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ORIGINAL CONTRIBUTION

# Effects of maternal health anxiety on children's health complaints, emotional symptoms, and quality of life

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**Abstract** Little is known about family risk factors and intergenerational transmission of psychological disturbance in the development of health anxiety (HA). This study investigated HA and related concepts in 8- to 17-year-old children who had been exposed to different maternal health status. Using a family case–control design, three family groups were included: (1) 50 case children of mothers with severe (HA); (2) 49 control children of mothers with rheumatoid arthritis (RA); and (3) 51 control children of healthy mothers. Children and mothers completed a battery of standardised questionnaires. Case children reported significantly higher level of HA symptoms than children of mothers with RA but not compared to children of healthy mothers. There was no significant difference between the children's self-reports in the three groups with regard to anxiety symptoms in general, physical complaints, or quality of life. In contrast, mothers with HA reported their children as having more emotional and physical symptoms than mothers in one or both control groups. Compared to mothers with RA but not healthy mothers, mothers with HA also reported more visits to the general practitioner with their children during the past year. The findings suggest that maternal HA only weakly affects children's own report of HA and thereby may not be a strong risk factor for the development of HA symptoms in childhood. However, mothers with severe HA seem to conceive their children as more ill and present them more often in the health care system which could, therefore, be an important target for intervention in adult patients.

**Keywords** Health anxiety · Hypochondriasis · Child and adolescent · Parental health status · Family case–control study

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## Introduction

Severe health anxiety (HA) (also known as illness anxiety disorder [1] or hypochondriasis [2]) refers to apprehension and fear concerning one's health. The essential feature is an exaggerated rumination with worries of having a serious illness leading to a significant decrease in quality of life [3]. Severe and persistent HA in adults is estimated to 3.4% in the general population [4] and 9.5% in primary care consultations [3]. Severe HA is costly for society in terms of increased health expenditure [5] and reduced work capacity [6].

Growing research suggests that HA may originate in childhood [7–13], and studies have demonstrated that cognitive and behavioural features similar to those described for HA in adults may also be present in younger age groups [7, 11, 13]. The development of HA probably has a complex nature involving a number of interacting factors such as genetics and social learning [14]. It is possible that

genetic factors cause a vulnerability to develop anxiety disorders more broadly [15] and that the direction towards HA is influenced by an interaction of factors such as early illness experiences [16] and social learning from significant others (mainly parents), of maladaptive illness behaviours [17], and negative health beliefs [8].

To the best of the authors' knowledge, only three studies have highlighted a possible transmission of HA from parents to their children [8, 11, 18]. All found a positive association between child HA symptoms and parental HA symptoms but did not distinguish between maternal and paternal HA, though some studies on the intergenerational transmission of anxiety disorders in general have found that maternal anxiety is more strongly associated with child anxiety compared to paternal anxiety [19, 20].

The current research leaves a number of questions unanswered. The specific role of maternal HA for the development of childhood HA could be further elucidated by examining whether maternal HA also has a more overall negative effect on the child's emotional state and quality of life.

In addition, it should be clarified if any such effects on the child are specific to maternal HA or whether they are similarly found in children of mothers with other mental or chronic physical health conditions.

In the present study, we aimed to investigate the specific impact of maternal HA on children's emotional symptoms and health-related quality of life by comparing three groups: a case group of children raised by mothers with severe HA and two control groups of children raised by a mother with a chronic physical disease (rheumatoid arthritis (RA) and by a well mother, respectively. We hypothesised that the case group would especially be more likely to display higher levels of HA and physical complaints as well as more maladaptive illness behaviour compared to the two control groups.

## Methods

### Participants and procedure

Using a family case–control study design, three groups of children and their parents were recruited: (1) a case group of children with mothers diagnosed with severe HA, (2) a first control group of children with mothers diagnosed with RA, and (3) a second control group of children with healthy mothers. The first control group was selected to control for factors that were general for a child having a mother with a chronic and impairing condition. We chose mothers with RA as comparison group because RA is a chronic illness characterised by clear physical signs and symptoms observable for others, including the child. In addition, this disorder is typically marked by recurrences of severe illness episodes and, therefore, similar in the fluctuating illness pattern also seen in HA.

