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THE PSYCHOSOCIAL SITUATION OF ADULTS WITH CONGENITAL HEART DISEASE IN IRAN

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زندگی صحنه یکتای هنرمندی ماست

هر کسی نغمه خود خواند و از صحنه رود

صحنه پیوسته به جاست

خرم آن نغمه که مردم بسپارند به یاد

شاعر: ژاله اصفهانی

Life is a one-time stage for our art

Everyone sings his song and leaves the stage

And what remains is the stage

Blessed the song that audiences remember

Poet: Zaleh Esfahani

To my parents, who always encourage me to fly as far and high as I dream.

To my beautiful motherland, Iran. I carry its love so strong and deep in my heart always and wherever I am.

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ABSTRACT

Background and objectives: Adults with congenital heart disease (CHD) are a new group of patients with a prevalence of 4 per 1000. They have evolved as a result of significant improvements in medical management during the past decades. However, adults with CHD experience various medical and social challenges that may influence their psychosocial functioning. Providing appropriate medical, rehabilitation and social care for adults with CHD, and indeed improving their well-being require the evaluation of their current psychosocial situation. This thesis aims to increase the understanding of the mental health, somatic symptoms, social support, style of coping, quality of life and life satisfaction of adults with CHD and to examine the possible contributing factors in the context of a developing country; issues not addressed in the current literature.

Methods: This thesis is based on four studies. Study I recruited 347 consecutive CHD patients (18-64 years) from two heart hospitals in Tehran, Iran. The study is cross-sectional and focused on sex differences in socio-economic status, lifestyle and medical characteristics of adults with CHD. Studies II, III and IV have a cross-sectional case-control design comparing the aforementioned CHD patients with 353 non-CHD participants, matched by sex and age. Outcome variables were anxiety, depressive and somatic symptoms (Study II), styles of coping (Study III), and life satisfaction/quality of life (Study IV). The data were analysed with bivariate and multivariate methods. Multivariate linear regression analyses were performed to scrutinize the association of demographic/socio-economic variables, social support, mental health, and medical variables with the aforementioned outcome variables among adults with CHD (Studies II, III, IV).

Results: Study I showed that women with CHD were more often married and had children and were less often employed, but had healthier behaviour compared to men. Even though most of the patients received regular medical

care from different types of medical professionals, half of them had no knowledge about the type of their cardiac defect. Study II showed that CHD patients experienced more anxiety and somatic symptoms than the healthy controls, whereas there were no differences in depressive symptoms. Perceived financial strain, lower social support and low annual income were positively associated with worse outcome in mental health and somatic symptoms. None of the medical variables were related to anxiety, depressive and somatic symptoms. Study III showed that the styles of coping of the CHD patients were comparable to those of the control group and CHD per se was not associated with a certain style of coping, except for palliative reaction pattern. Problem-focused styles of coping were associated with being never married, parenthood, higher level of anxiety and somatic symptoms, lower level of depressive symptoms and higher social support. Emotion-focused styles of coping were associated with annual income and higher level of anxiety. None of the adopted coping strategies were related to the heart disease variables. Study IV showed that adults with CHD had poorer quality of life and lower life satisfaction than the control group. However, CHD was associated only with decreased overall quality of life and its physical health domain, and life and health satisfaction. Among CHD patients, higher quality of life was associated with female sex, younger age, employment status, having less emotional distress and higher social support, while life satisfaction was associated with female sex, being employed, less emotional distress and higher social support.

Conclusions: The results support the notion that psychosocial factors contribute to the well-being of adults with CHD. Socio-economic factors, emotional health and social support are significant determinants in nearly all outcomes of interest which need to be considered by health care providers and policy makers in their efforts to improve the health of adults with CHD. However, longitudinal studies are warranted to establish causal links and qualitative studies are recommended to deepen the understanding of coping and quality of life.

Key words: Developing country, grown-up with congenital heart disease, life satisfaction, mental health, quality of life, social determinants, social support, somatic symptoms, style of coping.

PREFACE

I remember the first book that I read was named "Attila", I was 8 years old and that was the most inappropriate book for a child in that age. It was a hot summer in Shiraz, a beautiful city in south of Iran, where we spent holidays in my aunt's house. It was a lonely summer for me, because my two cousins and my sister were teenagers and did not like to share their secrets with me, and I did not want the company of my other 4 years old cousin. I could read only 70 pages of that book and it was enough to make the most important decision of my life that I want to become a hero. There was a Roman soldier in that book who was smart and brave, and fighting with bad guys. I liked it.

*****

The first time that I found out that being hero does not work was when I finished medical school and immediately started to work as a family physician in a rural primary health care centre in a remote area of Iran. I was young and ambitious. I had the feeling that I will be the second Aviccena[1]. I was thinking that I am able to solve all the health problems of the people. Quickly I found out that I could cure some patients for sure but I could not give them health.

There were serious problems: being poor, being illiterate, having no leisure activity and being dominated by a cumbersome tradition, would not let them to be truly healthy. It happened several times that some patients had no medical problems but they were coming to health centre regularly[2], especially women. They needed to be in other environment than their routines; they needed to have other social roles than just being a wife, mother, and daughter in law. That could be a patient role, the only available alternative.

Then I felt that, I am not satisfied by being just a doctor, sitting in my office and prescribing medications. I wanted to make a bigger change; I wanted to influence positively more people's lives; I wanted to find answers for my very questions that how it is possible to provide better health even for a person living in a very small village in a remote area.

[1] Famous Persian physician (c. 980 – 1037)

[2] Start of my work co-occurred with the start of rural insurance program which provided a very cheap medical service.

In this regard, I decided to study Public Health Sciences. I may achieve my aim, may not, but I always dream about it, that one day, there is health and happiness for everybody. Maybe I have changed the role of being a hero to a "dreamer", but I know that "I am not the only one"[3].

[3] From "Imagine" Song, John Lennon

LIST OF PAPERS

This thesis is based on the following papers, which are referred to in the text by their Roman numerals:

Paper I Eslami, B., Sundin, O., Macassa, G., Khankeh, H. R., Soares, J. J. (2013). Gender differences in health conditions and socio-economic status of adults with congenital heart disease in a developing country. *Cardiology in the Young*. 23(2):209-218.

Paper II Eslami, B., Sundin, O., Macassa, G., Khankeh, H. R., Soares, J. J. (2013). Anxiety, depressive and somatic symptoms in adults with congenital heart disease. *Journal of Psychosomatic Research*. 74(1):49-56.

Paper III Eslami, B., Macassa, G., Khankeh, H. R., Sundin, O., Soares, J. J. Style of Coping and its Determinants in Adults with Congenital Heart Disease in a Developing Country. *Congenital Heart Disease*. (Accepted for publication, 06 October 2013)

Paper IV Eslami, B., Macassa, G., Sundin, O., Khankeh, H. R., Soares, J. J. Quality of life and life satisfaction among adults with and without congenital heart disease in a developing country (Submitted).

LIST OF ABBREVIATIONS

BMI	Body Mass Index
CHD	Congenital Heart Disease
HADS	Hospital Anxiety and Depression Scale
QoL	Quality of Life

DEFINITIONS

Determinant factor	factor associated with outcome of interest
Explanatory factor	factor associated with outcome of interest

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1. INTRODUCTION

1.1. Congenital Heart Disease

Congenital heart disease (CHD) refers to "a gross structural abnormality of the heart or intra-thoracic great vessels that is actually or potentially of functional significance" (Mitchell el al., 1971). Congenital heart disease varies from small lesions (e.g. ventricular septal defect) which may undergo self-resolution spontaneously to severe and potentially life-threatening defects (e.g. single ventricle), which may require several cardiac interventions. Patients with severe CHD develop symptoms early in life, usually during the new-born or infancy periods. Patients with moderate CHD usually are detected in clinical examinations and need professional care, while patients with minor lesions may be asymptomatic their entire life (Hoffman & Kaplan, 2002).

Congenital heart disease is the most common form of major birth defects, accounting for 40% (Marelli et al., 2007; Romano-Zelekha et al., 2001), with a birth prevalence of 4-14 per 1000 (Hoffman & Kaplan, 2002). The birth prevalence of CHD varies according to geographical area, applied surveillance methods and the included type of heart lesions (Marelli, 2012). It is noteworthy that birth prevalence is the most accurate proxy for incidence of CHD.

Only 10-20% of the CHD cases are related to known risk factors. In 17-18% of the cases the cause is chromosomal anomaly (e.g. Down's syndrome) and in 2% single gene defect (e.g. mutation in the PTPN 11 gene). In addition to genetic factors, environmental influences may also increase the risk of CHD incidence. About 1% of the cases are associated with maternal disease (e.g. diabetes mellitus) and "substance" exposure (e.g. Angiotensin-Converting Enzyme Inhibitors consumption during pregnancy). The CHD risk due to hereditary factors is in general small. A parental couple that has had a child with CHD runs a risk of 2-4% that the next child will also have a heart problem, not necessarily the same problem as the parents. If the father has a heart disease the risk for his child is 2-4% and in the mother's case 5-14% (Chen et al., 2009; Connolly & Warnes, 2011; Garg, 2006; Romano-Zelekha et al., 2001). Pregnant patients with severe CHD have a maternal mortality rate of about 7% and a foetal mortality rate of 30% has been found in women with the severest CHD cases (Connolly & Warnes, 2011).

In general, infant mortality rate has declined during past decades due to better maternal care, increases in the quality of, and access to, medical care, and public health services. The medical management of CHD has also improved dramatically. Progress in diagnostic techniques (e.g. echocardiography) has facilitated the early/accurate detection of CHD and follow-up of its course. Innovation in surgical procedures has been significant over the years, particularly in relation to corrective/palliative interventions for complex defects that previously would lead to a rapid death. Catheter-based interventions (e.g. balloon angioplasty) for CHD have rapidly developed and can in many cases serve as an alternative to cardiac surgery or are used together with surgery. Additionally, improved prenatal care and early detection of CHD, and dramatic advances in anesthesia, intensive care, and pharmacological treatment have increased the survival rate of infants with CHD to 85-95% (Baumgartner et al., 2010; Bedard et al., 2008; Perloff & Warnes, 2001; Warnes, 2005). As a result, the size of CHD population and the proportion of patients with complex lesions have increased and mortality occurs usually in older ages (Deanfield et al., 2003; Marelli, 2012; Warnes et al., 2001; Warnes, 2005). In this regard, it has been reported that in United States, the mortality of infants and children with CHD aged between 0-10 years old has decreased from 100/100000 to <1/100000 during 1979-1997. In Canada, the median age of death due to CHD has increased from 2 years of age in 1997-1998 to 23 years of age in 2004-2005 (Marelli, 2012).

1.2. Adults with CHD

Dramatic advances in the medical care of children with CHD resulted in the emergence of a new and constantly growing adult patient population with CHD. The population of adults with CHD may consist of those being medically observed since infancy/childhood, operated during infancy/childhood or those who were diagnosed to have cardiac malformation later in adult life. The exact number of adults with CHD is unknown, but size estimations based on the incidence rate (or birth prevalence), survival through childhood and new cases diagnosed in adulthood (e.g. atrial septal defect) indicate that there are approximately 20,000 adults with CHD in the Netherlands (Deanfield et al., 2003), 124,000 in Canada (Warnes et al., 2001) and between 959,000 and 1.5 million in United States (CHD Public Health Consortium Fact Sheet, 2012). However, researchers from Canada (Marelli et al., 2007) and Japan (Shiina et al., 2011) reported that the prevalence of adults with CHD is about 4 per 1000. Although the prevalence rate is higher in children than adults, the absolute number of adults is presently at least equal to the

number of children with CHD (Marelli et al., 2007; Marelli, 2012). Currently, more deaths due to CHD occur in adulthood and in the near future there will be more adults than children with CHD (Deanfield et al., 2003). Despite the fact that longevity has increased, i.e. mean life-expectancy is 57.3 years (Warnes et al., 2008), many adults with CHD may still experience serious cardiac complications (e.g. arrhythmia, ischemia, endocarditis), may require re-operations (e.g. valve replacement) and may need life-long medical follow-ups because of the natural course of the cardiac defect or the residua and/or the sequelae of previous treatments (Deanfield et al., 2003; Warnes, 2005; Warnes et al., 2008). It is important to consider that most therapeutic procedures are palliative, not curative, and that the repaired heart does not necessarily imply a "normal" heart (Warnes, 2005).

It is noteworthy that in young and middle-aged adults with CHD, health outcome is mostly related to cardiac anomaly complications (e.g. pulmonary hypertension, arrhythmia), while in older adults with CHD, acquired medical conditions (e.g. chronic kidney disease, gastrointestinal bleeding) play an important role on their health status and mortality (Afilalo et al., 2011).

1.2.1. Adults with CHD in developing countries

The medical care of patients with CHD in developing countries has improved considerably during the past years (Gnanappa et al., 2011; Vida et al., 2006), albeit not at the same scale and pace as in developed countries. Edwin et al. (2010) reported that well-equipped cardiac centres and skilled personnel trained in the care of adolescents and adults with CHD are lacking in the West Africa sub-region. There, the cardiologists and cardiovascular surgeons in charge of the care of adults with acquired cardiac diseases are also responsible for the care of adolescents and adults with CHD. They have reported that most repairing cardiac operations occur in adolescence and adulthood, and socio-economic factors are important obstacles. Otherwise, good surgical outcomes with low mortality and morbidity are provided (Edwin et al., 2010; Mocumbi et al., 2011).

2. BACKGRUND

2.1. Living with CHD

Congenital heart disease is a chronic disease because of its long-term nature, uncertain course, and daily life difficulties imposed by physical limitations (Claessens et al., 2005). Rolland (1987) has described that chronic diseases have significant psychosocial influences on affected individuals and their families. Acute onset conditions require crisis-management skills, role flexibility, and ability to use external resources. Progressive course diseases need constant adaptation and role changing. Diseases with constant-course may lead to family exhaustion, while episodic-course diseases create serious uncertainty. Finally, the diseases which may shorten life or cause sudden death, with an inherent "It could happen" nature, may result in family overprotection. Rolland has also explained that the crisis phase of a chronic disease, referring to the symptomatic period before diagnosis, induces the patient to tackle several tasks such as learning to deal with illness-related symptoms on the one hand, and medical interventions and environment on the other. The chronic phase with the prominent psychological component of "day-to-day living with chronic illness" also requires fostering the ability of living normally under abnormal conditions. Accordingly, CHD can psychosocially influence the affected individuals with any of the aforementioned patterns.

Several investigators have shown that CHD patients may have diverse experiences of their childhood due to the onset and the course of the disease. Some patients may have experienced considerable physical performance restrictions, which could prevent them from participating in group activities (Claessens et al., 2005; Horner et al., 2000; McMurray et al., 2001) or have developed the feeling of lack of emotional support and empathy (McMurray et al., 2001). In some CHD patients, body features such as small stature, cyanosis, digital clubbing or surgical scars could be the cause of bullying or being excluded from social circles, or be interpreted as signs of "being different" (Claessens et al., 2005; Horner et al., 2000; McMurray et al., 2001). Some have lived with the fear of undergoing surgical intervention (McMurray et al., 2001), and others have undergone several hospitalizations and consequently cannot attend school regularly (Horner et al., 2000). The aforementioned issues may create social isolation, lack of control, frustration and decreased self-esteem (Claessens et al., 2005; Horner et al., 2000).

In some adult patients, cardiac-related complications (e.g. arrhythmias, heart failure, secondary pulmonary hypertension) may develop after periods of stability in their health condition (Deanfield et al., 2003; Marelli, 2012; Warnes, 2005), which may create or reinforce the feeling of uncertainty about the future such as worries about sudden death, deterioration of health and more physical disabilities (Horner et al., 2000). Thus, some CHD patients may start to become involved in their medical decision-making only as adults (Claessens et al., 2005; Horner et al., 2000), a new responsibility previously shouldered by their parents. They may need to choose among treatment options, which can be as uncertain as the disease course (Horner et al., 2000). Moreover, some CHD patients may experience job related discriminations, for example being excluded from a job (Kamphius et al., 2002b), which may negatively influence their social integration.

2.2. Mental health and somatic symptoms

Stressful life events, acute or chronic, may predispose or precipitate emotional and physical ill-health, e.g. depression and cardiovascular disease (Cohen, 2004; McEwen, 2004). Some of the mechanisms hypothesized to explain the link between stressful life events and ill-health are allostasis/allostatic load and overload (McEwen & Wingfield, 2003). The body has regulatory systems, i.e. neuroendocrine, autoimmune nervous system, metabolic and immune systems, through which it responds to the internal and external demands. These responses are involved in the adaptation to physical, psychological and environmental challenges (Logan & Barksdale, 2008; McEwen, 1998). These adaptive responses and how an individual deals with challenges refer to allostasis. The cumulative state of allostasis refers to allostatic load, which may overload on the superimposition of extra events. It is the price that the body pays for adaptation (McEwen, 1998; McEwen & Wingfield, 2003). Allostatic overload can occur in frequent and intensive challenges (Type 1); over-activity of regulatory systems and their failure to shut-down (Type 2); and in inadequacy of responses (Type 3) (McEwen, 1998, 2004; McEwen & Wingfield, 2003).

In this framework, chronic demands may result in a physiological "wear and tear" condition and contribute to bio-psychological distress, a phenomenon which may result in physical and mental ill-health (Compas, 2006; Logan & Barksdale, 2008; McEwen, 1998), a condition which may occur in CHD patients due to the long-term and uncertain pattern of the disease and its influences on patients' daily lives.

