Introduction

Health anxiety refers to anxiety or concerns regarding bodily symptoms and the belief that these symptoms may be indicative of a serious illness (American Psychiatric Association (APA), 2013; Asmundson et al., 2010). Health anxiety is thought to exist on a continuum ranging from lack of concern about one’s health to severe preoccupation (Asmundson et al., 2010). When health anxiety becomes excessive and impacts day-to-day functioning, it may warrant a diagnosis. Current diagnostic classifications of clinical levels of health anxiety are housed within the somatic symptom and related disorders section of Diagnostic and Statistical Manual of Mental Disorders (5th ed.; DSM-5; APA, 2013).

Examination of the phenomena of health in children has grown considerably within the literature in recent years (e.g. Delparte et al.,...
While we have observed growth in the area, we still have a limited understanding of the phenomena, in particular how and if health anxiety is experienced in specialized child or adolescent physical health populations. Health anxiety has been explored and observed in adults with various physical health conditions, such as chronic pain (Rode et al., 2006), fibromyalgia (Uçar et al., 2015), Ménière’s disease (Kirby and Yardley, 2009), multiple sclerosis (Kehler and Hadjistavropoulos, 2009), and non-cardiac chest pain (Bozkurt Zincir et al., 2014) demonstrating significantly higher levels of health anxiety compared to controls.

While there is no research specifically exploring health anxiety in a child or adolescent physical health population, Rask et al. (2016) was able to examine the association between self-reported health anxiety and physical health conditions in children 11–12 years of age, in the context of a larger study. Rask and colleagues found no relationship between health anxiety and presence of chronic physical conditions. However, their overall sample was categorized as either having a chronic physical health condition or not, with the most common chronic physical condition being asthma (i.e. 12.9%), without a priori participant recruitment or selection of participants with specific chronic physical health conditions. It is unknown if children and adolescents with specific chronic physical health conditions that may have more significant impact on day-to-day functioning (e.g. diabetes, childhood arthritis, and congenital heart disease) may demonstrate similar patterns of health anxiety observed in some adult chronic physical health populations.

Congenital heart disease (CHD) refers to a diverse group of diseases characterized by having a structural heart defect or abnormality present at birth (Karsdorp et al., 2007). The overall prevalence of CHD is increasing due to improved survival rates, with CHD prevalence recently having been shown to have increased by 11 percent in children and 57 percent in adults from 2000 to 2010 (i.e. 2000–2010; Marelli et al., 2014). Most children with significant structural heart defects require surgery and, in some cases, may require a heart transplantation (Warnes et al., 2001). Therefore, CHD represents a growing clinical population with significant health care exposure in early life that may precipitate disruptions in psychological functioning. However, our knowledge about the psychological functioning of this specialized population is limited and, at times, contradictory. For instance, there are mixed findings as to whether adolescents with CHD are at greater risk of developing internalizing disorders (e.g. anxiety and social withdrawal) and to a lesser extent, externalizing disorders (e.g. aggression and hyperactivity) than healthy children (e.g. Karsdorp et al., 2007; Rassart et al., 2014).

This cohort study (i.e. a study designed to follow a group of individuals with the same disease phenomenon; Sessler and Imrey, 2015) sought to quantitatively explore health anxiety and associated constructs (i.e. intolerance of uncertainty, anxiety sensitivity, and Diagnostic and Statistical Manual of Mental Disorders (4th ed.; DSM-IV) anxiety symptom categories) in children and adolescents with CHD. We hypothesized that children and adolescents with CHD would demonstrate significantly higher levels of health anxiety and associated constructs than typically developing children. We also hypothesized that significant associations would be observed between health anxiety and the associated constructs.

**Methods**

**Participants and recruitment procedure**

A total of 84 children and adolescents ages 7–16 years (42 children with CHD; mean age=11.12, standard deviation (SD)=2.72, and 42 typically developing children; mean age=12.19, SD=2.24) participated in this study. The majority of participants self-identified as Caucasian (i.e. CHD group=85.7%, n=36; typically developing group=97.6%, n=41).
Participants with CHD were recruited from the Department of Cardiology at the Royal University Hospital in Saskatoon, Saskatchewan, Canada and typically developing participants were recruited from the local community. Approximately 80 percent of parents of children or adolescents with CHD contacted provided consent for their child to participate in the study. Declining study participation was due to scheduling conflicts and distance from site of study. Approximately 75 percent of parents of typically developing children contacted provided consent for their child to participate. Declined participation was related to lack of interest.

