These often will not show until school age, when the academic demands start having an impact on their lives. Many families rationalize their child's developmental delay to the disease and the several hospitalizations.[18]

Some studies show that unsatisfactory educational background can be translated into lower educational and occupational achievement.[7]

This study showed a 21.8% prevalence of psychiatric disorder in our patients. Females showed a higher percentage of psychiatric disorder with 31%, and males only had 14%.

When compared to the reference value of the World Health Organization (WHO), 10% of the world population, it seems that adolescents and young adults with CHD have an increased proneness for psychiatric diagnosis.[19] However, a study of six different European countries showed a prevalence of 25% in the general population, which is closer but higher than the results for CHD patients in our study.[20] Another study estimated that the life time prevalence of psychopathology is 19.4% in Spain, 18.1% in Italy (countries that can be considered culturally close to Portugal), and 25.2% in Germany, but in striking contrast, 37.9% in France and 47.4% in the United

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States of America.[21]

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COMPETING INTERESTS
There are no competing interests.

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DATA SHARING STATEMENT

I am available to share any still unpublished data from the study. Any people interested may contact me to metega@sapo.pt
Maria Emília Areias (corresponding author)

CONTRIBUTORSHIP STATEMENT
Authors 1, 2 and 3 are last year students of the masters in clinical and health psychology; they performed the field work, interviewing participants and their relatives, they encoded data and performed the statisctical analysis; author number 1 revised the state-of-the-art about this subject matter and wrote the article.
Author 4 is pediatric cardiologist and advised in collecting clinical data of participants from files and interpreting them.
Author 5 is clinical psychologist; he advised on the methods and strategies for statistical analysis.
Author 6 is pediatric cardiologist and he contributed in planning the research, in all the clinical aspects and procedures in the department.
Author 7 is clinical psychologist and the supervisor of the master students (authos 1, 2 and 3); she planned the design of the study, the methods, chose the assessment instruments to be used, she planned the steps of the statistical analysis, she reviwed the state-of-the-art of this subject matter, and revised this manuscript.

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STROBE 2007 (v4) Statement—Checklist of items that should be included in reports of *cross-sectional studies*

Section/Topic	Item #	Recommendation	Reported on page #
Title and abstract	1	(*a*) Indicate the study's design with a commonly used term in the title or the abstract	Yes (Abstract/ Methods)
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found	Yes (Abstract/ Methods/ Results)
Introduction			
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	Yes (Introduction)
Objectives	3	State specific objectives, including any prespecified hypotheses	Yes (Abstract/ Introduction/ Methods)
Methods			
Study design	4	Present key elements of study design early in the paper	Yes (Abstract/ Methods)
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	Yes (Abstract/ Methods)
Participants	6	(*a*) Give the eligibility criteria, and the sources and methods of selection of participants	Yes (Methods, Participants)
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	Yes (Methods/ Results)
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	Yes (Methods/ Results)
Bias	9	Describe any efforts to address potential sources of bias	*Yes (Methods)*
Study size	10	Explain how the study size was arrived at	Yes (Methods)
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why	Yes (Methods)
Statistical methods	12	(*a*) Describe all statistical methods, including those used to control for confounding	Yes (Methods; Methods of

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			Statistical Analysis)
		(*b*) Describe any methods used to examine subgroups and interactions	Yes (Methods)
		(*c*) Explain how missing data were addressed	….
		(*d*) If applicable, describe analytical methods taking account of sampling strategy	….
		(*e*) Describe any sensitivity analyses	….
Results			
Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	There is only one stage
		(b) Give reasons for non-participation at each stage	Yes (Methods)
		(c) Consider use of a flow diagram	No
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	Yes (Methods/ Results)
		(b) Indicate number of participants with missing data for each variable of interest	The only missing are some relatives of patients) who did not accept to participate in the study
Outcome data	15*	Report numbers of outcome events or summary measures	Yes (Methods/ Results)
Main results	16	(*a*) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	…..
		(*b*) Report category boundaries when continuous variables were categorized	Yes (Methods/ Results)
		(*c*) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	……
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	Yes (Methods/ Results)
Discussion			
Key results	18	Summarise key results with reference to study objectives	Yes (Results/ Discussion)

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Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias	Yes (Discussion)
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	Yes (Discussion)
Generalisability	21	Discuss the generalisability (external validity) of the study results	Yes (Discussion)
Other information			
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	Yes (Funding/ Aknowledgements)

*Give information separately for cases and controls in case-control studies and, if applicable, for exposed and unexposed groups in cohort and cross-sectional studies.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at http://www.plosmedicine.org/, Annals of Internal Medicine at http://www.annals.org/, and Epidemiology at http://www.epidem.com/). Information on the STROBE Initiative is available at www.strobe-statement.org.

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