2.2.1. Mental health, somatic symptoms and adults with CHD

Few studies have addressed the psychological experiences of adults with CHD and most of them are from Western/developed countries (Brandhagen et al., 1991; Bromberg et al., 2003; Horner et al., 2000; Kovacs et al., 2009; Popelova et al., 2001). In this respect, different psychological challenges such as negative thoughts, heart-focused anxiety and difficulties in transition to adulthood have been reported (Kovacs et al., 2009; Ong et al., 2011; Rietveld et al., 2002).

Some studies have contrasted adults with CHD with general population normative data (Utens et al., 1998; van Rijen et al., 2003), while others have considered the emotional functioning of CHD patients with different diagnoses (van Rijen et al., 2005). In the first category, some studies have shown that adults with CHD had more favourable scores in hostility, self-esteem and neuroticism (Utens et al., 1994; van Rijen et al., 2003), while others have revealed negative outcomes such as higher scores in somatic complains (Utens et al., 1998) and psychological problems such as anxiety, depression and hostility (Brandhagen et al., 1991).

Other comparative studies have reported equivocal findings. For instance, Cox et al. (2002) have observed better emotional functioning in a small sample of adults with CHD comparing with a sample of orthopaedic outpatients, whereas Pike et al. (2012) have reported higher levels of depression in CHD patients compared to a sample of volunteers matched by age, sex, marital status, education and ethnicity.

In some observational studies, only the emotional well-being of adults with CHD has been addressed without using comparison groups, either through self-report questionnaires (Kovacs et al., 2009; Popelova et al., 2001) or psychiatric interviews (Bromberg et al., 2003; Horner et al., 2000; Kovacs et al., 2009). Their main findings were that nearly a third of adults with CHD experienced anxiety or depressive symptoms, and that 50% experienced at least one life-time mood or anxiety disorder.

Furthermore, studies addressing the association of emotional distress and severity of disease are also inconsistent and show that the severity of CHD is both related (Bromberg et al., 2003; Ong et al., 2011) and unrelated to psychopathology (Brandhagen et al., 1991; Utens et al., 1994, 1998).

Despite these efforts to describe the emotional distress experienced by adults with CHD, the findings are not consistent. This may be due to small sample sizes, lack of comparison groups or appropriate comparison groups, low response rates and differences in measurement (i.e. socio-cultural factors, disease severity, and instrument to measure emotional distress).

It is noteworthy that data on factors associated with emotional distress are crucial for establishing appropriate interventions. This has not been considered by many previous investigators. A few studies have shown that emotional distress was associated with female sex, younger age, health status, low exercise capacity, restrictions imposed by physicians, loneliness and fear of negative evaluation (Kovaks et al., 2009; van Rijen et al., 2004a, 2005). Other important factors, for instance employment that may interfere with psychological well-being, are not taken into account with appropriate statistical analyses, i.e. multiple regression analyses.

In this thesis, the psychological strains of CHD were measured in terms of anxiety, depressive and somatic symptoms, in comparison with randomly selected adults without CHD, which were matched by sex and age while controlling for major potential confounders. This is addressed in Study II.

2.3. Social Support

Cohen (2004) defines social support as the "social network's provision of psychological and material resources intended to benefit an individual's ability to cope with stress". Social support is often differentiated in terms of types of resources including appraisal, instrumental, informational and emotional (Berkman et al., 2000; Cohen, 2004). Emotional support involves expression of caring and sympathy; informational support is related to advice and guidance; instrumental support implies aid and assistance with tangible needs; and appraisal support helps in decision-making (Berkman et al., 2000; Cohen, 2004; Jones & Bright, 2001). According to the social network theory, social ties and networks are important entities for well-being (Berkman et al., 2000; Jones & Bright, 2001), because the structure of social institutions form the available resources, and consequently the behaviour and attitudes of individuals (Berkman et al., 2000). Social networks are placed in, and influenced by, larger social and cultural contexts (Berkman et al., 2000).

Uchino et al. (2012) conceptualize social support in terms of perceived and received support: perceived, "the perception that support will be available if needed" and received, "the exchange of support resources", the perceived social support being more effective.

Several psychological mechanisms have been postulated to explain the link between social support and health. For example, social networks entail social influences as the members share values and norms, hence social support may have positive or negative effects on health behaviours, e.g. tobacco smoking cessation. Social support may enhance self-worth and self-efficacy by strengthening the level of social engagement and the value of social roles (Berkman et al., 2000; Cohen, 2004). Social support may increase the sense of "mattering", which reflects one's importance and meaning to others, consequently giving purpose to one's life (Thoits, 2011). Additionally, social support may directly influence emotional well-being by, for instance, reducing depression (Berkman et al., 2000; Kim et al., 2010; Park et al., 2008). Social support has been shown to buffer stressors and thereby help individuals to cope with negative life events (Ibarra-Rovillard & Kupier, 2011; Stansfeld, 2006). The stress moderating effect of social support may function in two different ways, stress appraisal or coping facilitator. In the former, a person who is aware of available social support appraises stressful threats to be less important and less harmful. In the latter, social support provides more opportunities and material resources, thus empowering the person to face and overcome the stressful event (Cohen, 2004; Jones & Bright, 2001). Some studies of cancer and chronic cardiac patients have shown that high levels of perceived social support may increase emotional well-being or may lead to positive health behaviour changes through the choice of specific styles of coping (Holland & Holahan, 2003; Kim et al., 2010; Park et al., 2008). Moreover, it has been shown that social support may operate indirectly in coping by enhanced self-efficacy (Berkman et al., 2000). Additionally, one needs to consider that the stress buffering role of social support has an effect on health only in stressful conditions (Cohen, 2004). However, Uchino et al. (2012) have mentioned that mental health may moderate the relation of social support with physical health as, for instance, depression obscures the perception of support.

2.3.1. Social support and adults with CHD

Previous studies with young CHD patients have shown that they may be vulnerable to experiencing social isolation due to family overprotection, impaired

peer relations and delayed transition to adult life (Horner et al., 2000; Kovacs et al., 2009).

Few studies have addressed the issue of social support per se among adults with CHD. A study from the Netherlands suggests that CHD patients perceive less support than the normal population (van Rijen et al., 2004b). In contrast, Rose et al. (2005) have stated that CHD patients experience higher levels of social support, while Pike et al. (2012) have reported that adults with CHD perceive a similar level of social support to that of the normal population. Inconsistent findings may pertain to small sample sizes, including selected types of cardiac defects, and the use of different assessment instruments and population normative data as comparison group.

Very few attempts have been made to investigate the association between social support and health outcomes among adults with CHD. In this regard, Kovacs et al. (2009) found that loneliness and fear of negative evaluation were significant predictors of anxiety and depressive symptoms. Other researchers (Chen et al., 2011; Pike et al., 2012; Rose et al., 2005) have reported that social support was positively associated with satisfaction with life as well as high quality of life (QoL) in psychological, social and environmental domains.

In this thesis, social support was conceptualized as perceived support, assessing the individual's perception of support from the social networks in terms of satisfaction, sufficiency and availability. The issue of the social support of CHD patients in comparison with non-CHD individuals is addressed in Study III. Additionally, social support is addressed as a potential determinant factor in mental health, somatic symptoms, styles of coping, QoL and life satisfaction in Studies II, III and IV.

2.4. Style of coping

Coping is defined as "constantly changing cognitive and behavioural efforts to manage specific external and/or internal demands that are appraised as taxing or exceeding the resources of the person" (Lazarus & Folkman, 1984). Thus, coping is a process that changes with the situation over time. Coping has two dimensions: dealing with the emotions caused by stressors and managing the stressors. Both dimensions are influenced by the cognitive appraisal and reappraisal of the

stressors. When an individual appraises that it is not possible to change a stressful situation and has to accept it, the coping style will be emotion-focused, in which the emotions are regulated to make the stressful situation more endurable. On the other hand, when a person appraises that it is possible to change or control the stressful situation, the problem-focused coping style will be applied, in which the person struggles to find and solve the problem (Jones & Bright, 2001; Lazarus & Folkman, 1984; Lazarus, 1999). People may use both strategies to deal with stressful life events as both are essential and may facilitate each other (Jones & Bright, 2001; Lazarus & Folkman, 1984; Lazarus, 1999). For example, when a person is first diagnosed with CHD, she/he tries to seek the best available treatment. In this stage, the patient appraises whether she/he can control the situation by looking for the best therapy (problem-focused coping style) and, for instance, decides to undergo a cardiac intervention. Immediately after the procedure, the patient is not involved in the difficulties of the procedure and the outcome, and thus may try to express hers/his emotions by showing regret, anger, sorrow or totally denying the situation as in this stage the patient is not the agent who can change the situation (emotion-focused coping style). When the acute phase of intervention has passed, the patient has to change her/his life-style or participate in rehabilitation programs in order to prevent future potential cardiac problems or ameliorate the current disabilities, respectively. In this stage, the patient can again control the situation and act aim-oriented (again problem-focused coping style).

It has been stated that the available internal and external resources contribute to the ways people cope. In this regard, some investigators have introduced health, positive beliefs, social skills, age and socio-economic status as factors which may contribute to the adopted coping strategies (Lazarus & Folkman, 1984; Mayes et al., 2000). Among different resources, Thoits (1986) has argued that social support operates as a coping "assistance" strategy.

2.4.1. Style of coping and adults with CHD

As mentioned earlier, adults with CHD have unique life experiences. For instance, they may need long-lasting medical follow-ups and have a wide range of concerns related to adult life such as having difficulties in education, finding a job, getting life/health insurance, sexuality, making their own family and family planning (Kovacs et al., 2005; Simko et al., 2006). The way CHD patients deal with problems is not well documented. It has been suggested that CHD patients adopt a denial

coping strategy to normalize functioning (Horner et al., 2000). On the other hand, Brandhagen and colleagues (1991) have claimed that adults with CHD are "high achievers" as they seem to be "successful" in life. Claessens et al. (2005) have found that CHD patients struggle to be considered normal through the process of "normalization" because they always have the feeling of "being different" from others. These authors argued that adults with CHD use normalization as a dominant strategy to manage life. McMurray and co-workers (2001) have reported that adolescents with CHD avoid or ignore their physical limitations and/or perceived discrimination in order to "get on with life". However, van Rijen et al. (2004b) have found few differences in coping styles between adult CHD patients and a normal population. Saliba and colleagues (2001) have argued that adults with CHD reset their values and self-expectations based on their disabilities.

As the chronic stressors may impair the ability of effective coping (Compas, 2006), in this thesis, emotion- (e.g. expression of emotion) and problem- (e.g. active problem solving) focused styles of coping are addressed (Study III). Further, the styles of coping among adults with and without CHD were compared, while considering the different factors (e.g. socio-economic) associated with adoption of a certain style of coping. In this thesis coping was conceptualized as a person's endeavours to take away the threatening event or reduce its influences on her/his life and the attempts to decrease the negative feelings that arise in response to that threat.

Knowledge regarding the coping strategies of adults with CHD may help to better understand how this group of patients experience life's uncertainties. Moreover, information about the contribution of resources (e.g. social support) in selecting a certain coping strategy may help to establish appropriate interventions to empower patients to better integration of the disease in their daily life.

2.5. Quality of life

Quality of life has become one of the outcomes of interest for health care, beyond the traditional health indicators such as mortality and functional status, particularly in relation to patients with chronic conditions. For health care planers/providers and policy makers, assessment of QoL may be important in their work to improve the quality of care or rehabilitation programs given, to facilitate communication with future patients, to address late problems in relation to expectations and to medical decision-making (Fayers & Machin, 2007).

However, there is neither a concrete definition nor a uniform consensus regarding the concept of QoL to be applied for all circumstances and contexts, particularly its operationalization, making the QoL an ambiguous notion. In this regard, there is a need to clearly specify what the term QoL means when it is used. Traditionally, many scholars use the term QoL interchangeably with health status and functional capacity. However, they are conceptually distinct issues and patients with impaired health and functional status do not necessarily experience poor QoL, a phenomenon that Albrecht and Devlieger (1999) have defined as "disability paradox". Although somatic ill-health may restrict physical or role function of the affected individuals, and physical impairment may interfere with social participation and interpersonal relationships, patients' perception of disease is often in discord with their subjective perception of QoL. This is so particularly among patients with chronic diseases where QoL usually goes far beyond disease-related concerns and patients use their health condition to reinterpret their lives (Albrecht & Devlieger, 1999). Indeed, physical functioning may affect one's QoL, but is not necessarily the real reflection of QoL, similar to other concepts such as happiness or achievement of personal goals, which are used to conceptualize QoL, concepts that are worth-while investigating and measuring, but not necessarily indicative of what QoL is (Bradley, 2001; Fayers & Machin, 2007).

In this regard, world health organization has developed a multi-dimensional definition for the concept of QoL as "individuals' perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns embedded in a cultural, social and environmental context" taking a holistic approach to health and health care (Skevington et al., 2004). It is stated that physical and social conditions may impede empowering people and not allow them to act as agents in their own lives, thus not having high QoL (Albrecht & Devlieger, 1999). According to world health organization, QoL is not a construct to measure "objectively the symptoms or disabilities, but a construct assessing the effect of disease and health care on one's life" (Skevington et al., 2004).

2.5.1. Quality of life and adults with CHD

Interest in the QoL of adults with CHD has increased. Similar to other patients with chronic conditions, adults with CHD can face physical health issues and psychosocial challenges. Most studies addressing QoL are from Western and/or developed countries and the majority involves health-related QoL. Yet, findings

are inconclusive or inconsistent. Some studies have shown that physical functioning and general health perception are the most affected domains of health-related QoL among adults with CHD compared with normal population samples (Kamphius et al., 2002a; Lane et al., 2002; Mokhles et al., 2011; Pike et al., 2012), whereas others report that the psychosocial well-being is also negatively affected by CHD (Rose et al., 2005) or patients have poorer QoL in all domains, except for social relationships (Rose et al., 2005). At odds with these findings, some investigators observed that adult CHD patients have excellent health-related QoL contrasted with standard population samples (Immer et al., 2005; Loup et al., 2009; Moons et al., 2006a), while others (Saliba et al., 2001) did not find any difference between CHD patients and healthy adults in terms of health-related QoL.

One of the interests in QoL research among adults with CHD has been the association of cardiac defect and perceived QoL, with the assumption that having more severe and complex heart defects are associated with poorer QoL. In this regard, Moons et al. (2005) have investigated the association of disease severity with QoL in terms of primary heart defect diagnosis, illness course and functional status. These authors found that the severity of CHD was marginally associated with QoL and health status of adults with CHD, although cyanotic patients had significantly lower health status than acyanotic patients. On the other hand, Loup and colleagues (2009) showed that cyanotic and acyanotic CHD patients had similar QoL. However, patients with the same diagnosis (e.g. transposition of the great arteries), but different surgical treatments (Senning/Mustard versus Switch procedure), experienced different levels of QoL, vitality and psychosocial functioning. Lane et al. (2002) also reported that patients with inoperable conditions had a poorer QoL than patients who underwent surgery or those who received medical therapy alone. Similarly, Vigl and colleagues (2011) observed that having a more severe heart defect was related to poorer health-related QoL, except in mental health domains.

Contradictory findings may pertain to differences in the conceptualization of QoL (e.g. health-related or overall QoL) and methodological issues such as small sample sizes, low response rates, heterogeneous measures (e.g. The Short Form (36) Health Survey, Analogue Linear Scale), different comparison groups (e.g. volunteers, normative data, age/gender matched) and included cardiac defects (e.g. single diagnosis, different diagnoses, treatment mode). Interestingly, although many

scholars are interested in QoL of adults with CHD, none of them have scrutinized whether "the presence of CHD" per se was independently associated with QoL.

To date, only a few studies have addressed the association of other factors than health-related variables such as demographics, socio-economic status, emotional distress or social support with QoL among adults with CHD. In this respect, some studies have shown that younger age was positively associated with better QoL in terms of physical, mental, social and general health components of health related QoL (Saliba et al., 2001; Vigl et al., 2011), while higher educational level was associated with better scores in physical, mental and general components of health related QoL (Saliba et al., 2001; Vigl et al., 2011) as well as health and life satisfaction (Vigl et al., 2011). According to a study by Moons et al. (2009), education, employment, health status and cardiac disease instability were negatively associated with poorer QoL.

In this thesis, the operational definition of QoL proposed by WHO was used. The issue of QoL of adults with CHD in comparison with non-CHD participants, while considering different factors (e.g. social support) associated with QoL of CHD patients is addressed in Study IV.

2.6. Life satisfaction

Satisfaction with life refers to "a subjective appraisal of one's personal life" indicating how satisfied a person is with her/his entire life (Moons et al., 2006b). Moons and colleagues (2006b) have argued that life satisfaction is the most suitable concept of QoL as it is a global, subjective and constant construct although it changes over time. These authors have defined QoL as " the degree of overall life satisfaction which is positively or negatively influenced by the individual's perception of life domains important to them, including matters both related and unrelated to health" (Moons et al., 2006a).

2.6.1. Life satisfaction and adults with CHD

Life satisfaction has been scarcely scrutinized in the research of adults with CHD. Pike et al. (2002) have shown that CHD patients who underwent Fontan surgery were as satisfied with their lives as were healthy counterparts, while Vigl and co-workers (2011) found that patients with severe cardiac defects experienced less life satisfaction. Moons et al. (2006a) have reported that CHD patients are more satisfied with their lives than healthy peers. The factors associated with more life

satisfaction have been shown to have younger age, higher education, and being employed (Vigl et al., 2011), and the absence of depression, better health condition and better social support (Pike et al., 2012).

In this thesis, satisfaction with life was conceptualized as subjective appraisal of personal life and the extent to which a person is satisfied with her/his entire life (Moons et al., 2006b). The issue of satisfaction with life of adults with CHD in comparison with non-CHD participants, while considering important factors (e.g. social support) associated with life satisfaction of CHD patients, is addressed in Study IV.