**Exclusion criteria**

The exclusion criteria for this study were established by the criteria from a larger study (i.e. study associated with the Children’s Healthy Heart Activity Monitoring Program in Saskatchewan (CHAMPS)), that had an intervention component necessitating the following exclusion criteria: (1) cardiac surgery within the last 6 months, (2) persisting cyanosis, and (3) intellectual disability. A pediatric cardiologist pre-screened all participants with CHD to ensure criteria were met. Typically developing children were recruited from the local community through advertisements and word of mouth requesting healthy participants.

**Measures**

**Demographic form.** Parent/legal guardian completed a basic demographic form for their child. Items included questions regarding age, sex, date of birth, ethnicity, and location of residence. Participants’ medical history and diagnoses were also obtained from parents.

**Childhood Illness Attitude Scales.** The Childhood Illness Attitude Scales (CIAS; Wright and Asmundson, 2003) is 35-item self-report measure used to evaluate fears, beliefs, and attitudes associated with health anxiety in school-aged children (Wright and Asmundson, 2005; for example, Item 1—Do you worry about your health?; Item 2—Are you worried that you might get really sick in the future?). Most items are rated on a 3-point Likert-type scale, ranging from 1 (none of the time) to 3 (a lot of the time). Items 29 through 31 evaluate the frequency of treatment experiences and are rated on a 3-point scale, ranging from 1 (zero times) to 3 (three or more times), and items 28 and 32 are not included, as they are open-ended questions used to gain supplementary information (Wright and Asmundson, 2003). It is recommended to exclude items 8, 11, 15, and 25 when scoring as these items either cross-loaded on multiple factors or did not load significantly on any particular factor (Wright and Asmundson, 2005). Thus, total scores range from 29 to 87, with higher scores indicating higher levels of health anxiety and illness behavior. The CIAS comprises four subscales including fears, help-seeking, symptom effects, and treatment experiences (Wright and Asmundson, 2003). The CIAS has demonstrated strong construct validity with associated measures (i.e. $r=.71$; Delparte et al., 2015), high re-test reliability after a 10- to 14-day interval (i.e. $r=.86$; Wright and Asmundson, 2003), and excellent internal consistency in terms of its total score (i.e. $\alpha=.88–.89$; Wright and Asmundson, 2005 and $\alpha=.80$; Rask et al., 2016). For this study, the CIAS total score demonstrated good internal consistency ($\alpha=.84$) and acceptable to good internal consistency for the CIAS subscale scores: $\alpha=.82$ (fears) and $\alpha=.84$ (treatment experiences). However, the help-seeking and symptom effects subscales were just below the acceptable value ($\alpha=.68$ and $\alpha=.69$, respectively).

**Childhood Anxiety Sensitivity Index.** The Childhood Anxiety Sensitivity Index (CASI; Silverman et al., 1991) is an 18-item child self-report measure. Each item (e.g. Item 3—It scares me when I feel “shaky”; Item 5. It is important for me to stay in control of my feelings) is rated on a 3-point Likert-type scale ranging from 1 (none) to 3 (a lot). Total scores range from 18 to 54, where higher scores indicate higher levels of anxiety sensitivity (Silverman et al., 1991). The CASI total score has demonstrated good
internal consistency ($\alpha = .87$) and acceptable levels for test–retest reliability (Silverman et al., 1991). For this study, we employed the CASI total score, and the CASI total score demonstrated good internal consistency ($\alpha = .82$).

**Intolerance of Uncertainty Scale—Revised.** Intolerance of Uncertainty Scale—Revised (IUS-R; Walker et al., 2010) is a 12-item questionnaire designed to assess intolerance of uncertainty (IU) across the lifespan (i.e., allows comparison across children, adolescents, and adults) and is an adaptation of the 12-item version of IUR (Carleton et al., 2007). Items (e.g., Item 1—When things happen suddenly, I get very upset; Item 5. I always want to know what will happen to me in the future) are rated on a 5-point Likert-type scale ranging from 1 (not at all like me) to 5 (entirely like me). Total scores range from 12 to 60, with higher scores indicating a greater degree of intolerance of uncertainty. For this study, we employed the IUS-R total score, and the IUS-R total score demonstrated good internal consistency ($\alpha = .81$).