If a mother with HA had more than one child available for inclusion, one of them was randomly selected to participate. The children in the two control groups were matched with the children in the case group on age and gender.

Data were collected during the period April 2013 to September 2015. Mothers with HA were recruited from three university hospitals in Denmark (Aarhus, Køge, and Bispebjerg) at departments specialised in outpatient treatment of somatoform disorders including hypochondriasis or severe HA. All patients assigned to the department in Aarhus were, at some point during January 2012 to May 2015, identified from lists of outpatients diagnosed with severe HA according to the criteria described by Fink et al. [3].

Patients who had completed treatment were invited to participate by a letter or phone call by the first author, whereas current patients were consecutively invited to participate in relation to their visits at the departments. Mothers diagnosed with RA (ICD diagnosis of seronegative or seropositive rheumatoid arthritis) were recruited from outpatient lists from departments at three university hospitals and one regional hospital in Denmark and invited to participate by phone or letter. The healthy mothers were recruited by announcements in waiting rooms in eight general practices, on school intranets at six public schools in the Central Denmark Region, and on Facebook.

Mother and child separately completed electronic questionnaires at a hospital department or in their own home after being instructed in the technical procedure. A trained research assistant was present during the whole assessment to administer and assist if there were any questions. The fathers received an e-mail with a link to an electronic questionnaire. To increase participation, all participating families received a compensation of 75 dollars, and there was a draw among all the children to win two iPads.

### Inclusion and exclusion criteria

Children in the age group 8–17 years and their parents had to understand and speak fluent Danish. The family was excluded if the child was adopted or if one of the members (mother, father or child) had a known diagnosis of a severe mental disorder (e.g. psychosis or bipolar affective disorder) or a severe chronic physical disease, which required regular examination and follow-up visits in a hospital setting (e.g. diabetes, epilepsy, cancer). An exception was the first control group, where the mother was diagnosed with RA.

The assessment of mental and physical health status differed between the groups of participants. Mothers with HA underwent a thorough clinical assessment for mental

problems performed by a medical doctor or a trained psychologist with a modified version of the semi-structured psychiatric interview, Schedules for Clinical Assessment in Neuropsychiatry (SCAN) [3, 21]. To be eligible, the mothers had to fulfill diagnostic criteria for severe HA according to empirically established positive research criteria as described by Fink et al. [3]. These criteria include exaggerated rumination with intrusive worries about harbouring serious illness and persistent preoccupation with one's health leading to significant impairment. Inclusion criterion was severe HA of at least 2 years' duration from the first episode. In case of other psychiatric disorders such as depression or anxiety, these did not explain the HA symptoms and, HA was considered to be the dominant disorder. In addition, a review of the HA mother'-s' medical records was performed by a doctor in order to exclude severe comorbid physical disorders.

RA was diagnosed according to The American College of Rheumatology (ACR) criteria [22] in a hospital setting. Inclusion criterion was RA of at least 2 years' duration with a severity that required follow-up at an outpatient clinic on a regular basis. The medical records were reviewed by a doctor to ensure exclusion of diagnoses of mental disorders, including HA, and other severe physical disorders than RA.

Healthy mothers' health status was confirmed by contact to their general practitioner (GP).

The children's health status was confirmed by their mothers, i.e. the child did not have a diagnosed severe physical or mental disorder prior to study participation.

Father'-s' well health status was confirmed by self-reports or by reports from the mother.

## Assessment

### *Parental assessment*

Mothers' and fathers' HA and mental and physical functioning was assessed by the following:

*The Whiteley 7-Index (WI-7)* was used as an indicator of degree of parental HA. It measures illness worry, e.g. "do you worry a lot about your health" and has demonstrated satisfactory psychometric properties in primary care sample with a satisfactory internal validity (Cronbach's $\alpha = 0.68$) [5] and very good external validity for screening DSM-IV hypochondriasis/HA [23, 24]. Each item is Likert-scored from 1 (not at all) to 5 (a lot) with a total sum-score ranging from 7 to 35. In the present study, scores were transformed to a 0 to 100 score point scale with high scores indicating a high level of HA [5] to facilitate comparison with other studies using other versions of the Whiteley Index.