2.7. Socio-economic status

Socio-economic status is an important determinant of health. Those who are worse off socio-economically have worse health. It has been shown that unemployment is associated with physical and mental ill-health. Being unemployed negatively affects health through imposing poverty and financial strain, changing health-related behaviour, and is a stressful life event per se. Conversely, being employed has its own psychological benefits by providing physical and mental activity, interpersonal contacts and respect from others, better self-esteem and, better control over life and social status (Bartley et al., 2006; Green & Tones, 2012). Poverty and unemployment lowers the standard of living, predispose individuals to redundant exposure to physical and social health hazards, and restrict the access to essential health care. The financial strain caused by unemployment reduces social integration. These effects become particularly prominent in societies where low social benefits cannot compensate for the economic inconveniences of unemployment (Bartley et al., 2006; Green & Tones, 2012; Marmot, 2004) and result in excessive income inequalities.

On the other hand, where a person stands in the social gradient determines her/his health and life expectancy in all socio-economic strata and in terms of any social indicator such as housing tenure, educational group, and social and occupational class (Adler, 1994; Marmot, 2004; Shaw et al., 2006).

Indeed, relative poverty, referring to the destitute position of an individual compared to the norms of the community where she/he belongs, may exclude people from social and community life. Again, this phenomenon becomes

prominent in societies with higher levels of economic and social inequalities. Social exclusion itself may lead to extra psychosocial distress and material deprivation resulting in adoption of health-threatening behaviour. On the other hand, social exclusion by lowering social trust may lead to low social equity and cohesion which are of important determinants of health (Green & Tones, 2012; Marmot, 2004; Shaw et al., 2006).

2.8. Iran: the context of the study

Iran, located in the Middle East, is the 17th largest country in the world with an area of 1,648,195 Km^2 (Mehdrad, 2009). Iran has an estimated population of 74.8 million (UNFPA, 2011) with more than 9 million living in its capital, Tehran (Mehdrad, 2009). A third of the total population is younger than 15 years old and only 5% over 60 years old (Mehdrad, 2009), with an annual population growth rate of 1% (UNFPA, 2011). Iran has a gross domestic product (GDP) per capita of US $ 4,526 (World Bank, 2012) and a life expectancy at birth of 72/75 years (male/female) (World Health Organization, 2011). Although Iran is a middle-income country (World Bank, 2012), its economic situation has deteriorated during the past years due to political problems and the resulting international sanctions, which created high inflation, increased unemployment rate and poverty (e.g. http://www.guardian.co.uk/commentisfree/2012/oct/07/iran-santions-suffering, retrieved 11-06-2013), and thereby negatively influenced the citizens' welfare. Moreover, economic crises may influence the allocation of resources to social and health care services, which also influence the welfare of the citizens, mostly those with chronic disease in need of care continuity.

Figure 1. Map of Iran

2.8.1. Health care system in Iran

The Ministry of Health and Medical Education is responsible for health-related policy making and supervision in public and private sectors, and also for medical education. The public sector provides primary, secondary and tertiary health services through a nation-wide network, alongside the private sector which plays a considerable role in medical and rehabilitation care in urban areas (Mehdrad, 2009). In Iran, 71% of the population is urban dwellers (UNFPA, 2011). In the urban health care system, there are no gate-keeping, referral or counter-referral systems. There are multiple sub-health systems with different health plans and health providers, which prevent achieving a meaningful continuity of care (World Bank, 2008). A person can enter the health care system at any link in the chain. There are no available data about the magnitude of referrals, which may result in duplication of medical examinations, inadequate care and poor management of ill-health, specifically chronic conditions that need good continuity of care (Moghadam et al., 2012). The private sector, particularly in large cities such as Tehran, is in a dominant position concerning health care services. There is a lack of adequate and developed systems for regulating the capacity and behaviour of private health providers (Moghadam et al., 2012).

2.8.2. Health financing in Iran

Iran is considered to be a high middle-income country. In spite of high economic growth during the past years, inflation rate was 20.6%, the government spent 5.6%

of the GDP on health, according to the World Bank reports (2012). International sanctions are also new economic burdens.

Health services are either financed totally by government as in primary health care, which includes basic preventive services (e.g. immunization of children and pregnant women) or are financed by different health insurance organizations (e.g. social security organization) in medical benefit packages (Ibrahimipour et al., 2011). Although some data have reported (Mehrdad, 2009) that more than 90% of Iranians are covered by at least one kind of health insurance, out of pocket expenditure on health is high (Ibrahimipour et al., 2011). Furthermore, Ibrahimipour and colleagues (2011) showed that there is a lack of information on insurance rates that prevents effective policy making. They also have argued that regressive financing, high out of pocket expenditure on health and a shortage of standard benefit packages have resulted in lack of fairness in the insurance system.

2.8.3. Adults with CHD in Iran

The birth prevalence of CHD in Iran is reported as 8.9 per 1000 in Gorgan, a city in north of Iran (Nikyar et al., 2011) and as 12.3 per 1000 in Ahvaz, a city in southwest of Iran (Rahim et al., 2008). Although geographical variation exists, which could be due to the cultural differences such as more consanguinity marriages, prevalence is quite high in both regions.

The exact number of adults with CHD in Iran is unknown, as in other countries. One reason could be that many CHD defects, if not the majority, may be asymptomatic. Thus patients with CHD may not notice their symptoms and therefore not come into contact with health care until the cardiac defect is discovered accidentally or through late presentation of complications (Mocumbi et al., 2011). It may also be because there are no referral centres responsible for the care of adults with CHD, or that there is a lack of national or regional CHD databases or monitoring.

Similar to other countries, the medical management of CHD patients (e.g. cardiac surgery) has improved significantly during the past years and their survival into adulthood has increased in Iran (Nobakht et al., 2008). However, there are no data regarding the survival rate of children with CHD into adulthood thus making it difficult to estimate the prevalence of adults with CHD.

In contrast to advances in cardiology, there is a shortage of human resources trained to take care of adults with CHD, particularly in terms of their psychosocial situation. At best, they are treated by general/paediatric cardiologists or cardiovascular surgeons, and non-medical rehabilitation is almost non-existent. Furthermore, there is lack of data about the knowledge of health care staff (e.g. doctors) about the QoL and psychosocial well-being of adults with CHD. In fact, there are no studies in Iran addressing these issues. Studies about the well-being of adults with CHD (e.g. QoL) have been conducted mostly in Western/developed countries, and the findings are limited and conflicting. Furthermore, the transferability of current findings to Iran can be questioned as it differs from Western/developed countries in various ways such as health care system and culture; thus what may pertain to Western/developed countries may not be applicable to Iran. As an example, some data from Guatemala (Vida et al., 2006) have shown that atrial septal defect percutaneous occlusion compared to surgical closure in a low-income country is not as cost effective as the data reported from high-income countries. Additionally, there are hardly any studies on these topics from other low/middle-income countries that may serve as an example for Iran.

Review of the current literature reveals that the aforementioned issues have not attracted much attention among researchers, health care staff and health policy makers, not only in Iran but also in other developing countries. This shortcoming is likely to have serious negative consequences such as the absence of relevant services to meet the patients' needs.

2.9. Conceptual framework

The main conceptual foundation of this thesis is the biopsychosocial model of health. According to biomedical model, which defines health as the absence of the disease, none of the CHD patients may be considered healthy. While in terms of human health, the abnormality may be present, the patient may be healthy nevertheless. On the other hand, at best, the presence of a biomedical defect may be necessary, but not sufficient for the human experience of disease and illness (Engel 1977). Other elements (e.g. psychological and social) than a biomedical defect may contribute to the individual experience of disease and/or health.

It is a fact that the life expectancy of CHD patients has increased due to the enormous development of the biomedical approach. If today a new group of

patients exist, known as adults with CHD, this is the result of the medical approach which is able to palliate the anatomical and functional cardiac lesions among CHD patients. However, within this discipline, there is no room for social, psychological, and behavioural dimensions of health (Engel 1977). How health is conceptualized among adults with CHD determines the proper borders of health care professionals' responsibilities and also influences their attitudes and behaviour towards patients (Engel 1977). How health is conceptualized also determines how policy makers address the needs of this steadily growing group of patients, especially in the context of developing countries with limited resources.

To provide appropriate care to maintain health in the presence of a chronic condition such as CHD, the patients' personal experiences of disease and their social context and social lives should be taken into account. The biopsychosocial model, considering the influence of various elements (e.g. social support, socio-economic status) rather than giving primacy to biological factors alone, may possibly help us to understand why some CHD patients fare well and others not.

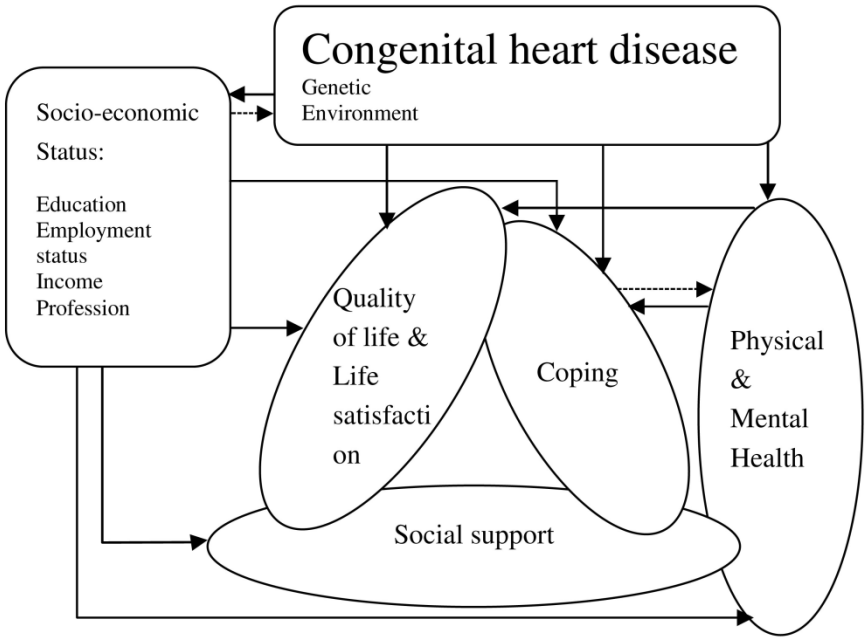

Figure 2. Conceptual map

According to this conceptual framework, it is hypothesized that CHD per se and CHD-related issues (e.g. undergoing cardiac operation) may have direct and indirect effects on the psychosocial situation of the affected individuals. Congenital heart disease, by superimposing internal (e.g. experienced uncertainties) and external (e.g. need for cardiac operation) demands, may lead to allostatic overload and may result in mental (i.e. anxiety and depressive symptoms) and physical (i.e. somatic symptoms) health problems, as well as temporarily influencing how people cope with uncertainties and unpleasant life events.

On the other hand, it is hypothesized that CHD, by inducing performance restrictions, difficulties in social integration and experienced discrimination (Claessends et al., 2005; Horner et al., 2000; McMurray et al., 2001), may impact on socio-economic status, QoL and life satisfaction of affected individuals. Socio-economic status per se is an important determinant of health, as higher socio-economic status corresponds to better health behaviours, and mental and physical health (Adler, 1994; Marmot, 2004). In this regard, it is hypothesized that socio-economic status can impact on mental and physical health, QoL and life satisfaction as well as styles of coping of adults with CHD. Accordingly, Lazarus and Folkman (1984) have described that the ways people cope are determined by the available internal and external resources.

Additionally, this conceptual map considers social support as an important asset in perceived mental and physical health, QoL, life satisfaction and styles of coping. However, social support itself can be influenced by socio-economic status.
Finally, it is important to state that how people cope with stressful life events may influence their perception of Qol and life satisfaction and might impact on their mental and physical health, a subject not addressed in this thesis.

2.10. Rationale

With a relatively high birth prevalence of CHD in Iran (mean 10.6%) in a population of 74.8 million, and increased survival rate of children born with CHD into adulthood, the well-being of adults with CHD becomes a considerable public health problem. Adults with CHD are a newly emerging but steadily growing group of patients in Iran and other developing countries as well as developed countries.

The main purpose of public health practice is to improve and promote the population's health, prevent diseases and minimize their consequences, prolong life and reduce health inequalities (Detels, 2009). It might not be possible to prevent the occurrence of CHD in the short-term but by an overall understanding of the psychosocial situation as well as quality of life of adults with CHD, and understanding the factors which contribute in their health outcomes, it will be possible to improve their health, to prevent potential psychosocial problems and to reduce the experienced health inequalities.

Therefore, this thesis aims to investigate the psychosocial situation of adults with CHD in Iran, a subject in which research is lacking. Most of the existing studies have been conducted in Western/developed countries, but their findings are limited and inconsistent. Further, questions can be raised about the transferability of such findings to Iran and even to other developing countries.

One expectation is that the data derived from this thesis will be able to help shed light on the current situation of adults with CHD in Iran, and serve as a reference value for future studies. In addition, this research may be useful to develop effective methods to treat/rehabilitate adults with CHD with psychosocial morbidities, but also to build-up preventive interventions for those at risk. Besides, such data may be valuable to develop methods to empower CHD patients in order to cope better with their physical and social challenges. This research may help us to understand the contribution of socio-economic factors on the different health outcomes among this group of patients, an important interest in public health practice. Also, the data from this thesis may help health policy makers and health providers in their efforts to provide the appropriate medical and social services for adults with CHD, services that are responsive to the real needs of this group of patients. Finally, the findings from this research might be used as a model for other developing countries suffering from economic deprivation, social inequities and health inequalities.

3. OBJECTIVES

The main objectives of this thesis were to understand the psychosocial situation of adults with CHD in Iran and to determine the factors which may contribute to enhancing or reducing their psychosocial well-being.

3.1. Specific objectives

(a) To study the CHD-related health condition and socio-economic status of adults with CHD with respect to sex differences (Study I).

(b) To assess and compare the mental health (e.g. depression) and somatic symptoms experienced by adults with and without CHD, and to identify factors associated with the mental health (e.g. depression) and somatic symptoms of all participants and separately for each group (Study II).

(c) To assess and compare the coping strategies used by adults with CHD and non-CHD persons and to identify factors (e.g. social support) associated with the styles of coping of all participants and separately for each group (Study III).

(d) To assess and compare the quality of life and life satisfaction of adults with and without CHD and to identify factors (e.g. demographics) associated with quality of life and life satisfaction of all participants and of CHD patients separately (Study IV).

4. METHODS

4.1. Design

This research uses a quantitative approach through a cross-sectional case-control design.

4.2. Setting and participants

The research was conducted at the Tehran Heart Center in collaboration with Shahid Rajaee Heart Hospital which are two public and university affiliated specialized hospitals in Tehran. The participants were female and male adults with CHD who met the following criteria: (a) hospitalized due to CHD between April 2002 and March 2010; (b) aged 18-64 years at the time of study; (c) not suffering from Marfan syndrome; (d) not mentally impaired (e.g. Down's syndrome); (e) living in Tehran province; (f) able to read and write Persian; and (g) willing to participate in the study.

The archives of the aforementioned hospitals provided a list, phone number and addresses of the patients with these given inclusion criteria. Sixty-one patients suffering from isolated bicuspid aortic valve, isolated dextrocardia, isolated patent foramen oval, common atrium, malformation of coronary arteries, malformation of the cardiac septum, aorto-pulmonary fistula, arterio-venous fistula, partial anomalous pulmonary venous connection, small ventricular septal defect, hypertrophic cardiomyopathy or primary pulmonary hypertension were excluded because of the very small number of patients in each group or due to difficulty in categorizing them. The number of available adults with CHD became 853. Of these patients, it was not possible to access 480 of them because of incorrect telephone numbers or because they had moved without leaving forwarding information. Although several attempts (several calls at different times of the day on different days of the week) were made, it was not possible to reach these patients. Of those patients that could be contacted, 26 refused to participate. Thus, the final sample of adults with CHD consisted of 181 females and 166 males (total 347, Figure 3) with a response rate of 93% (Study I-IV).

For the comparison group, 353 adults without CHD who were matched by sex and age (± 2 years) to the patients were recruited. This comparison group was randomly selected from the same residential area as the patients. Furthermore,

these participants were not suffering from cognitive deficiencies and were able to read and write Persian. Thus, the final overall sample of the study consisted of 700 persons (Study II-IV).

Figure 3. Patient flow diagram

4.3. Procedure

Patients meeting the inclusion criteria were contacted consecutively and provided with detailed information about the purpose and the process of the study. If they agreed to participate, an appointment was made for a trained researcher to visit them at their home to provide complementary oral and written information. If they still wished to participate, they were given the questionnaire and consent form. The participants could keep the questionnaire for 4-7 days in order to answer the questions. At the end of the first meeting a new appointment was made to collect the questionnaires (Study I-IV).

A systematic randomization procedure was used to select adults without CHD. First, it was determined "how many" non-CHD participants, "which sex", "which age range" were needed in each residential area. Then one alley in each residential area was chosen at random. The first call invited a man, and the second a woman, participant and so on. Each contact could have three outcomes: the right participant in the right age range was found; the person with that specific inclusion criterion was not living in that address; or the person was not interested in participating in the study. If the appropriate participant was not found or if she/he

was not interested in participating in the study, the process continued as described above until all needed non-CHD participants were found. This process was performed only during weekends in order to make sure that employed and unemployed individuals had an equal chance of being contacted.
After selecting non-CHD participants, the procedure of delivering and collecting the questionnaires were the same as with the CHD patients (Studies II-IV).