**Spence Children’s Anxiety Scale.** The Spence Children’s Anxiety Scale (SCAS; Spence, 1998) is a 44-item self-report measure used to assess symptoms regarding anxiety clusters in children (Spence, 1998). It comprises six subscales, including separation anxiety, social phobia, obsessive-compulsive disorder, panic/agoraphobia, generalized anxiety, and fears of physical injury (Spence, 1998). Items (e.g., Item 1—I worry about things; Item 9. I feel afraid that I will make a fool of myself in front of people) are rated on a 4-point Likert-type scale, ranging from 1 (never) to 3 (always). Total scores vary from 0 to 114, with higher scores indicating higher levels of anxiety. High internal consistency has been observed for the SCAS total score (i.e., $\alpha = .92$, Essau et al., 2002; Spence, 1998; Spence et al., 2003, 2003). Acceptable test–retest reliability for the SCAS total score over a 6-month period has been observed; however, test–retest reliability was poorer for the subscale totals (Spence, 1998; Spence et al., 2003). This scale has also displayed high concurrent validity (Spence et al., 2003). For this study, the SCAS total score demonstrated excellent internal consistency ($\alpha = .91$) and acceptable internal consistency for three of the SCAS subscales: $\alpha = .76$ (generalized anxiety), $\alpha = .76$ (separation anxiety), and $\alpha = .79$ (panic/agoraphobia). The remaining subscales demonstrated poorer internal consistency values: $\alpha = .60$ (physical injury fears), $\alpha = .69$ (social phobia), and $\alpha = .68$ (obsessive compulsiveness).

**Procedure**

Ethical approval was provided by the University of Saskatchewan Biomedical Research Ethics Board. Consent was obtained from parents/legal guardians of the participants, and assent was obtained from the child/adolescent participants. All participants completed the same battery of questionnaires. For participants with CHD, research assistants facilitated administration of measures approximately 1 week prior to involvement in the larger study. Similarly, research assistants facilitated administration of the measures to the typically developing participants in a similar setting on a prescheduled date.

**Statistical analyses**

Statistical analyses were performed using the IBM SPSS Statistics software package (SPSS: version 23). Descriptive statistics were computed for demographic information and questionnaire subscales and total scores. The statistical analyses included (1) a series of bivariate correlations computed between age and total scores on measures of health anxiety, anxiety sensitivity, intolerance of uncertainty, and DSM-IV anxiety disorder symptom categories to examine potential age differences in the constructs of interest; (2) a series of independent sample $t$-tests computed to examine potential gender differences in total scores on measures of health anxiety, anxiety sensitivity, intolerance of uncertainty, and DSM-IV anxiety.
disorder symptom categories; (3) a series of univariate analyses of variance (ANOVA) conducted to assess for potential group differences across total and subscale scores on measures of health anxiety, anxiety sensitivity, intolerance of uncertainty, and DSM-IV anxiety disorder symptom categories; and (4) two sets of bivariate correlations conducted separately to examine the associations between the CIAS and constructs of interest for each group.

Results

Descriptive statistics

Descriptive statistics were computed for demographic information and questionnaire subscales and total scores (see Table 1). A total of 84 children and adolescents aged 7–16 years (mean age = 11.65 years, SD = 2.53) participated in this study (36 females; mean age = 11.97 years, SD = 2.60 and 48 males; mean age = 11.42 years, SD = 2.48). No significant age differences were observed across groups, t(82) = 1.971, p = .052, or gender, t(82) = .000, p = 1.000. Various types of CHD were observed in the CHD group, with the most common types being Tetralogy of Fallot (21.4%; n = 9), hypoplastic left heart syndrome (16.7%; n = 7), pulmonary stenosis (4.8%; n = 2), coarctation of the aorta (4.8%; n = 2), transposition greater arteries (9.5%; n = 4), and tricuspid/pulmonary atresia (7.1%; n = 3). Aside from CHD conditions, 16.7 percent (n = 7) had one additional health condition, 9.5 percent (n = 4) had two additional health conditions, 2.4 percent (n = 1) had three additional health conditions, and 2.4 percent (n = 1) had four additional health conditions. The most common additional health conditions included having a high blood pressure—7.1 percent (n = 3), stroke—4.8 percent (n = 2), and celiac disease—4.8 percent (n = 2). Mental health conditions included attention deficit hyperactivity disorder (ADHD) 4.8 percent (n = 2), Aspergers 2.4 percent (n = 1), depression and anxiety 2.4 percent (n = 1), and post-traumatic stress disorder (PTSD) 2.4 percent (n = 1). One participant reported being currently prescribed anti-anxiety medication. In our typically developing group, there were minimal health conditions present: four children had a self-reported health condition.
(i.e. asthma, not specified, and a peanut allergy). In addition, one participant had a heart defect that self-resolved early in life (i.e. small ventricular septal defect). One typically developing participant did not complete the entire battery of measures and was excluded from the primary analyses. One participant in the CHD group only completed the measure of health anxiety (i.e. CIAS), therefore was only included in the associated primary analyses.

Bivariate correlations were computed between age and total scores on measures of health anxiety, anxiety sensitivity, intolerance of uncertainty, and DSM-IV anxiety disorder symptom categories to examine potential age differences in the constructs of interest. No statistically significant associations between age and the aforementioned measures were found. A series of independent samples t-tests were computed to examine potential gender differences in total scores on measures of health anxiety, anxiety sensitivity, intolerance of uncertainty, and DSM-IV anxiety disorder symptom categories. Results demonstrated that there were no statistically significant differences found, although the SCAS physical injury fears subscale scores did trend toward significance $t(1.95)=81; p=.055$.

**Group differences in health anxiety and associated constructs**

A series of ANOVAS was conducted to assess for potential group differences across subscale and total scores on the CIAS and associated constructs (see Table 1). Results demonstrated that children and adolescents with CHD demonstrated significantly higher CIAS total scores, $F (1, 82)=9.28$, $p=.003$, $\eta^2=.10$, CIAS treatment experiences subscale scores, $F (1, 82)=50.63$, $p<.001$, $\eta^2=.38$, and CIAS symptom effects subscale scores, $F (1, 82)=4.24$, $p=.043$, $\eta^2=.05$. Furthermore, children and adolescents with CHD demonstrated significantly higher CASI total scores, $F (1, 81)=4.59$, $p=.035$, $\eta^2=.05$, IUS-R total scores, $F (1, 81)=6.36$, $p=.014$, $\eta^2=.07$, and SCAS total scores, $F (1, 81)=6.02$, $p=.016$, $\eta^2=.07$. With respect to SCAS subscales, separation anxiety subscale scores, $F (1, 81)=6.63$, $p=.012$, $\eta^2=.08$, and physical injury fears subscale scores, $F (1, 81)=11.08$, $p=.001$, $\eta^2=.12$, were significantly higher in the CHD group. There was also a trend toward significant group differences for the SCAS social phobia subscale $F (1, 81)=3.63$, $p=.060$, $\eta^2=.04$.

**Association between health anxiety and associated constructs**

Bivariate correlations were computed between CIAS subscale and total scores and total scores of the CASI, IUS-R, and SCAS, as well as the SCAS subscales scores (see Table 2—with correlation coefficients for CHD group on the left hand side and correlation coefficients for the typically developing group on the right). Results showed statistically significant associations between the CIAS total score and all total scores from measures of interest for both groups, with the exception of IUS-R where the correlation was significant only for the typically developing group. Significant associations were also observed between the majority of CIAS subscales and constructs of interest. However, the CIAS help-seeking and treatment experience subscales were associated with very few of the total scores from the measures of interest.