4.4. Measures

The participants answered a self-report questionnaire covering various areas such as:

4.4.1. Socio-demographic variables consisted of age, sex, marital status (never married, married, divorced/widowed), having children (yes/no), educational level (categorized as illiterate or some informal education or primary school or similar, secondary school or high school or similar, university or similar), being employed (yes/no), being on sick-leave during past year (yes/no), profession (categorized as blue-collar worker (e.g. simple worker), low white-collar worker (e.g. clerk), middle/high white-collar worker (e.g. nurse), annual income (categorized as less than 48,000,000 Iranian Rials (IRR), between 48,000,000 and 72,000,000 IRR, between 72,000,001 and 96,000,000 IRR, more than 96,000,000 IRR, and don't know/don't want to disclose; 1 US dollar≈10,000 IRR at the time of data collection), and financial strain (categorized as never, quite often, often, always).

In Studies III and IV, the following variables were re-categorized: educational level (informal/primary/guidance school/similar = low, high school/similar = middle, university/similar = high), annual income (less than 48,000,000 Iranian Rial (IRR) = low; 48,000,000 to 96,000,000 IRR = middle; more than 96,000,000 IRR = high) and financial strain (quite often/often/always=yes, never=no). This was carried out because of the small number of participants in some of the categories or for analysis purposes (e.g. better interpretation of findings).

4.4.2. Health behaviour variables were: body mass index (BMI) calculated for each participant with the standard formula as weight (kg.) divided by the square of height (m.) based on self-reported height and weight; dieting categorized as consumption of fruit/vegetables, meat/fish, dairy (e.g. milk), grains (e.g. rice) at least three times a week; physical activity (e.g. walking; at least 30 minutes, three

times a week); and substance use including cigarette smoking (daily), regular tobacco smoking by water-pipe, regular alcohol and daily opium use.

4.4.3. Anxiety and depressive symptoms were assessed with the Persian version of The Hospital Anxiety and Depression Scale, HADS (Montazeri et al., 2003; Zigmond & Snaith, 1983). The scale comprises 14 items; each item is graded on a four-point (0-3) scale response category. Seven items are about anxiety symptoms (e.g. I get sudden feelings of panic) and seven items about depressive symptoms (e.g. I feel as if I am slowed down). The range for each subscale is 0-21. A score of 0-7 corresponds to no cases, 8-10 to possibly cases and 11-21 to probable cases of anxiety and depression. High scores correspond to high level of anxiety and depressive symptoms. Cronbach α was 0.86 for total HADS, 0.81 for anxiety, and 0.79 for depressive symptoms.

4.4.4. Somatic symptoms were assessed using the short version of the Giessen Complaint List (Brähler & Scheer, 1995). The scale includes 24 items and each item is scored on a 5-point scale from 1 (not at all) to 5 (very much). The symptoms are organized in four domains with six items in each: exhaustion (e.g. tiredness); gastrointestinal (e.g. nausea); musculoskeletal (e.g. pains in joints or limbs); and heart distress (e.g. heavy, rapid or irregular heart-throbbing). The total score ranges from 0-96, and 0-24 in each symptom domain. The higher the score the more one is affected (total/ domains). Cronbach α was 0.92 for the total scale, 0.83 for exhaustion, 0.74 for gastrointestinal, 0.81 for musculoskeletal and 0.80 for heart distress symptoms.

4.4.5. Social support was addressed with the Multidimensional Scale of Perceived Social Support (Zimet et al., 1988). This consists of 12 items covering three domains: family (4 items, e.g. I can talk about my problems with my family), significant other (4 items, e.g. There is a person in my life caring about my feelings) and friends (4 items, e.g. I can count on my friends when things go wrong). Each item is scored on a 7-point scale from 1 (very strongly disagree) to 7 (very strongly agree) and the total score ranges 12-84. The higher the score, the higher the perceived social support (total/domains). Cronbach α for the total scale was 0.88.

4.4.6. Style of coping was assessed with Utrecht Coping List-short form (Schreurs et al., 1988), with 19-items organized into five domains: active problem focusing (5

items, viewing the situation from different angles and try to solve the problem targeted); palliative reaction pattern (4 items, diverting attention from the problem); seeking social support (5 items, asking for help or comfort and understanding); avoidance behaviour (3 items, avoiding the problematic situation or let the situation run its course); and expression of emotion (2 items, showing frustration or anger). The first three domains indicate problem-focused and the last two domains represent emotion-focused styles of coping. Each item is scored on a 4-point scale from 1 (seldom or never) to 4 (very often). Cronbach α for total coping scale was 0.73.

4.4.7. Quality of life was assessed via The World Health Organization Quality of Life Short-Form (Skevington et al., 2004) including 26 items rated on a 5-point scale from 1 to 5. The first two items are about overall QoL (i.e. How would you rate your quality of life?) and satisfaction with health (i.e. How satisfied are you with your health?). The remaining 24 items are structured according to four-domains: physical capacity (7 items, e.g. Do you have enough energy for everyday life?); psychological well-being (6 items, e.g. How much do you enjoy life?); social relationships (3 items, e.g. How satisfied are you with your personal relationships?); and environmental health (8 items, e.g. How available to you is the information that you need in your day-to-day life?). After transforming the scores into a scale from 0 to 100 for each domain, high scores correspond to a high level of QoL. Cronbach α for the total QoL scale was 0.92, 0.81 for physical capacity, 0.81 for psychological well-being, 0.59 for social relationships and 0.80 for Environmental health. The relatively low Cronbach alpha for social relationships can be due to small number of items (3 items) in this domain.

4.4.8. Life satisfaction was addressed by The Satisfaction With Life Scale (Diener et al., 1985), which contains 5 statements (e.g. In most ways my life is close to my ideal) graded on a 7-point scale ranging from 1 (strongly disagree) to 7 (strongly agree). The total score ranges 5-35. High scores correspond to high life satisfaction. Cronbach α was 0.88.

4.4.9. Medical variables for CHD patients were obtained from the hospital records such as type and number of heart defects, or were collected through self-reported questions as follows: the number of surgical operations or angiographies; cause (e.g. arrhythmia) and number of hospitalisations other than surgery or

angiography; type and number of cardiac medications used; time of CHD diagnosis which was categorized into infant (<2 years), child (2-11 years), teenager (12-17 years) and adult (≥18 years); place of first time CHD diagnosis categorized as hospital, primary care centre, and other places; the presence of other diseases than CHD including asthma/allergy, diabetes mellitus, cancer, hepatic, gastrointestinal and pulmonary or other diseases; the presence of CHD in the family/relatives; out-patient visit intervals; type of specialist responsible for the CHD patients' health care; and light physical activity (e.g. walking, at least 30 minutes three times a week) and heavy physical activity (e.g. volleyball) limitations due to CHD. Physical activity was considered limited if patients reported moderate to considerable limitations.

To assess whether CHD patients had knowledge about their heart defect, a list of Latin and Persian names of different types of CHD was provided. Thereafter, the patients were asked to select one or several alternatives. In case the patients could not find the name of their disease, they could write it down or explain it in their own words.

Participants without CHD also answered questions regarding the presence of chronic health problems such as asthma, diabetes mellitus, cancer, hepatic, gastrointestinal and pulmonary diseases or other diseases (e.g. arthritis).

4.5. Classification of the CHD

Because of the diversity in the complexity of CHD and the limited number of patients in each cardiac defect category, a common problem which exists in almost all studies related to this patient population, CHD patients were classified in three hierarchical groups (or blocks) as inspired by Marelli et al. (2007), which fitted current sample well. The groups were as follows: Group I (n=83), severe lesions included atrio-ventricular canal defect, tetralogy of Fallot, univentricular heart, transposition of the great vessels, truncus arteriosus and hypoplastic left heart syndrome; Group II (n=186), shunt lesions included atrial septal defect, ventricular septal defect, patent ductus arteriosus, aortic coarctation and Ebstein's anomaly; Group III (n=78), valve anomalies included anomaly of pulmonary artery and valve, congenital tricuspid valve disease, congenital aortic and mitral valve stenosis and insufficiency.

Table 1. Overview of designs and methods used in the thesis

Study	I	II	III	IV
Outcome	Sex differences in adults with CHD	Anxiety, depressive and somatic symptoms and their determinants	Styles of coping and their determinants	Life satisfaction and quality of life and their determinants
Design	Cross-sectional -descriptive	Cross-sectional, case-control -descriptive -analytical	Cross-sectional, case-control -descriptive -analytical	Cross-sectional, case-control -descriptive -analytical
Data collection	Hospital records Questionnaire	Hospital records Questionnaire	Hospital records Questionnaire	Hospital records Questionnaire
Sample	347 adults with CHD	-347 adults with CHD -353 non-CHD participants	-347 adults with CHD -353 non-CHD participants	-346 adults with CHD -353 non-CHD participants
Data analysis	Chi-square test Fisher's exact test Student's *t*-test	Chi-square test Student's *t*-test Mann-Whitney U test Block-wise Linear Regression	Chi-square test Student's *t*-test Mann-Whitney U test Block-wise Linear Regression	Chi-square test Student's *t*-test Mann-Whitney U test Linear Regression

CHD indicates congenital heart disease

4.6. Statistical analyses

The Predictive Analytics Software statistic packages 18.0 and 19.0 (IBM/SPSS Inc., Chicago, IL) were used to analyse the data. The categorical variables are presented with absolute frequencies and percentages, the normally distributed continuous variables by mean and standard deviation, and non-normally distributed continuous variables by median.

4.6.1. Bivariate analyses

Categorical variables were compared with *chi*-square test or *Fisher's* exact test. Normally distributed continuous variables were compared with Student's *t*-test or one-way analyses of variance (ANOVA) across the groups, and non-normally distributed variables with Mann-Whitney U test. The significance level was set at $p<0.05$.

In Study I, Women were compared with men, and also the groups defined by defect category. In Study II, III and IV, adults with CHD were compared with non-CHD participants.

4.6.2. Multivariate analyses

To scrutinize factors associated with certain styles of coping, life satisfaction, quality of life, and anxiety, depressive and somatic symptoms, multiple linear regression analyses were performed (Studies II, III and IV). Dummy coding was used for categorical variables with more than two categories. The data were expressed in the form of Betas (β), 95% Confidence Intervals, p-values and R square. The significance level was set at $p<0.05$.

Study II. Block-wise multiple linear regression analyses were conducted to estimate the association of independent variables with anxiety, depressive and somatic symptoms (dependent variables). As the somatic symptoms variable was not normally distributed, logarithm transformation was performed.

For the regressions, the independent variables were factors that differentiated significantly the patients and the comparison group in the bivariate analyses. The independent variables consisted of parenthood, educational level, employment, annual income, profession, sick-leave during the past year, financial strain, BMI and water-pipe use. Moreover, age, gender, social support and when applicable concomitant anxiety/depressive/somatic symptoms were added as independent variables into the models. First, a block-wise multiple linear regression analysis was performed among all participants (n=700) with the aforementioned independent variables fitting in four blocks as *demographics/socio-economics*, *health behaviour*, *health* (i.e. anxiety, depression, somatic symptoms) and *perceived social support*. Additionally, *the presence of CHD* was added as a fifth block to examine whether CHD in itself was independently associated with anxiety, depressive and somatic symptoms. In the next step, block-wise multiple linear regression analyses were performed separately for the CHD patients and for the non-CHD participants with the same independent variables in the same blocks (except the presence of CHD block). The patient group had an extra block including CHD-related variables, i.e. the severity of the cardiac defects, time of CHD diagnosis, history of CHD hospitalization, operation and angiography, and use of heart-related medications.

Study III. Block-wise multiple linear regression analyses were performed to scrutinize factors (independent variables) associated with each domain of coping style (dependent variables). The dependent variables were: active problem focusing, palliative reaction pattern, seeking social support, avoidance behaviour, and expression of emotion. The independent variables were selected based on a review of the literature (Lazarus & Folkman, 1984; Mayes et al., 2000) as well as the factors differentiated significantly the CHD and non-CHD participants in previous analyses (Study II).

Independent variables were introduced into the models in three blocks as: *demographic/socio-economic block* including age, sex, marital status, parenting, educational level, employment, profession, annual income, financial strain; *psychological adjustment/social support block* including anxiety, depressive and somatic symptoms, perceived social support; and a *CHD block* in block-wise multiple linear regression analyses which were performed for all participants to scrutinize whether CHD per se was associated with adoption of a certain style of coping. Two more block-wise multiple linear regression analyses were performed for both CHD patients and non-CHD participants. In the former, independent variables in the CHD block included severity of the disease, time of CHD diagnosis, history of previous surgery, catheterization, and hospitalization due to cardiac defect, and cardiac medication use. In the latter, models were constructed based on two blocks as *demographic/socio-economic* and *psychological adjustment/social support*.

Study IV. To estimate the association of independent variables with QoL/life satisfaction (dependent variables), multiple linear regression analyses were conducted in two steps. First, a regression analysis was conducted among all participants (n=699). The independent variables were factors that differentiated the CHD patients and non-CHD participants in the bivariate analyses (i.e. parenting, educational level, employment, annual income, profession, financial strain, BMI and water-pipe use). Additionally, age, sex, emotional distress (i.e. anxiety and depression), somatic symptoms and perceived social support were added as independent variables into the models. To scrutinize whether the perception of QoL/life satisfaction was independently associated with the disease itself, "the presence of CHD" was entered as an independent variable into the model. In the second step, a regression analysis was performed with only CHD patients (n=346) using the same independent variables. To scrutinize the association of the CHD-

related variables with QoL/life satisfaction, the medical variables (i.e. severity of the cardiac defects, time of CHD diagnosis, the history of CHD hospitalization, operation and angiography, use of heart-related medications) were introduced into the models.

Missing data regarding the different scales were handled according to the instructions of each instrument. However, the participants and researchers went through the questions at the time of collecting the questionnaires, which reduced the probability of missing data as much as possible. Nonetheless, one patient was excluded from the analyses in study IV due to a considerable amount of missing data in the quality of life scale. Thus, the number of CHD participants in Study IV was 346.

4.7. Ethical considerations

All studies in this thesis were approved by the Ethical Review Board of the Tehran Heart Center, Tehran, Iran (Ref nr. 74). The studies considered the ethical principles in medical research involving human participants according to World Medical Association Declaration of Helsinki (2000), including avoiding any potential harm, intentions to benefit and respect the autonomy of the participant as well as maintaining confidentiality. In this regard, all the participants were informed orally and in writing about the aims and the process of the research in a language understandable by lay people. It was clearly stated that their participation was voluntary and they could decline participation or withdraw their consent at any point in time with no consequences on their future medical care provided by the hospitals. Anonymity and confidentiality were emphasized. The participants received informed consent with the questionnaires. The CHD patients provided written consent and non-CHD participants gave oral consent.

5. RESULTS

The main findings are presented in Tables 2, 3, and 4 in the appendix.

5.1. Demographic and socio-economic status

Three hundred and thirty four adults with CHD (mean age, 33.24 ± 12.11 years; median age, 29 years) were enrolled in this study (52.2% women). Contrasted to men, women were older, more often married and more likely to have children, although educational level and annual income were similar. The main source of financial support for men was work, whereas women were financially supported by their spouses. Men were more often employed and on sick-leave than women. Lower employment rate was observed among men with more complex heart defects, whereas the employment rate of women was similar across the defect categories (Study I).

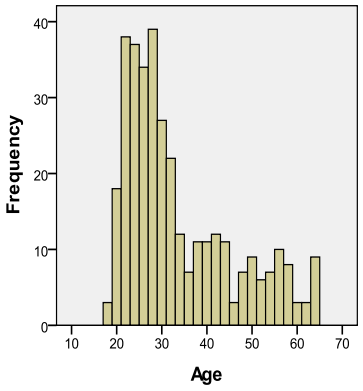

Figure 4. The age distribution of adult CHD patients (n=347)

Three hundred fifty three adults without CHD (mean age 33.49 ± 12.18 years; median age, 29 years) participated in this study (51.6% women). The non-CHD participants compared with the CHD patients were more likely to have children, had a higher educational level, greater annual income, better job position, and experienced greater financial strain. They were also more often employed and had been less often on sick leave (Studies II, III and IV).

5.2. Health behaviour and clinical variables

Health behaviour was similar among adults with and without CHD, except for those non-CHD participants who more often smoked tobacco by water-pipe and had a higher BMI (Studies II and IV).

Among adults with CHD, men had worse health behaviour (e.g. smoking cigarettes) and diet than women. Body mass index was similar across the two sexes. However, it was lower in patients with a high educational level, particularly women, and among those with a lower job position (Study I).

Among adults with CHD, more men than women had complex cardiac defects (i.e. disease block I) and valve diseases (i.e. disease block III). Body mass index was lower in patients with more complex cardiac defects compared with other CHD patients. Women and men were statistically similar in terms of the number of cardiac defects, undergone cardiac operations and angiographies, medication use and the number of medications.

The mean age for CHD diagnosis was 17.79±16.75 years. Diagnosis in men was made at a younger age. Congenital heart disease was diagnosed mostly in an inpatient setting (57%), medical care was mostly provided by the cardiologists (65%) and about 11.5% of patients did not have regular clinical follow-up visits. A majority of patients (87.4%) had out-patient visits at least once a year (similar for both sexes). Half of the patients knew the type of their cardiac defect, while the rest either lacked correct information or had no idea about it (similar for both sexes). Women with CHD reported more limitation in light physical activities, while both sexes experienced similar level of limitation in heavy physical activities (Study I).

There were no differences between adults with and without CHD regarding illness such as diabetes mellitus, cancer, hepatic and gastrointestinal diseases. However, patients with CHD experienced more pulmonary (4.6% vs. 1.1%, p=0.006) and other (21.3% vs. 15.3%, p=0.039) diseases than non-CHD participants.

5.3. Anxiety, depressive and somatic symptoms

The CHD patients (n=347) reported higher scores in anxiety (9.22 ± 4.40 vs. 8.11 ± 4.22) and somatic symptoms (median 14 vs. 8) than non-CHD participants (n=353). Depressive symptoms were similar across the groups (6.51 ± 4.30 vs. 6.36 ± 3.98).

However, multiple linear regression analyses among all participants showed that CHD was associated independently only with somatic symptoms (β =0.19, CI= 0.26 – 0.55, $p<0.001$).

Factors associated with anxiety, depressive and somatic symptoms

Among CHD patients, anxiety and depressive symptoms were associated with perceived financial strain, lower perceived social support and greater scores in somatic symptoms. The model explained 44.3% and 42.7% of the variance in anxiety and depressive symptoms, respectively. In the comparison group, in addition to aforementioned factors, anxiety symptoms were associated with female sex and depressive symptoms were associated with lower level of education. Among CHD patients, somatic symptoms were associated with lower annual income, perceived financial strain, and greater scores in anxiety and depressive symptoms. The model explained 42.9% of the variance in somatic symptoms. In the comparison group, in addition to these factors, being female was associated with somatic symptoms while primary education was protective. None of the medical variables were associated with anxiety, depressive and somatic symptoms.

5.4. Style of coping and social support

The CHD patients (n=347) were similar to non-CHD participants (n=353) in different styles of coping, except for palliative reaction pattern (8.31 ± 2.03 vs. 8.72 ± 1.93). Moreover, both groups were similar in perceived social support (total and each domain). Multiple linear regression analyses among all participants revealed that there is no association between CHD per se and style of coping, except for palliative reaction pattern (β =-0.10, CI= -0.73 – -0.10, $p<0.05$).

Factors associated with styles of coping

Among CHD patients, an active problem-focusing style was associated with being never married, having children, higher anxiety and somatic symptoms, lower depressive symptoms, and higher social support. Palliative reaction pattern was significantly associated with being unemployed, lower depressive symptoms and higher social support. Seeking social support style was significantly associated with higher anxiety symptoms and perceived social support. Avoidance behaviour style was significantly associated with not having a high annual income. Expression of emotion style was significantly associated with experiencing financial strain and having higher level of anxiety. Having a high or low annual income was negatively associated with expression of emotion style. The models

could explain 18.6% of the variance in active problem focusing, 16.6% in palliative reaction pattern, 20.9% in seeking social support, 10.4% in avoidance behaviour and 18.9% in expression of emotion.

Among non-CHD participants, active problem focusing was positively associated with female sex and employment status, and negatively associated with low educational level. The model explained 10.9% of the variance in active problem focusing. Palliative reaction pattern was significantly associated only with higher scores in social support and seeking social support with female sex, younger age, and higher scores in social support. The models could explain 4.9% and 12.6% of the variance in the palliative reaction pattern and in the seeking social support, respectively. However, avoidance behaviour was significantly associated only with lower scores in social support and expression of emotion with female sex and higher scores in anxiety symptoms. The models could explain 8.8% and 14.6% of the variance in avoidance behaviour and the expression of emotion, respectively.

5.5. Life satisfaction and quality of life

The CHD patients (n=346) compared to non-CHD participants (n=353) had lower scores in overall QoL (3.36 ± 0.82 vs. 3.57 ± 0.78) and health satisfaction (3.31 ± 0.93 vs. 3.71 ± 0.86). They also scored lower in the physical (59.65 ± 17.69 vs. 66.19 ± 15.39), psychological (56.33 ± 18.72 vs. 60.34 ± 16.82), and environmental (50.95 ± 15.86 vs. 53.50 ± 15.72) domains of QoL. Additionally, the CHD patients were less satisfied (19.48 ± 7.80 vs. 21.33 ± 7.60) with their lives.

Multiple linear regression analyses among all participants showed that CHD was associated independently only with overall QoL (β =-0.08, CI= -0.23 – -0.01, $p<0.05$), physical health (β =-0.08, CI= -4.92 – -0.77, $p<0.01$), health (β =-0.15, CI= -0.40 – -0.14, $p<0.001$) and life (β =-0.08, CI= -2.24 – -0.27, $p<0.05$) satisfaction.

Factors associated with life satisfaction among CHD patients

Overall QoL was positively associated with being female, being employed, no financial strain, lower scores in emotional distress, and higher scores in social support. Health satisfaction was negatively associated with financial strain, high scores in emotional distress, and high scores in somatic symptoms, but it was positively associated with having children.

Physical health was negatively associated with older age, high scores in emotional distress and somatic symptoms and a history of hospitalization due to cardiac complications, but positively associated with being employed. Psychological health was positively associated with younger age, having children, no financial strain, lower scores in emotional distress, and higher scores in social support. Social relationships were positively associated with being female, younger age, having children, lower scores in emotional distress, higher scores in social support, and a negative history of hospitalization due to CHD. Environmental health was positively associated with being female, employment, no financial strain, lower scores in emotional distress and in somatic symptoms and higher scores in social support, but negatively associated with low educational level and a history of cardiac surgery. Life satisfaction was positively associated with being female, employment, not having a better job position, higher BMI, lower scores in emotional distress, higher scores in social support, and a history of cardiac catheterization. The models could explain 44.1% of the variance in overall QoL, 36.4% in health satisfaction, 53.5% in physical health, 62.4% in psychological health, 41.7% in social relationships, 50.8% in environmental health and 51.9% in life satisfaction.

6. DISCUSSION AND CONCLUSIONS

6.1. Reflections on main findings

Overall, adults with and without CHD were similar in terms of health-related behaviours (Table 2, Appendix), although the majority of patients (≈ 90%) visited health care professionals at least once a year and medical care was provided mostly (≈ 75%) by specialists (Study I). The data yielded from Study I showed that only half of the patients knew about the type of their disease. According to these findings, one can question the education about CHD provided to the patients. Of course it is not possible to assess patient education or perceived informational support only by asking one question. However, this single item can be used as a proxy to enquire, if health care providers have not given the information about the disease itself, how probable it is that they could supply the information about the course of the disease which is one of the sources of uncertainties and stress for the patients. Furthermore, how can one be sure that enough information about potential cardiac complications (e.g. infective endocarditis), family planning and risk of pregnancy, and healthy behaviour (e.g. safe and appropriate level of physical activity compatible with limitations) is provided. However, it is possible that information was provided, but not in a language readily understandable by the patients. Rönning and colleagues (2008) showed that a two-way communication with a simple, clear and understandable language is what patients require.

Moreover, similar to a previous study (Goossens et al., 2013), CHD patients with a higher educational level were more knowledgeable about their disease than those with a lower educational level in the present sample. With present data it is not possible to ascertain whether the provided information was more understandable for the highly educated patients, or whether they were more eager to ask about their diseases, or had obtained information from other resources than health care professionals. Goossens and colleagues (2013) found that even highly educated CHD patients who received structured education, have relatively low levels of knowledge, which emphasizes the need for alternative and complementary forms in patient education other than verbal methods.

Health education is an initial and important factor in health promotion and public health practice, which needs to be addressed precisely by health care professionals.

However, health care professionals lacking specific training caring for adults with CHD may not be able to provide the appropriate information; this requires attention from policy makers.

6.1.1. Anxiety, depressive and somatic symptoms

Adults with CHD reported greater levels of anxiety and somatic symptoms than non-CHD participants, while there were no differences regarding depressive symptoms (Study II). However, the presence of cardiac disease contributed independently only in relation to somatic symptoms. These findings could simply indicate that adults with CHD are not receiving adequate medical care and suffer from marked somatic symptoms. It is also possible that the findings reflect a distorted body image perception, in particular body rejection, which has been shown to be associated with somatic symptoms (Geyer et al., 2006). Moreover, the findings could relate to CHD patients being hypersensitive/hypervigilant to their body sensations or interpreting their body feelings incorrectly (Karsdorp et al., 2008; Norozi et al., 2006). The independent association of CHD with mental health and somatic symptoms was not addressed in previous studies, making the comparison of findings a difficult task. However, the present results are in line with some studies (Brandhagen et al., 1991; Kovacs et al., 2009; Popelova et al., 2001; Saliba et al., 2001) and at odds with others (Bromberg et al., 2003; Ong et al., 2011). Discrepancies between the present findings and those of others may be due to methodological differences (e.g. sample characteristics).

In this context, it is important to consider that although anxiety was not independently associated with CHD, a substantial proportion of adults with CHD (≈ 40%) reported symptom levels indicating the presence of probable clinical cases of anxiety, calling for the implementation of appropriated interventions (e.g. cognitive behaviour therapy).

6.1.2. Style of coping

The current results indicate that adults with and without CHD adopted similar styles of coping (Study III), which is in line with a study from the Netherlands (van Rijen et al., 2004b). Moons & Norekval (2006) have suggested that those grown-up with CHD develop a strong sense of coherence, which may help them to cope well with their disease. One may argue that having a life-threatening disease and being involved in making serious health- and/or life-related decisions, e.g. undergoing operation (Kovacs et al., 2009) may result in *comprehensibility*. Moreover, being a

"high achiever" and having a good social support, e.g. family protection (Horner et al., 2000) may make the disease and life *manageable*. Also, surviving a life-threatening disease may increase life appreciation and *meaningfulness*. The aforementioned three components may thus help to develop a strong sense of coherence (Antonovsky, 1987; Moons & Norekval, 2006). On the other hand, Saliba and colleagues (2001) have argued that CHD patients adapt well with their disabilities because they set new values in their lives.

6.1.3. Social support

Adults with and without CHD did not differ in terms of perceived social support (Study III). Few studies have addressed this issue, but the present findings are in accord with those of a study from the United States (Pike et al., 2012) and contrary to those of a study from the Netherlands (van Rijen et al., 2004b) reporting that adults with CHD receive less social support than their healthy counterparts. This contradiction in findings may pertain to, in the study from the Netherlands, social support being measured as interactions and discrepancies in receiving social support. Another explanation may be cultural differences, which could also explain the results of the study from United States where nearly 40% of the sample consisted of people from other cultures than Anglo-Saxon (e.g. Hispanics). It has been reported that in collectivistic cultures, for instance Iranian, family attachment and interpersonal relationships are highly valued, while individualistic cultures are oriented around the self, and independence and individualism are central (Adams & Plaut, 2003; Markus & Kitayama, 1991). Additionally, the findings of Study III are reinforced by those of Study IV showing that adults with CHD do not differ from non-CHD participants regarding satisfaction with their social relationships.

6.1.4. Quality of life and life satisfaction

The results indicate that adults with CHD had a poorer QoL (overall, health satisfaction, physical, psychological and environmental domains) and life satisfaction in comparison with the non-CHD participants (Study IV), which is in line with a previous study (Rose et al., 2005) and at odds with others (Loup et al., 2009; Saliba et al., 2001; Silva et al., 2011). The disparate findings may be due to, for example, differences in sample characteristics (e.g. age of the participants) and in the measures used (health-related QoL vs. subjective QoL). The current findings indicate that adults with and without CHD differed notably in the psychological and physical domains of QoL, validating the results of Study II, in which patients

experienced higher levels of anxiety and somatic symptoms than non-CHD participants. Accordingly, the presence of cardiac disease was independently associated only with the overall, and general and physical health domains of QoL and life satisfaction. These results indicate that there is a need for measures to improve the QoL of adults with CHD, particularly in relation to psychological and physical well-being.

6.1.5. Factors associated with anxiety, depression, somatic symptoms, style of coping, life satisfaction and quality of life

6.1.5.1. Demographic and socio-economic factors

The results indicate that women and men with CHD differed only in age, marital status, parenthood and employment (Study I). Women with CHD were more often older, married and parents than men, which is in line with a Finnish study (Nieminen et al., 2003). Although educational levels were similar among women and men with CHD, women were less often employed and more financially dependent than men (Study I). Nevertheless, female sex was positively associated with greater life satisfaction and better overall QoL as well as in the social and environmental domains (Study IV). One possible explanation could be that women with CHD in the current sample were less influenced by direct and indirect economic burdens of the disease because they were financially supported mostly by their parents or spouses and experienced less financial strain than men. Thus, their perception of life satisfaction and QoL may be less distorted by economic problems. The current findings are at odds with some previous results showing that women with CHD have poorer QoL in physical (Chen et al., 2011) and mental health domains (Vigl et al., 2011). One could hypothesize that these findings pertain to cultural differences as the Iranian patriarchal culture where women are expected to have a high level of role functioning to prove their competence. The high expectations in role functioning may result in high levels of self-worth and self-satisfaction, when the expected responsibilities are accomplished. However, it is possible that the aforementioned high role expectations resulted in the overestimation of QoL among the women with CHD. Additionally, the current findings could reflect that social roles such as being spouse and parent mediate the association between sex and better QoL/life satisfaction among women with CHD: Women who experienced parenthood, to be involved in children's concerns, for example school issues, are more socially engaged, and thus they may have better perception of social relationships.

The results of Studies II and III showed that among adults with CHD gender was not related to anxiety, depression and somatic symptoms as well as styles of coping, which is in line with some previous studies (Cox et al., 2002; Geyer et al., 2006; Kovacs et al., 2009; Ong et al., 2011). Only two studies from the Netherlands (Utens et al., 1998; van Rijen et al., 2004a) have reported that female sex was associated with emotional problems and somatic symptoms. The same investigators also found that women and men with CHD differed in terms of coping strategies as women adopted more active problem solving style of coping than men (van Rijen et al., 2004b). The inconsistent findings may be due to the latter study differing in methodology, e.g. the regression analyses consider only medical variables, sex and age, while the other determinants which may confound the relations were neglected.

Although there was no association between age and anxiety, depressive and somatic symptoms among adults with CHD in Study II, younger patients experienced a better QoL in the domains of social relationships, physical and psychological health in Study IV. These findings suggest that the subjective assessment of QoL compared to health-related QoL may be more reflective of true well-being. Another possible explanation could be that older adults with CHD experience aging-related health conditions that may negatively influence their QoL. Afilalo and colleagues (2011) have shown that acquired medical conditions (e.g. gastrointestinal problems) play an important role in the health status of older adults with CHD. On the other hand, although old people are highly respected in the Iranian culture, there are limited social facilities, for example leisure activities, for this age group which may influence their QoL.

Parenthood was an experience reported by nearly half (43%) of the current sample (Study I), a rate higher than that found in other studies (Simko & McGinnis, 2003; van Rijen et al., 2003). It is worth noting that parenthood was associated with a problem-focused style of coping (Study III) and better QoL in the health satisfaction, psychological health and social relationship domains (Study IV). Overall, the findings seem to indicate that the experience of parenthood among adults with CHD had a protective social role, even though Horner and colleagues (2000), reported that patients with CHD may have some worries about parenthood, in particular women with complex CHD, about passing on the CHD to their children and not living long enough to raise them. On the basis of the present

findings, one could hypothesize that parenthood may be experienced as a sign of healthy condition and good functioning for adults with CHD.

Socio-economic factors encompassing education, employment status, income and profession were more or less common explanatory factors in different outcomes in this thesis. The results showed that adults with CHD had a lower educational level than non-CHD participants (Studies II, III and IV). However, higher educational levels were more common in the present CHD sample (Study I) compared with samples from, for instance, Finland and Netherlands (Nieminen et al., 2003; van Rijen et al., 2003). This could reflect cultural norms in Iran where education is highly valued as it is a very important indicator of social status. On the other hand, this could be due to a higher level of economic and social inequalities which exist in Iran compared to those European countries. Thus, patients with CHD might try to obtain higher education in order to have more chance to have control over their lives. In this regard, having a higher education provides not only a better social status per se, but also helps the individuals to get better jobs with a higher income. Hence, having lower education levels may indirectly influence the standard of living conditions such as safety, availability of resources (e.g. information, services) and financial comfort. In line with this, the present data (Study IV) show that having a low level of education and experiencing financial strain were associated with poorer QoL in the environmental health domain, and the opposite in relation to being employed. Cohen (2004) argued that having several network connections may provide various sources of information and improve health-relevant behaviours, and thus may lead to more efficient use of available health resources. This author also stated that having various social ties may protect a person from facing stressful situations; a condition which may be more likely for an employed rather than an unemployed person. Moreover, the current results revealed that adults with CHD were less often employed, and had lower job positions and annual income than those without CHD (Studies II, III and IV). The unemployment rate among the present sample of CHD patients was higher than rates among samples from other countries (Moons et al., 2004; Nieminen et al., 2003; van Rijen et al., 2003). Claessens and co-workers (2005) have reported that due to stigmatization some CHD patients may experience difficulties in their career choice, whereas McMurray et al. (2001) observed that employment problems could occur due to social misjudgement because society may emphasize CHD patients' limitations more than their capabilities. On the other hand, when there are limited jobs in a society, the physical or psychological factors become a source

of employment discrimination (Green & Tones, 2010), a condition which could be ascribed to a country with a high rate of inflation, poverty and social inequality such as Iran.

The multivariate analyses suggested that employment was a factor positively associated with a higher level of life satisfaction and a better overall QoL as well as in the physical and environmental health domains (Study IV). Low annual income, on the other hand, was associated with higher levels of somatic symptoms (Study II) and adopting emotion-focused style of coping (Study III). Interestingly, financial strain, independent of employment and income, was negatively associated with broad psychosocial outcomes in all studies of this thesis such as higher levels of anxiety, depressive and somatic symptoms (Study II), more emotion-focused style of coping (i.e. expressing frustration and anger, Study III), and poorer overall QoL as well as in the general and psychological health domains, in line with previous results (Bartley et al., 2006). These findings may indicate that CHD indirectly affects the mental and physical health as well as QoL of the patients through financial strain. One needs to consider that CHD may be an expensive disease as current data show that more than two thirds of this sample of CHD patients underwent cardiac interventions (i.e. ≈77 % and 88%, diagnostic procedures and operation, respectively), about 23% were hospitalized because of disease complications and 57% used cardiac medications (Study I). Thus, the need for medical care may result in extra expenditure and financial burdens on adults with CHD and their families. This becomes more highlighted in a country with a high rate of inflation (World Bank, 2012) and high out of pocket expenditure on health (Ibrahimipour et al., 2011), a condition which should be noted by policy makers in providing appropriate social and health care services, as well as fair insurance benefit packages.