**Discussion**

This study is the first of its kind to examine self-reported health anxiety and associated constructs in children and adolescents with CHD. Consistent with our primary hypothesis, children and adolescents with CHD reported significantly higher levels of health anxiety (as measured by CIAS total score) compared to typically developing children. Our findings differ from Rask et al. (2016) findings that demonstrated that self-reported health anxiety symptoms at ages 11–12 were not associated with chronic physical health conditions. A noted limitation was that Rask et al. (2016) did not recruit children or adolescents with specific chronic physical health conditions and the most common physical condition in their sample was asthma. As such, the latter findings may not be
generalizable to populations of children with chronic physical health conditions (such as CHD) that may be associated with more significant functional limitations. Our current findings are consistent with those of adult populations with certain chronic physical health conditions (i.e. chronic pain, fibromyalgia, Ménière’s disease, multiple sclerosis, and noncardiac chest pain; Bozkurt Zincir et al., 2014; Kehler and Hadjistavropoulos, 2009; Kirby and Yardley, 2009; Rode et al., 2006; Uçar et al., 2015).

Our findings also demonstrated that those with CHD experience a greater number of treatment experiences compared to our control group (as measured by the CIAS treatment experiences subscale). This finding may be intuitive given that children with CHD are followed by medical professionals (e.g. cardiologists) regularly throughout their development. Our findings also demonstrated that those with CHD endorsed significantly higher scores on the CIAS symptom effects subscale, suggesting that those with CHD experience significantly more functional impairment from the awareness of bodily changes (e.g. unusual sensation), which may or may not be medically explained.

Children and adolescents with CHD reported significantly higher levels of anxiety sensitivity and intolerance of uncertainty. Our findings appear to parallel some of the related existing research in child/adolescent and adult samples with chronic physical health conditions where anxiety sensitivity was explored (e.g. Anderson et al., 2014; Asmundson et al., 2000; Lavoie et al., 2003; Lipsitz et al., 2004). However, there are mixed findings across this subset of the literature (e.g. Pritchard et al., 2003). Having a physical health condition, such as a CHD, where symptoms (prognosis) and surgical procedures may be unpredictable, may lead to the elevations in intolerance of uncertainty we also observed. This pattern has been observed in an adult sample with Ménière’s disease, where those with Ménière’s disease demonstrated significantly higher levels of intolerance of uncertainty than controls (Kirby and Yardley, 2009). Furthermore, exploration of the source of uncertainty in child and adolescents with CHD (e.g. symptoms, upcoming surgeries, and types of surgeries) is warranted, as has not been explored to date.

Consistent with existing research examining general psychological functioning of children and adolescents with chronic health conditions (e.g. Pinquart and Shen, 2011; Sztein and Lane, 2016), participants with CHD demonstrated significantly higher anxiety disorder symptoms (as measured by the SCAS) than typically developing controls. In particular, children and adolescents with CHD reported significantly higher

<table>
<thead>
<tr>
<th>Measure</th>
<th>CIAS total score</th>
<th>CIAS fear</th>
<th>CIAS HS</th>
<th>CIAS TE</th>
<th>CIAS SE</th>
</tr>
</thead>
<tbody>
<tr>
<td>CIAS total score</td>
<td>.65**</td>
<td>.58**</td>
<td>.65**</td>
<td>.54**</td>
<td>.49*</td>
</tr>
<tr>
<td>SCAS total score</td>
<td>.57**</td>
<td>.42**</td>
<td>.69**</td>
<td>.42**</td>
<td>.34*</td>
</tr>
<tr>
<td>Separation anxiety</td>
<td>.52**</td>
<td>.22</td>
<td>.62**</td>
<td>.32*</td>
<td>.20</td>
</tr>
<tr>
<td>Social phobia</td>
<td>.25</td>
<td>.26</td>
<td>.37*</td>
<td>.22</td>
<td>.11</td>
</tr>
<tr>
<td>Obsessive compulsive</td>
<td>.41**</td>
<td>.41**</td>
<td>.42**</td>
<td>.43**</td>
<td>.26</td>
</tr>
<tr>
<td>Panic/agoraphobia</td>
<td>.43**</td>
<td>.39*</td>
<td>.53**</td>
<td>.34*</td>
<td>.21</td>
</tr>
<tr>
<td>Physical injury Fears</td>
<td>.35*</td>
<td>.14</td>
<td>.40**</td>
<td>.18</td>
<td>.03</td>
</tr>
<tr>
<td>Generalized anxiety</td>
<td>.61**</td>
<td>.48**</td>
<td>.74**</td>
<td>.46**</td>
<td>.24</td>
</tr>
<tr>
<td>IUS-R total score</td>
<td>.26</td>
<td>.48**</td>
<td>.22</td>
<td>.42**</td>
<td>.07</td>
</tr>
</tbody>
</table>

CIAS: Childhood Illness Attitude Scales; HS: help-seeking; TE: treatment experience; SE: symptom effects; CASI: Children’s Anxiety Sensitivity Index; SCAS: Spence Children’s Anxiety Scale; IUS-R: Intolerance of Uncertainty Scale–Revised.