Additionally, having CHD could be a source of worry, such as not getting a job due to cardiac defect induced disability or job discrimination, and/or not getting life/health insurance to be able to afford treatment expenses. One can speculate that the aforementioned concerns are also relevant for the present sample.

These findings emphasize the need for inter-sectorial co-operation in public health practice. In this regard, policy makers need to create conditions so that appropriate employment, compatible with the physical limitations of adults with CHD, is made available. Moreover, they should legislate labour laws to support this group of

patients (e.g. in the Netherlands there are financial benefits for employers of people with disabilities or in the United States discrimination because of the pre-existing condition is forbidden, Kamphius et al., 2002b), and monitor the implementation of these laws. Policy makers should also create conditions for the availability of vocational training that takes into consideration the health status of adults with CHD. Apart from job and vocational training, creating conditions for a secure work environment should be a major concern for policy makers.

Finally, one could argue that health care provided by professionals who are not exclusively trained for adults with CHD may result in extra costs both for the patients and the health care system. Thus investment in training the health care professionals in the care of adults with CHD could be beneficial.

6.1.5.2. Social support

Perceived social support was an important determinant factor for all outcome variables in this thesis. The findings showed that higher levels of social support were associated with better mental health (Study II), adopting more frequently problem-focused coping style (Study III), and having greater levels of life satisfaction and QoL (Study IV). This is in accord with previous studies (Chen et al., 2011; Kovacs et al., 2009; Pike et al., 2012; Rose et al., 2005). However, few studies have addressed directly the issue of social support among adults with CHD.

One explanation for the findings could be that social support itself acted as a stress-buffering component (Cohen, 2004; Ibarra-Rovillard & Kupier, 2011; Stansfeld, 2006). On the other hand, having social support provides larger social networks and consequently more available resources. These two mechanisms may help individuals to appraise their stressful life events (i.e. chronic disease, daily hassles) as less threatening and/or more controllable (Berkman et al., 2000; Cohen, 2004). Thus, a person with high levels of social support may have the perception that she/he has higher autonomy and control over her/his physical and mental health, and social functioning, a mechanism which may protect a person from allostasis overload. This perception may also lead to life satisfaction and a better QoL. Moreover, higher social support by providing larger social networks, more access to resources and material goods may empower the patients in their efforts to face and overcome problems because the patients appraise that there are possibilities to change or control the stressful situation and solve the problems

(Cohen, 2004; Jones & Bright, 2001). The data from Study III showing that higher perceived social support was associated with active problem focusing give room to this argument.

Although it has been reported that CHD patients may feel socially isolated (Horner et al., 2000; Kovacs et al., 2009), the present findings (Study III) revealed that the levels of social support were similar among adults with and without CHD. According to present results, one can hypothesize that in cultures with highly valued interpersonal relationships, for instance Iranian, social support can be used as a "natural" booster to ameliorate physical and mental health among adults with CHD. Social support is beneficial in terms of health not only for the support receiver, but also for the support provider (Post, 2005), when it is voluntary. When this is the case, reinforcing social support can improve health not only among adults with CHD, but also among their families, friends and significant others. However, reinforcing social support can act as a double-edged sword; thus one needs to be careful of social strain which may be caused by imposing excessive demands on support providers and/or invasion of privacy and meddling for the support receiver as both may have negative psychological and physiological outcomes (Green & Tones, 2010).

6.1.5.3. Anxiety, depressive and somatic symptoms

Emotional distress (i.e. anxiety and depression) was independently associated with poorer QoL and life satisfaction (Study IV) among CHD patients, similar to a previous study (Chen et al., 2011). One could hypothesize that higher levels of emotional distress may influence the appraisal of health and life conditions, and thus may negatively impact on the perceptions of QoL and life satisfaction. Additionally, emotional distress per se may interfere with social role functioning, resulting in social isolation and consequently poorer QoL. However, due to the cross-sectional design of the studies in current thesis, it was not possible to differentiate whether poor QoL and life satisfaction lead to emotional distress or vice versa. The findings of Study III showed that CHD patients scoring high on depressive symptoms adopted less frequently the problem-focused style of coping, while those with high scores in anxiety symptoms adopted both emotion- and problem- focused styles of coping. These results suggest that emotional symptoms such as irritability (anxiety) or indifference and apathy (depression) may provoke or suppress emotion-focused (e.g. expression of emotion) or problem-focused styles of coping, respectively. One could assume that emotional distress impacts on

the appraisal of the restrictions and resources that exist. Thus, distorted appraisal may influence the level of perceived control over life and adopting an effective style of coping.

The current results revealed that emotional distress was strongly associated with somatic symptoms (Study II), indicating that somatic symptoms may be the presentation of emotional distress (Sayer et al., 2003; Simon et al., 1999). Moreover, one could hypothesize that the expression of somatic symptoms may be a strategy that patients use to protect themselves from the stigmatization or discrimination related to having psychological problems found in several cultures, e.g. Iranian (Ghanean et al., 2011; Mohammadi et al., 2006; Simon et al., 1999).

Another explanation could be that patients express emotional distress somatically because they may presume that somatic symptoms are more understandable by health care providers (Mohammadi et al., 2006). The findings from Study III show that the patients with higher levels of somatic symptoms used a problem-focused style of coping more frequently and this may support the argument. Moreover, some investigators have stated that people from developing/non-western countries and/or with low-socio-economic status may have difficulties in expressing their emotional distress (Mohammadi et al., 2006; Piccinelli & Wilkinson; 2000). Thus, health care professionals working with adults with CHD, particularly in the context of developing countries/non-Western cultures, should address psychological problems.

6.1.5.4. Congenital heart disease-related factors

One frequently discussed issue is whether the severity of heart lesion or CHD-related factors are associated with psychological well-being, style of coping, life satisfaction or perceived QoL of adults with CHD. One could hypothesize that patients with more complex defects experience more emotional distress, have poorer QoL, are less satisfied with their lives and have more difficulties in coping. The present results showed that although the type of the CHD was not directly associated with any of the outcome variables in this thesis, the positive history of cardiac surgery and hospitalization due to cardiac complications were negatively associated with the physical, environmental and social domains of QoL (Study IV). The patients' positive history of surgical interventions could thus reflect that their cardiac lesions were more severe.

In any case, the results indicate that the history of undergoing angiography was positively associated with life satisfaction. On the other hand, the history of hospitalization due to cardiac complications may be truly the manifestation of more severe conditions, which interfere with physical activities and social life. The present findings were in line with some previous studies reporting that experiencing physical restrictions may impede CHD patients from physical and social roles functioning (Claessens et al., 2005; Horner et al., 2000; McMurray et al., 2001). McMurray and colleagues (2001) also reported that some CHD patients lived with the fear of undergoing surgical interventions. The aforementioned associations could be also explained in terms of the experienced uncertainties due to the lack of information. Indeed, about half of the CHD patients in the present sample could neither name their cardiac defects nor describe them (Study I). Thus, patients and their families should be carefully informed about, for instance, the CHD lesion, natural course of the disease and potential complications.

6.2. Methodological and other considerations

The strengths of the present thesis lie in its relatively large sample size, high response rate, the presence of randomly selected age- and sex-matched participants without CHD serving as comparison group, applying well-known and widely used instruments, and testing the association between a large numbers of factors with different outcome variables. To the best of my knowledge, the studies in this thesis are the first to address the psychosocial situation of adults with CHD within the context of a developing country.

However, this thesis has several limitations. The sample was selected from two university affiliated heart hospitals in Tehran. Thus, this patient population may not represent the entire population of adults with CHD in Iran. In urban areas, the private sector plays an important role in providing medical care and rehabilitation, which was not addressed in this thesis. This may result in selection bias, especially when the socio-economic status is related to health outcome and the purchased health care services. Moreover, it was not possible to contact nearly 480 patients meeting the primary inclusion criteria because of the difficulty in locating them, which may have influenced the sample size as well as contributing to selection bias. The possible explanations for the unavailability of these patients could be: the extensive phone number changes that have occurred in Tehran during the past years without the provision of facilities to track the new numbers; the altered socio-economic situation of the patients/the families which obliged them to move

to a wealthier or poorer residency area; or the worsening of the economy of the family which could no longer afford a telephone.

Nearly all types of CHD have been included in this thesis, but still one needs to acknowledge that some selection bias may have occurred as the CHD patients with cognitive disabilities were excluded. With respect to collecting the data through self-reporting questionnaires, sufficient cognitive ability was required. However, the number of adults with CHD with cognitive deficiencies was very low in the original sample.

The participants completed a self-report questionnaire, and thus the data obtained were based on the subjective perception of participants, except for the type and the number of cardiac lesions, which were obtained from hospital records. Objective measurements to corroborate the subjective responses were not performed. Similarly, no interviews were performed with the participants using, for instance, the Diagnostic and Statistical Manual of Mental Disorders-Forth Edition (DSM-IV), to confirm objectively the presence of anxiety and depressive symptoms. Thus, a general and/or differential misclassification may have occurred.

The applied measurement scales (e.g. HADS) are well established and widely used. In this thesis, all the applied instruments were translated into Persian and were adapted for the conditions in Iran (i.e. Giessen Complaint List, The Satisfaction With Life Scale, Utrecht Coping List-short form, The Multidimensional Perceived Social Support Scale). Complete psychometric studies were not conducted, however, except for The Hospital and Anxiety Scale and The World Health Organization Quality of Life Short-Form which have been shown by others to have good reliability and validity in the Iranian context (Montazeri et al., 2003; Yousefy et al., 2010).

In the present research, a cross-sectional design approach was used; therefore it was not possible to establish firm causal links.

There was no control group with other chronic diseases. Thus, it was not possible to scrutinize whether the current psychosocial situation of adults with CHD is exclusive for this group of patients or is similar to that of patients with other chronic diseases. However, selecting a control group with another chronic disease

is a serious challenge as it is rare for any other chronic disease to have the same pattern at onset, course and outcome as CHD.

The heterogeneity of CHD makes it difficult to classify patients based on the severity of the disease. The disease severity classification in this thesis has been based on initial diagnosis and just anatomy. A modified version of hierarchical classification of cardiac lesions, reported by Marellie et al. (2007), was used.

In the assessment of coping styles among adults with CHD, the participants were asked how much they "usually" use each option (of the instrument) with the assumption that the answers may provide and indicate coping strategies that people use in certain situations. But there is likely a difference between people's perception of coping and how they think they act with how they really behave in particular situations. Despite this limitation, for practical reasons, many researchers use questionnaires asking people how they "usually" cope (Jones & Bright, 2001).

6.3. Conclusions

Overall, most medical care of CHD patients was provided by cardiologists and most of the patients visited health care providers regularly, but only half of them had knowledge about their cardiac disease (i.e. type of cardiac lesion) with no differences among women and men. Adults with CHD experienced more socio-economic problems (e.g. unemployment) than non-CHD peers. However, they had similar health behaviour.

Adults with CHD experienced greater anxiety and somatic symptoms than non-CHD persons. They perceived the same level of social support from family, friends and significant others, and they also adopted different styles of coping as frequent as non-CHD peers. Nonetheless, CHD patients had a poorer QoL and experienced less satisfaction with their lives. Only somatic symptoms, palliative reaction pattern style of coping, poor life satisfaction and general QoL were independently associated with the presence of CHD.

Interestingly, there was no independent association among any of the CHD-related factors (e.g. type of cardiac lesion) and mental health condition (e.g. anxiety), somatic symptoms, styles of coping, life satisfaction and QoL, except for the

positive history of hospitalization and cardiac operation which were associated with poorer QoL.

Anxiety, depressive and somatic symptoms were predicted by financial problems, social support, and concomitant somatic symptoms/mental health, respectively. In addition to these predictors, sex and parenthood had a role in how frequently a certain style of coping is adopted, whereas, age, sex, socio-economic factors (e.g. employment), emotional distress, and perceived social support were important determinants of life satisfaction and QoL.

The results of this thesis confirm the necessity of biopsychosocial approach in caring of adults with CHD.

6.4. Implications for interventions

In spite of the improvements in medical care of adults with CHD, patients have poor QoL, are not satisfied with their lives, and suffer from anxiety and somatic symptoms. Besides improving patients' survival rates, helping them to integrate the disease with their daily lives should be a main purpose for health care professionals and policy makers. This will not be achieved unless the different entities within society cooperate with the health care sector.

One of the practical difficulties in conducting these studies was locating the CHD patients. It was not possible to access 480 patients even after several attempts, because there was no regist of CHD patients. The question is when it is not possible to access the patients, how health care providers can follow-up the patients to assess the quality of care. Therefore, establishment of a databank for CHD patients would be beneficial, especially in hospitals which receive the patients from the entire country. This may facilitate future research as well as the quality of health care provided.

Adult CHD has its own medical characteristics, which need professionals trained exclusively for this condition. The data indicate that although the majority of the CHD patients received regular medical care from different types of health professionals, they still suffered from somatic symptoms. Interestingly, somatic symptoms were independently associated with the presence of CHD and were a factor associated with nearly all outcome variables in this thesis as well as a strong association with emotional distress. Thus, the findings emphasize the importance

of establishing specialized centres for the medical care of adults with CHD, and training experts including physicians, nurses, physiotherapists, and psychologists to provide the most appropriate care for this group of patients.

Moreover, the data indicate that a relatively large number of adults with CHD experience anxiety symptoms. The suggested interventions to address this issue include:

- A comprehensive psychological assessment should be performed for each CHD patient. In addition, the establishment of specific clinics or units is needed to provide mental health consultations and services.

- Educational packages including information about the disease itself, disease course, and potential complications, health-enhancing behaviours (e.g. physical activity) should be developed and distributed among the patients and their families (e.g. written materials and/or computer programs adapted to personal capabilities and facilities).

- Social skills education, anxiety management, cognitive behaviour therapy and mindfulness interventions could be offered to patients through individual and group meetings, written materials and/or computer programs adapted to personal capabilities and facilities.

The data show that socio-economic factors are important determinants of various outcome variables in this thesis. With respect to improving the psychosocial well-being of adults with CHD, in addition to health care providers, policy makers in different levels such as government, state, community should be involved. For instance, providing job opportunities and vocational training, legislating labour laws to protect adults with CHD in the labour market, helping with unemployment and sick insurance benefits, can improve the QoL and life satisfaction of adults with CHD. Additionally, one could provide educational packages for societal organizations (e.g. job centres) and the public at large (e.g. by involving mass media) to decrease the likelihood of discrimination and stigmatization. Therefore, one needs to emphasize the necessary contribution of health care professionals in counselling.

The data indicate that social support is a factor associated with good outcomes for different outcome variables in this thesis. Meanwhile, providing and strengthening

social networks could be useful. However, this requires large-scale interventions focusing on families, communities, schools and workplaces. One strategy could be to develop support groups at different levels. Support groups are quite common in developed countries, but this phenomenon is not common in the Iranian context. Thus, patient support groups could help members discover that they are not alone in what they experience and provide a forum for disseminating information based on their own experiences. Additionally, this type of group can help patients to enhance the sense of self-worth and of being important to others, feelings that may give more purposes to their lives (Thoits, 2011). Throit (2011) has argued that both close (e.g. friends) and formal (e.g. health care providers) relationships may be helpful in facing stressful life events. Those with close relationships can provide emotional support more effectively while the members of formal relationships can supply the most beneficial informational support. Support groups in communities may help to reduce stigmatization or discrimination. Support groups in workplaces may lower the level of job discrimination and insecurity.

6.5. Future directions

The current thesis provides insight into the psychosocial situation of adults with CHD in the context of a developing country. However, further research is warranted to overcome the limitations of current studies, to assess the efficiency of suggested interventions and to address other issues not covered in this thesis.

This research had a cross-sectional design which impedes establishing firm causal links, thus future studies using a longitudinal approach are warranted.

The current sample was from an urban area, studies from both rural and urban areas are needed to investigate the impact of urbanization on the access to health care services and the experienced social inequalities and health.

Demographic changes, aging and its consequent needs should be taken into account in future research. It is reported that, in older adults with CHD, acquired medical conditions (e.g. chronic kidney disease, gastrointestinal bleeding) play an important role in their health status and mortality (Afilalo et al., 2011), while in younger patients, health outcome is mostly related to cardiac anomaly complications (e.g. pulmonary hypertension, arrhythmia). These issues need to be considered by health care providers, policy makers and researchers. Accordingly,

providing better health and social services will help adults with CHD have a healthier and happier old age.

Herewith, only anxiety and depressive symptoms were addressed, thus it is essential to conduct studies that consider a broader spectrum of potential psychological challenges.

As the concepts of coping and quality of life are subjective, complex and context bound, qualitative research may help to deepen our understanding.

The adults with CHD perceived social support as high as non-CHD persons, but it was unclear under what circumstances. There is a lack of data regarding the psychological situation of families of adults with CHD, for instance, spouses, siblings, and children. Thus, there is a need for future studies addressing the psychological situation of families of adults with CHD.

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8. REFERENCES

Adams, G., & Plaut, V. C. (2003). The cultural grounding of personal relationship:Friendship in North American and West African Worlds. *Personal Relationships*. 10: 333-347.

Adler, N. E., Boyce, T., Chesney, M. A., Cohen, S., Folkman, S., Kahn, R. L., Syme, S. L. (1994). Socioeconomic status and health. The challenge of the gradient. *American Psychology*. 49(1): 15-24.

Afilalo, J., Therrien, J., Pilote, L., Ionescu-Ittu, R., Martucci, G., Marelli, A. J. (2011). Geriatric congenital heart disease: burden of disease and predictors of mortality. *Journal of American College of Cardiology*. 58(14): 1509-1515.

Albrecht, G. L., & Devlieger, P. J. (1999). The disability paradox: high quality of life against all odds. *Social Science & Medicine*. 48(8): 977-988.

Antonovsky, A. (1987). *Unraveling the Mystery of Health: How People Manage Stress and stay well* (1st ed.). San Francisco, California: Jossy-Bass.

Bartley, M., Ferrie, J., Montgomery, S. M. (2006). Health and labour market disadvantage: unemployment, non-employment, and job insecurity. In: Marmot, M. & Wilkinson, R. G. (Eds). *Social determinants of Health* (2nd ed.). New York: Oxford University Press**.** pp.78-96.

Baumgartner, H., Bonhoeffer, P., De Groot, N. M.,de Haan, F., Deanfield, J. E., Galie, N., Gatzoulis, M. A., Gohlke-Baerwolf, C., Kaemmerer, H., Kilner, P., Meijboom, F., Mulder, B. J., Oechslin, E., Oliver, J. M., Serraf, A., Szatmari, A., Thaulow, E., Vouhe, P. R., Walma, E. (2010). ESC Guidelines for the management of grown-up congenital heart disease (new version 2010). *European Heart Journal*. 31(23): 2915-2957.

Bedard, E., Shore, D. F., Gatzoulis, M. A. (2008). Adult congenital heart disease: a 2008 overview. *British Medical Bulltin*. 85(1): 151-180.

Berkman, L. F., Glass, T., Brissette, I., Seeman, T. E. (2000). From social integration to health: Durkheim in the new millennium. *Social Science & Medicine*. 51(6): 843-857.

Bradley, C. (2001). Importance of differentiating health status from quality of life. *Lancet*, 357(9249): 7-8.

Brähler, E. & Scheer, J. (1995). Der Gießener Beschwerdebogen (GBB). Bern, Huber.

Brandhagen, D. J., Feldt, R. H., Williams, D. E. (1991). Long-term psychologic implications of congenital heart disease: a 25-year follow-up. *Mayo Clin Proceedings*. 66(5): 474-479.

Bromberg, J. I., Beasley, P. J., D'Angelo, E. J., Landzberg, M., DeMaso, D. R. (2003). Depression and anxiety in adults with congenital heart disease: a pilot study. *Heart Lung*. 32(2): 105-110.

Chen, F., Delot, E., Perloff, J. K. (2009). Genetics, Epidemiology, and Counseling. In: Perloff, J. K., Child, J. S., Aboulhosn, J. (Eds). *Congenital Heart Disease in Adults* (3rd ed.). Philadelphia: Saunders Elsevier. pp. 221-247.

Chen, C. A., Liao, S. C., Wang, J. K., Chang, C. I., Chiu, I. S., Chen, Y. S., Lu, C. W., Lin, M. T., Chiu, H. H., Chiu, S. N., Hua, Y. C., Lue, H. C., Wu, M. H. (2011). Quality of life in adults with congenital heart disease: biopsychosocial determinants and sex-related differences. *Heart*. 97(1): 38-43.

Claessens, P., Moons, P., de Casterle, B. D., Cannaerts, N., Budts, W. Gewillig, M. (2005). What does it mean to live with a congenital heart disease? A qualitative study on the lived experiences of adult patients. *European Journal of Cardiovascular Nursing*. 4(1): 3-10.

Cohen, S. (2004). Social relationships and health. *American Psychology*. 59(8): 676-684.

Compas, B. E. (2006). Psychobiological processes of stress and coping: implications for resilience in children and adolescents-comments on the papers of Romeo & McEwen and Fisher et al. *Annals of the New York Academy Sciences*. 1094: 226-234.

Congenital Heart Disease Public Health Consortium, Fact sheet. (2012). Retrived 20 February 2013 from http://www.chphc.org/hostedsites/Pages/CHDTOOLS-CHD-Fact-Sheet.aspx.

Connolly, H. M. & Warnes, C. A. (2011). Pregnancy and contraception. In: Gatzoulis, M. A., Webb, G. D., Daubeney, P. E. F. (Eds). *Diagnosis and Management of Adult Congenital Heart Disease* (2nd ed.). Philadelphia: Elsevier sanders. pp. 158-164.

Cox, D., Lewis, G., Stuart, G., Murphy, K. (2002). A cross-sectional study of the prevalence of psychopathology in adults with congenital heart disease. *Journal of Psychosomatic Research*. 52(2): 65-68.

Deanfield, J., Thaulow, E., Warnes, C., Webb, G., Kolbel, F., Hoffman, A., Sorenson, K., Kaemmer, H., Thilen, U., Bink-Boelkens, M., Iserin, L., Daliento, L., Silove, E., Redington, A., Vouhe, P., Priori, S., Alonso, M. A., Blanc, J. J., Budaj, A., Cowie, M., Deckers, J., Fernandez Burgos, E., Lekakis, J., Lindahl, B., Mazzotta, G., Morais, J., Oto, A., Smiseth, O., Trappe, H. J., Klein, W., Blomstrom-Lundqvist, C., de Backer, G., Hradec, J., Parkhomenko, A., Presbitero, P., Torbicki, A. (2003). Management of grown up congenital heart disease. *European Heart Journal*. 24(11): 1035-1084.

Detels, R. (2009). The scope and concerns of public health. In: Detels, R., Beaglehole, R., Lansang, M. A., Gulliford, M. (Eds). *Oxford Texbook of Public Health* (5th ed.). New York: Oxford University Press. pp. 3-19

Diener, E., Emmons, R. A., Larsen, R. J., Griffin, S. (1985). The Satisfaction With Life Scale. *Journal of Personality Assessment*. 49(1): 71-75.

Edwin, F., Sereboe, L. A., Tettey, M. M., Aniteye, E. A., Kotei, D. A., Tamatey, M. M., Entsua-Mensah, K., Frimpong-Boateng, K.L. (2010). Experience from a single centre concerning the surgical spectrum and outcome of adolescents and adults with congenitally malformed hearts in West Africa. *Cardiology in the Young*. 20(2): 159-164.

Engel, G. L. (1977). The need for a new medical model: a challenge for biomedicine. *Science*. 196(4286):129-136.

Fayers, P. M., & Machin, D. (2007). Introduction. In: *Quality of life: the assessment, analysis and interpretation of patient-reported outcomes* (2nd ed.). Chichester: John Wiley & Son Ltd. pp. 3-30.

Garg, V. (2006). Insights into the genetic basis of congenital heart disease. *Cellular and Molecular Life Sciences*. 63(10): 1141-1148.

Geyer, S., Norozi, K., Zoege, M., Kempa, A., Buchhorn, R., Wessel, A. (2006). Psychological symptoms in patients after surgery for congenital cardiac disease. *Cardiology in the Young*. 16(6): 540-548.

Ghanean, H., Nojomi, M., Jacobsson, L. (2011). Internalized Stigma of Mental Illness in Tehran, Iran. *Stigma Research and Action*. 1(1): 11-17.

Gnanappa, G. K., Ganigara, M., Prabhu, A., Varma, S. K., Murmu, U., Varghese, R., Valliatu, J., Kumar, R. N. (2011). Outcome of complex adult congenital heart surgery in the developing world. *Congenit Heart Disease*. 6(1): 2-8.

Goossens, E., Van Deyk, K., Zupancic, N., Budts, W., Moons, P. (2013). Effectiveness of structured patient education on the knowledge level of adolescents and adults with congenital heart disease. *European Journal of Cardiovascular Nursing*. Published online: 17 February. Doi: 10.1177/1474515113479231.

Green, J., & Tones, K. (2010). *Assessing health and its determinants. In: Health promotion: Planning and strategies* (2nd ed.). London: Sage publications Ltd. pp. 57-110.

Hoffman, J. I., & Kaplan, S. (2002). The incidence of congenital heart disease. *Journal of American College of Cardiology*. 39(12): 1890-1900.

Holland, K. D., & Holahan, C. (2003). The Relation of Social Support and Coping to Positive Adaptation to Breast Cancer. *Psychology & Health*. 18(1): 15-29.

Horner, T., Liberthson, R., Jellinek, MS. (2000). Psychosocial profile of adults with complex congenital heart disease. *Mayo Clinic Proceeding*. 75(1): 31-36.

Ibarra-Rovillard, M. S., & Kuiper, N. A. (2011). Social support and social negativity findings in depression: perceived responsiveness to basic psychological needs. *Clinical Psychology Review*. 31(3): 342-352.

Ibrahimipour, H., Maleki, M. R., Brown, R., Gohari, M., Karimi, I., Dehnavieh, R. (2011). A qualitative study of the difficulties in reaching sustainable universal health insurance coverage in Iran. *Health Policy and Planning*. 26(6): 485-495.

Immer, F. F., Althaus, S. M., Berdat, P. A., Saner, H., Carrel, T. P. (2005). Quality of life and specific problems after cardiac surgery in adolescents and adults with congenital heart diseases. *European Journal of Cardiovascular Prevention and Rehabilitation*. 12(2): 138-143.

Jones, F., & Bright, J. (2001). *Stress: Myth, Theory and Research*. Edinburg gate: Harlow, Pearson Education Limited.

Kamphuis, M., Ottenkamp, J., Vliegen, H. W., Vogels, T., Zwinderman, K. H., Kamphuis, R. P., Verloove-Vanhorick, S. P. (2002a). Health related quality of life and health status in adult survivors with previously operated complex congenital heart disease. *Heart*. 87(4): 356-362.

Kamphuis, M., Vogels, T., Ottenkamp, J., Van Der Wall, E. E., Verloove-Vanhorick, S. P., Vliegen, H. W. (2002b). Employment in adults with congenital heart disease. *Archive of Pediatric and Adolescence Medicine*. 156(11): 1143-1148.

Karsdorp, P. A., Kindt, M., Rietveld, S., Everaerd, W., Mulder, B. J. (2008). Interpretation bias for heart sensations in congenital heart disease and its relation to quality of life. *Internation Journal of Behavioral Medicine*. 15(3): 232-240.

Kim, J., Han, J. Y., Shaw, B., McTavish, F., Gustafson, D. (2010). The roles of social support and coping strategies in predicting breast cancer patients' emotional well-being: testing mediation and moderation models. *Journal of Health Psychology*. 15(4): 543-552.

Kovacs, A. H., Saidi, A. S., Kuhl, E. A., Sears, S. F., Silversides, C., Harrison, J. L., Ong, L., Colman, J., Oechslin, E., Nolan, R. P. (2009). Depression and anxiety in adult congenital heart disease: predictors and prevalence. *International Journal of Cardiology*. 137(2): 158-164.

Kovacs, A. H., Sears, S. F., Saidi, A. S. (2005). Biopsychosocial experiences of adults with congenital heart disease: review of the literature. *Americal Heart Journal*. 150(2): 193-201.

Lane, D. A., Lip, G. Y., Millane, T. A. (2002). Quality of life in adults with congenital heart disease. *Heart*. 88(1): 71-75.

Lazarus, R. S. (1999). Coping. In: *Stress and emotion, a new synthesis*. London: Free Association Books. pp. 101-125.

Lazarus, R. S., & Folkman, S. (1984). The Coping Process: An Alternative to Traditional Formulation. In: *Stress, Appraisal, AND Coping*. New York: Springer Publishing Company. pp.141-180.

Logan, J. G., & Barksdale, D. J. (2008). Allostasis and allostatic load: expanding the discourse on stress and cardiovascular disease. *Journal of Clinical Nursing*. 17(7B): 201-208.

Loup, O., von Weissenfluh, C., Gahl, B., Schwerzmann, M., Carrel, T., Kadner, A. (2009). Quality of life of grown-up congenital heart disease patients after congenital cardiac surgery. *European Journal of Cardiothoracular Surgery*. 36(1): 105-111.

Marelli, A. (2012). The Future of ACHD Care Symposium: Changing demographics of congenital heart disease. *Progress in Pediatric Cardiology*. 34(2): 85-90.

Marelli, A. J., Mackie, A. S., Ionescu-Ittu, R., Rahme, E., Pilote, L. (2007). Congenital heart disease in the general population: changing prevalence and age distribution. *Circulation*. 115(2): 163-172.

Markus, H., & Kitayama, S. (1991). Culture and the self: Implications for cognition, emotion, and motivation. *Psychological Review*. 98(2): 224-253.

Marmot, M. (2004). *Status Syndrome:how your social standing directly affects your health and life expectancy*. London : Bloomsbury.

Mayes, B. T., Johnson, T. W., Sadri, G. (2000). Personality, job level, job stressors, and their interaction as predictors of coping behavior. *Psychological Reports*. 87(1): 61-81.

McEwen, B. S. (1998). Stress, adaptation, and disease. Allostasis and allostatic load. *Annals of the New York Academic Sciences*. 840: 33-44.

McEwen, B. S. (2004). Protection and damage from acute and chronic stress: allostasis and allostatic overload and relevance to the pathophysiology of psychiatric disorders. *Annals of the New York Academic Sciences*. 1032: 1-7.

McEwen, B. S., & Wingfield, J. C. (2003). The concept of allostasis in biology and biomedicine. *Hormones and Behavior*. 43(1): 2-15.

McMurray, R., Kendall, L., Parsons, J. M., Quirk, J., Veldtman, G. R., Lewin, R. J. P., Sloper, P. (2001). A life less ordinary: growing up and coping with congenital heart disease. *Coronary Health Care*. 5(1): 51-57.

Mehrdad, R. (2009). Health System in Iran. *Japan Medical Association Journal*. 52(1): 69-73.

Mitchell, S. C., Korones, S. B., Berendes, H. W. (1971). Congenital heart disease in 56,109 births. Incidence and natural history. *Circulation*. 43(3): 323-332.

Mocumbi, A. O., Lameira, E., Yaksh, A., Paul, L., Ferreira, M. B., Sidi, D. (2011). Challenges on the management of congenital heart disease in developing countries. *Intnational Journal of Cardiology*. 148(3): 285-288.

Moghadam, M. N., Sadeghi, V., Parva, S. (2012). Weaknesses and challenges of primary healthcare system in Iran: a review. *The International Journal of Health Planning and Management*. 27(2): e121-131.

Mohammadi, M., Ghanizadeh, A., Davidian, H., Noorbala, A. A., Malekafzali, H., Naghavi, H. R., Pouretemad, H. R., Bagheri Yazdi, S. A., Rahgozar, M., Alaghebandrad, J., Amini, H., Razzaghi, E. M., Mesgarpour, B., Mohammadi, M., Ghaemi, S. N. (2006). Prevalence of Mood Disorders in Iran. *Iranian Journal of Psychiatry*. 1: 59-64.

Mokhles, M. M., van de Woestijne, P. C., de Jong, P. L., Witsenburg, M., Roos-Hesselink, J. W., Takkenberg, J. J., Bogers, A. J. (2011). Clinical outcome and health-related quality of life after right-ventricular-outflow-tract reconstruction with an allograft conduit. *European Journal of Cardiothoracic Surgery*. 40(3): 571-578.

Montazeri, A., Vahdaninia, M., Ebrahimi, M., Jarvandi, S. (2003). The Hospital Anxiety and Depression Scale (HADS): translation and validation study of the Iranian version. *Health and Quality of Life Outcomes*. 1: 14.

Moons, P., Budts, W., De Geest, S. (2006b). Critique on the conceptualisation of quality of life: a review and evaluation of different conceptual approaches. *International Journal of Nursing Studies*. 43(7): 891-901.

Moons, P., De Bleser, L., Budts, W., Sluysmans, T., De Wolf, D., Massin, M., Gewillig, M., Pasquet, A., Suys, B., Vliers, A. (2004). Health status, functional abilities, and quality of life after the Mustard or Senning operation. *Annals of Thoracic Surgery*. 77(4): 1359-1365.

Moons, P., & Norekval, T. M. (2006). Is sense of coherence a pathway for improving the quality of life of patients who grow up with chronic diseases? A hypothesis. *European Journal of Cardiovascular Nursing*. 5(1): 16-20.

Moons, P., Van Deyk, K., De Bleser, L., Marquet, K., Raes, E., De Geest, S., Budts, W. (2006). Quality of life and health status in adults with congenital heart disease: a direct comparison with healthy counterparts. *European Journal of Cardiovascular Prevention and Rehabilitation*. 13(3): 407-413.

Moons, P., Van Deyk, K., De Geest, S., Gewillig, M., Budts, W. (2005). Is the severity of congenital heart disease associated with the quality of life and perceived health of adult patients? *Heart*. 91(9): 1193-1198.

Moons, P., Van Deyk, K., Marquet, K., De Bleser, L., De Geest, S., Budts, W. (2009). Profile of adults with congenital heart disease having a good, moderate, or poor quality of life: a cluster analytic study. *European Journal of Cardiovascular Nursing*. 8(2): 151-157.

Nieminen, H., Sairanen, H., Tikanoja, T., Leskinen, M., Ekblad, H., Galambosi, P., Jokinen, E. (2003). Long-term results of pediatric cardiac surgery in Finland: education, employment, marital status, and parenthood. *Pediatrics*. 112(6 Pt 1): 1345-1350.

Nikyar, B., Sedehi, M., Mirfazeli, A., Qorbani, M., Golalipour, M. J. (2011). Prevalence and Pattern of Congenital Heart Disease among Neonates in Gorgan, Northern Iran (2007-2008). *Iranian Journal of Pediatrics*. 21(3): 307-312.