The values on the left hand side of the column are correlation coefficients for the CHD group and those on the right are for the typically developing group.

*p < .05; **p < .001.
levels of separation anxiety, physical injury fears, and levels of social anxiety trending toward significance (i.e. \( p = .06 \)). These findings may be partially explained by considering that this group of children and adolescents (as a function of their chronic physical health condition) may have had fewer substantive social interactions due to higher rates of medically related school absences and, therefore, less opportunities to develop appropriate social skills and sense of self-efficacy as it pertains to peer interaction and independence (Masi and Brovedani, 1999). Peer interaction via physical extra-curricular activities may have been limited by parents and/or child due to worries regarding child’s current physical health status being negatively impacted or exacerbated by engagement in physical activities (Ong et al., 2011). Furthermore, elevations in social anxiety could arise when these children and adolescents are in situations when surgical scars may become visible (e.g. changing into swim or gym clothing, swimming, and gymnastics). Individuals with visible differences (e.g. surgical scars) have reported having difficulties, including experiencing social anxiety, fear of negative social evaluations, lower self-esteem and body image, and social avoidance (Rumsey and Harcourt, 2004). Prior work has shown that adults with CHD may have body image concerns about surgical scars (Kovacs et al., 2005), as do adolescents (Masi and Brovedani, 1999). Elevations in these anxiety disorder symptom categories warrant future examination.

Our findings also supported our second hypothesis, in that we observed statistically significant associations between CIAS total score and the CASI, SCAS, and IUS-R total scores across both groups, with the exception of IUS-R where the association was significant only for the typically developing group. Our findings are generally consistent with the results found within adult medical populations (e.g. Jones et al., 2014; Kehler and Hadjistavropoulos, 2009). However, the majority of existing related research focuses on the relationship between health anxiety and psychopathology (e.g. depression and/or anxiety disorders; Bennett et al., 2016; Hadjistavropoulos et al., 2002; Kehler and Hadjistavropoulos, 2009; Szakats et al., 2015; Thew et al., 2016; Uçar et al., 2015) as opposed to the associations between health anxiety and intolerance of uncertainty and anxiety sensitivity, more specifically. Thus, our study extends the existing literature by adding initial findings regarding the association between health anxiety and intolerance of uncertainty in a child and adolescent sample with CHD.

Limitations and future directions

There are several limitations that require attention. First, this study is a smaller component of a larger study, as such certain study criteria were directed by the larger study (i.e. sample size). Our effect sizes ranged from small to large (partial \( \eta^2 = .07-.38 \)) suggesting that the group differences are fairly large in some cases, even with our sample size (i.e. 84). That said, we do have a relatively small sample size and our analyses may be underpowered. Future studies should seek to obtain a larger sample size to substantiate our findings. Second, we utilized self-reports, a commonly used approach, when assessing constructs of interest. Research assistants were available to facilitate with this process, in a nondirective manner (i.e. did not direct responses but provided explanation for measure items when requested). However, parents helped in certain situations resulting in responses that may have been impacted by the assistance provided or by parental presence.

Conclusion

This is the first study to examine health anxiety and associated constructs in children and adolescents with a specific chronic physical condition and, more specifically, in children and adolescents with CHD. Our findings highlight that children and adolescents with CHD experience higher levels of health anxiety and associated constructs than typically developing children. These findings appear to speak to the psychological needs of children and adolescents
with CHD, possibly highlighting the need for specialized intervention. More specifically, these findings may serve to guide the development and implementation of specialized interventions for this population (i.e. focus on health anxiety, anxiety sensitivity, intolerance of uncertainty, and specific anxiety disorders symptoms such as separation anxiety).

Declaration of Conflicting Interests
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