Nobakht, M., Aarabi, M. Y., Shahmohammadi, A., Davari, P., Meraji, S. M., Tabib, A., Mortazaeiyan, H. (2008). Ten years experience with transposition of the great arteries associated with ventricular septal defect and left ventricular outflow tract obstruction (LVOTO). *Heart, Lung and Circulation*. 17S: 49.

Norozi, K., Zoege, M., Buchhorn, R., Wessel, A., Geyer, S. (2006). The Influence of Congenital Heart Disease on Psychological Conditions in Adolescents and Adults after Corrective Surgery. *Congenital Heart Diseas.* 1: 282-288.

Ong, L., Nolan, R. P., Irvine, J., Kovacs, A. H. (2011). Parental overprotection and heart-focused anxiety in adults with congenital heart disease. *International Journal of Behavioral Medicine*. 18(3): 260-267.

Park, C. L., Edmondson, D., Fenster, J. R., Blank, T. O. (2008). Positive and negative health behavior changes in cancer survivors: a stress and coping perspective. *Journal of Health and Psychology*. 13(8): 1198-1206.

Perloff, J. K., & Warnes, C. A. (2001). Challenges posed by adults with repaired congenital heart disease. *Circulation*. 103(21): 2637-2643.

Piccinelli, M., & Wilkinson, G. (2000). Gender differences in depression. Critical review. *British Journal of Psychiatry*. 177: 486-492.

Pike, N. A., Evangelista, L. S., Doering, L. V., Eastwood, J. A., Lewis, A. B., Child, J. S. (2012). Quality of life, health status, and depression: comparison between adolescents and adults after the Fontan procedure with healthy counterparts. *Journal of Cardiovascular Nursing*. 27(6): 539-546.

Popelova, J., Slavik, Z., Skovranek, J. (2001). Are cyanosed adults with congenital cardiac malformations depressed? *Cardiology in the Young*. 11(4): 379-384.

Post, S. G. (2005). Altuism, happiness, and health: it's good to be good. *International Journal of Behavioral Medicine*. 12(2): 66-77.

Rahim, F., Ebadi, A., Saki, G., Remazani, A. (2008). Prevalence of congenital Heart Disease in Iran: A Clinical Study. *Journal of Medical Sciences*. 8(6): 547-552.

Rietveld, S., Mulder, B. J., van Beest, I., Lubbers, W., Prins, P. J., Vioen, S., Bennebroek-Evererz, F., Vos, A., Casteelen, G., Karsdorp, P. (2002). Negative thoughts in adults with congenital heart disease. *International Journal of Cardiology*. 86(1): 19-26.

Rolland, J. S. (1987). Chronic illness and the life cycle: a conceptual framework. *Family Process*. 26(2): 203-221.

Romano-Zelekha, O., Hirsh, R., Blieden, L., Green, M., Shohat, T. (2001). The risk for congenital heart defects in offspring of individuals with congenital heart defects. *Clinical Genetics*. 59(5): 325-329.

Rose, M., Kohler, K., Kohler, F., Sawitzky, B., Fliege, H., Klapp, B. F. (2005). Determinants of the quality of life of patients with congenital heart disease. *Quality of Life Research*. 14(1): 35-43.

Rönning, H., Nielsen, N. E., Swahn, E., Strömberg, A. (2008). Educational needs in adults with congenitally malformed hearts. *Cardiology in the Young*. 18(5): 473-479.

Saliba, Z., Butera, G., Bonnet, D., Bonhoeffer, P., Villain, E., Kachaner, J., Sidi, D., Iserin, L. (2001). Quality of life and perceived health status in surviving adults with univentricular heart. *Heart*. 86(1): 69-73.

Sayar, K., Kirmayer, L. J., Taillefer, S. S. (2003). Predictors of somatic symptoms in depressive disorder. *General Hospital Psychiatry*. 25(2): 108-114.

Shaw, M., Dorling, D., Smith, G. D. (2006). Poverty, social exclusion, and minorities. In: Marmot, M. & Wilkinson, R. G. (Eds). *Social determinants of Health* (2nd ed.). New York: Oxford University Press. pp.196-223.

Schreurs, P.J.G., Van de Willige, G., Tellegen, B. (1988). *Utrecht Coping List: a manual Lisse*. Swets en Zeitlinger.

Shiina, Y., Toyoda, T., Kawasoe, Y., Tateno, S., Shirai, T., Wakisaka, Y., Matsuo, K., Mizuno, Y., Terai, M., Hamada, H., Niwa, K. (2011). Prevalence of adult patients with congenital heart disease in Japan. *International Journal of Cardiology*. 146(1): 13-16.

Silva, A. M., Vaz, C., Areias, M. E., Vieira, D., Proenca, C., Viana, V., Moura, C., Areias, J. C. (2011). Quality of life of patients with congenital heart diseases. *Cardiology in the Young*. 21(6): 670-676.

Simko, L. C., & McGinnis, K. A. (2003). Quality of life experienced by adults with congenital heart disease. *American Association of Critical-Care Nurses Clinical Issues*. 14(1): 42-53.

Simko, L. C., McGinnis, K. A., Schembri, J. (2006). Educational needs of adults with congenital heart disease. *Journal of Cardiovascular Nursing*. 21(2): 85-94.

Simon, G. E., VonKorff, M., Piccinelli, M., Fullerton, C., Ormel, J. (1999). An international study of the relation between somatic symptoms and depression. *New England Journal of Medicine*. 341(18): 1329-1335.

Skevington, S. M., Lotfy, M., O'Connell, K. A. (2004). The World Health Organization's WHOQOL-BREF quality of life assessment: psychometric properties and results of the international field trial. A report from the WHOQOL group. *Quality of Life Research*. 13(2): 299-310.

Stansfeld, S. A. (2006). Social support and social cohesion Social determinants of Health. In: Marmot, M. & Wilkinson, R. G. (Eds). *Social determinants of Health* (2nd ed.). New York: Oxford University Press. pp.148-171.

Thoits, P. A. (1986). Social support as coping assistance. *Journal of Consulting and Clinical Psychology*. 54(4): 416-423.

Thoits, P. A. (2011). Mechanisms linking social ties and support to physical and mental health. *Journal of Health and Social Behavior*. 52(2): 145-161.

Uchino, B. N., Bowen, K., Carlisle, M., Birmingham, W. (2012). Psychological pathways linking social support to health outcomes: a visit with the "ghosts" of research past, present, and future. *Social Science & Medicine*. 74(7): 949-957.

UNFPA. *State of the World Population 2009*. Retrived 5 March 2013 from http://foweb.unfpa.org/SWP2011/reports/EN-SWOP2011-FINAL.pdf.

Utens, E. M., Bieman, H. J., Verhulst, F. C., Meijboom, F. J., Erdman, R. A., Hess, J. (1998). Psychopathology in young adults with congenital heart disease. Follow-up results. *European Heart Journal*. 19(4): 647-651.

Utens, E. M., Verhulst, F. C., Erdman, R. A., Meijboom, F. J., Duivenvoorden, H. J., Bos, E., Roelandt, J. R., Hess, J. (1994). Psychosocial functioning of young adults after surgical correction for congenital heart disease in childhood: a follow-up study. *Journal of Psychosomatic Research*. 38(7): 745-758.

van Rijen, E. H., Utens, E. M., Roos-Hesselink, J. W., Meijboom, F. J., van Domburg, R. T., Roelandt, J. R., Bogers, A. J., Verhulst, F. C. (2003). Psychosocial functioning of the adult with congenital heart disease: a 20-33 years follow-up. *European Heart Journal*. 24(7): 673-683.

van Rijen, E. H., Utens, E. M., Roos-Hesselink, J. W., Meijboom, F. J., van Domburg, R. T., Roelandt, J. R., Bogers, A. J., Verhulst, F. C. (2004a). Medical predictors for psychopathology in adults with operated congenital heart disease. *European Heart Journal*. 25(18): 1605-1613.

van Rijen, E. H., Utens, E. M., Roos-Hesselink, J. W., Meijboom, F. J., van Domburg, R. T., Roelandt, J. R., Bogers, A. J., Verhulst, F. C. (2004b). Styles of coping and social support in a cohort of adults with congenital heart disease. *Cardiology in the Young*. 14(2): 122-130.

van Rijen, E. H., Utens, E. M., Roos-Hesselink, J. W., Meijboom, F. J., van Domburg, R. T., Roelandt, J. R., Bogers, A. J., Verhulst, F. C. (2005). Longitudinal development of psychopathology in an adult congenital heart disease cohort. *International Journal of Cardiology*. 99(2): 315-323.

Vida, V. L., Barnoya, J., O'Connell, M., Leon-Wyss, J., Larrazabal, L. A., Castaneda, A. R. (2006). Surgical versus percutaneous occlusion of ostium secundum atrial septal defects: results and cost-effective considerations in a low-income country. *Journal of American College of Cardiology*. 47(2): 326-331.

Vigl, M., Niggemeyer, E., Hager, A., Schwedler, G., Kropf, S., Bauer, U. (2011). The importance of socio-demographic factors for the quality of life of adults with congenital heart disease. *Quality of Life Research*. 20(2): 169-177.

Warnes, C. A. (2005). The adult with congenital heart disease: born to be bad? *Journal of American College of Cardiology*. 46(1): 1-8.

Warnes, C. A., Liberthson, R., Danielson, G. K., Dore, A., Harris, L., Hoffman, J. I., Somerville, J., Williams, R. G., Webb, G. D. (2001). Task force 1: the changing profile of congenital heart disease in adult life. *Journal of American College of Cardiology*. 37(5): 1170-1175.

Warnes, C. A., Williams, R. G., Bashore, T. M., Child, J. S., Connolly, H. M., Dearani, J. A., del Nido, P., Fasules, J. W., Graham, T. P., Jr., Hijazi, Z. M., Hunt, S. A., King, M. E., Landzberg, M. J., Miner, P. D., Radford, M. J., Walsh, E. P., Webb, G. D., Smith, S. C., Jr., Jacobs, A. K., Adams, C. D., Anderson, J. L., Antman, E. M., Buller, C. E., Creager, M. A., Ettinger, S. M., Halperin, J. L., Krumholz, H. M., Kushner, F. G., Lytle, B. W., Nishimura, R. A., Page, R. L., Riegel, B., Tarkington, L. G., Yancy, C. W. (2008). ACC/AHA 2008 guidelines for the management of adults with congenital heart disease: a report of the American College of Cardiology/American Heart Association Task Force on Practice Guidelines (Writing Committee to Develop Guidelines on the Management of Adults With Congenital Heart Disease). Developed in Collaboration With the American Society of Echocardiography, Heart Rhythm Society, International Society for Adult Congenital Heart Disease, Society for Cardiovascular Angiography and Interventions, and Society of Thoracic Surgeons. *Journal of American College of Cardiology*. 52(23): e143-263.

World Bank. (2008). Islamic Republic of Iran Health Sector Review, Volume I: Main Report. Retrived 10 March 2013 from https://openknowledge.worldbank.org/handle/10986/7960.

World Bank Group. (2012). World Development Indicators 2012. Retrived 5 March 2013 from http://data.worldbank.org/sites/default/files/wdi-2012-ebook.pdf.

World Health Organisation statistics. Retrived 31 October 2012 from http://www.who.int/countries/irn/en/.

World Medical Association. (2000). Declaration of Helsinki. *The Journal of the American Medical Association*. 248(23): 3043-3045.

Yousefy, A. R., Ghassemi, G. R., Sarrafzadegan, N., Mallik, S., Baghaei, A. M., Rabiei, K. (2010). Psychometric properties of the WHOQOL-BREF in an Iranian adult sample. *Community Mental Health Journal*. 46(2): 139-147.

Zigmond, A. S., & Snaith, R. P. (1983). The hospital anxiety and depression scale. *Acta Psychiatrica Scandinavica*. 67(6): 361-370.

Zimet, G. D., Dahlem, N. W., Zimet, S. G., Farley, G. K. (1988). The Multidimensional Scale of Perceived Social Support. *Journal of Personality Assessment*. 52: 30-41.

APPENDIX

Table 2. Demographic, socio-economic, and health behaviour description of adult with and without CHD.

Variables	CHD	Non-CHD	*p*-value
	n (%)	n (%)	
Age (mean±SD)	33.24 ±12.11	33.49 ± 12.18	NS
Female	181 (52.2)	182 (51.6)	NS
Marital status			NS
Never married	148 (42.7)	125 (35.4)	
Married	185 (53.3)	214 (60.6)	
Divorced/widowed	14 (4)	14 (4)	
Having children	150 (43.2)	181 (51.3)	0.03
Educational level			0.000
High	84 (24.2)	164 (46.5)	
Intermediate	153 (44.1)	126 (35.7)	
Low	110 (31.7)	63 (17.8)	
Unemployed	216 (62.2)	179 (50.7)	0.002
On sick-leave [a]	43 (12.4)	35 (9.9)	0.01
Annual income			0.000
Not disclosed	106 (30.5)	113 (32)	
High	11 (3.2)	67 (19)	
Middle	69 (19.9)	36 (10.2)	
Low	161 (46.4)	29 (8.2)	
Profession [b]			0.002
Blue-collar worker	70 (20.2)	69 (9.5)	
Low white-collar worker	21 (6.1)	33 (9.3)	
Middle/high white-collar worker	40 (11.5)	72 (20.4)	
Financial strain			0.003
Never	70 (20.2)	62 (17.6)	
Quite often	106 (30.5)	131 (37.2)	
Often	127 (36.6)	81 (23)	
Always	44 (12.7)	79 (22.4)	
BMI (Mean ±SD)	23.98 ± 4.61	25.27 ± 4.11	0.000
Cigarette use	43 (12.4)	52 (14.7)	NS
Alcohol use	37 (10.7)	35 (9.9)	NS
Water-pipe use	69 (19.9)	105 (29.7)	0.003
Opium use	10 (3)	4 (1.1)	NS
Dieting			NS
Fruit/vegetables	321 (92.5)	333 (94.3)	
Meat/fish	292 (84.1)	307 (87)	
Dairy	318 (91.6)	332 (94.1)	
Grains	342 (98.6)	342 (96.9)	
Physical activity	106 (30.5)	125 (35.4)	NS

[a] = being on sick-leave during the past 12 months, [b] = only employed participants replied this question. CHD indicates congenital heart disease; SD, standard deviation; NS, non significant; BMI, body mass index.

Table 3. Overall summary of results of differences between adults with and without CHD

Variables	CHD		Non-CHD		*p*-value
	Mean (SD)	***Median***	***Mean (SD)***	***Median***	
Anxiety	9.22(4.40)		8.11 (4.22)		0.001
Depression	6.51 (4.30)		6.36 (3.98)		NS
Somatic symptoms					
Total		14		8	0.000
Exhaustion tendency		4		2	0.000
Gastrointestinal discomfort		1		1	0.014
Musculoskeletal discomfort		4		3	0.002
Heart distress		3		0	0.000
Coping strategies					
Active Problem Focusing	12.88 (2.67)		13.20 (2.50)		NS
Palliative Reaction Pattern	8.31 (2.03)		8.72 (1.93)		0.007
Avoidance Behaviour	6.37 (1.78)		6.13 (1.58)		NS
Seeking Social Support	11.39 (3.08)		11.56 (3.00)		NS
Expression of Emotions	4.21 (1.51)		4.08 (1.31)		NS
Social support					
Total score	58.31 (15.0)		57.80 (14.87)		NS
Family	21.69 (5.96)		21.38 (5.71)		NS
Friends	15.76 (7.16)		15.90 (6.88)		NS
Significant Other	20.86 (5.98)		20.53 (6.27)		NS
Quality of life					
Overall QoL	3.36 (0.82)		3.57 (0.78)		0.000
Satisfaction with health	3.31 (0.93)		3.71 (0.86)		0.000
Physical health	59.65 (17.69)		66.19 (15.39)		0.000
Psychological health	56.33 (18.72)		60.34 (16.82)		0.003
Social relationships	56.78 (19.28)		58.60 (18.32)		NS
Environment	50.95 (15.86)		53.50 (15.72)		0.033
Life Satisfaction	19.48 (7.80)		21.33 (7.60)		0.002

CHD indicates congenital heart disease; NS, non significant; SD, standard deviation; QoL, quality of life.

Table 4. Overall summary of factors associated with anxiety, depression, somatic symptoms, styles of coping, quality of life and life satisfaction among adults with CHD

		Study II			Study III		Study IV							
	Dependent variables →	*Anxiety*	*Depression*	*Somatic symptoms*	*Coping*		*Quality of Life*							*Life satisfaction*
	Independent variables ↓				Problem focused	Emotion focused	Overall	Health satisfaction	Physical	Psychological	Social relationship	Environmental	
Demographics	Sex (female)						+				+	+	+	
	Age								–	–	–			
	Being married				–									
	Having children				+			+		+	+			
Socio-economics	Education											+		
	Annual income			–		– †								
	Financial strain	+	+	+		+	–	–		–		–		
	Being employed				–		+		+			+	+	
	Profession												–	
Health & social support	Anxiety			↑	↑	↑	↓	↓	↓	↓	↓	↓	↓	
	Depression			↑	↓		↓	↓	↓	↓	↓	↓	↓	
	Somatic symptoms	↑	↑		↑			↓	↓			↓		
	Social support	↓	↓		↑		↑			↑	↑	↑	↑	
CHD-related variables	Defect severity													
	Time of diagnosis													
	History of hospitalization & cardiac intervention								–		–	–	+	
	Medication use													

† Emotion focused style of coping was associated with extreme level of annual income as low and high annual income; CHD indicates congenital heart disease.