In the current study, the measure demonstrated excellent internal consistency ($\alpha = 0.96$).

*The Medical Outcomes Study 36-item Short-Form Health Survey (SF-36)* which is a generic instrument to assess health-related quality of life. In the present study, the summary scores for the overall mental and physical health, respectively, were used [i.e. the physical (PCS) and the mental component summary (MCS)] [25]. The two subscales are scored using norm-based methods with a mean of 50 and a standard deviation of 10, where scores over 50 indicate better functioning than the average. It exists in a well-validated Danish version and Danish norm data are available [26, 27].

### *Child assessment*

The children underwent a brief medical examination (including height, weight, blood pressure, and stethoscopy of heart and lungs), conducted by a doctor or a trained medical student, to exclude undetected physical disease. The child's illness behaviour, i.e. doctors visits and school absence due to illness, was assessed by maternal proxy reports and information obtained from schools, whereas emotional symptoms and health-related quality of life were assessed by both maternal proxy and self-reports.

### *Maternal proxy reports*

*The Strength and Difficulties Questionnaire* (SDQ, parent version) [28] is a widely used screening instrument to assess emotional and behavioural problems in children and adolescents. It consists of 25 items scored on a 3-point rating scale and is divided into five subscales: emotional problems, hyperactivity, conduct problems, peer problems, and prosocial (the latter subscale is not included in the total sum-score). The total score ranges from 0 to 40, whereas the emotional problems scale ranges from 0 to 10 with high scores indicating high level of problems on both scales. Its psychometric properties are well established [29, 30], and Danish norm data exist [31]. In the current study the internal consistency was good ($\alpha = 0.76$).

*The Children's Somatization Inventory* (CSI-24, parent version) [32] assesses 24 non-specific physical symptoms experienced in the previous 2 weeks in children and adolescents. It is rated on a 5-point rating scale with a total sum score ranging from 0 to 96 and high scores indicating more symptoms or symptom severity. It has demonstrated excellent psychometric properties [32], and the internal consistency in the current study was good ($\alpha = 0.88$).

*Kidscreen-27* (parent version) [33] is a well-validated measure to assess health-related quality of life during the last week for children and adolescents. The physical (5 items) and the psychological (7 items) well-being subscales are scored on a 5-point scale. Each dimension is reported as $t$ values with scales means around 50 and standard

deviations around 10 with high values indicating high health-related quality of life. A Danish version exists, and the internal consistency in the present study was satisfactory ($\alpha = 0.89$).

*The Spence Children's Anxiety Scale* (SCAS-P, parent version) [34] is a 38-item measure of anxiety symptoms in children and adolescents. The items reflect symptoms of the main DSM-IV child anxiety disorders. Each item is rated on a 4-point scale with total scores ranging from 0 to 114 where high scores reflect high levels of anxiety. The items can be divided into six subscales corresponding to separation anxiety, social phobia, obsessive–compulsive disorder (OCD), and generalised anxiety (GAD) (all consisting of 6 items, scoring range 0–18), panic/agoraphobia (9 items, scoring range 0–27), and physical injury fears (5 items, scoring range 0–15). A Danish version exists [35], and the Cronbach's alpha in the present study was excellent ($\alpha = 0.91$).

### *School absence, medical consultations, and use of pain killers*

The mothers filled out study-specific questions regarding their child's: (1) number of visits to the GP (categorised as ≥3 and <3 times during the past year, respectively), (2) school absence due to illness during the year prior to study participation (categorised as none, <1 week and ≥1 week), and (3) the child's intake of pain killers during the past 14 days (categorised as yes/no). Additional data on school absence due to illness during the past year were obtained from the child's school.

### *Child self reports*

*The Childhood Illness Attitude Scales* (CIAS) [10] was used as an indicator of the child's HA symptoms. It consists of 35 items and is adapted from the Illness Attitude Scale used in adults [36]. It assesses fears, beliefs, and attitudes that are associated with hypochondriasis and abnormal illness behaviour in school-aged children. Items are rated on a 3-point rating scale with the exception of items 29–31 which are rated on a 3-point frequency scale. In total, 33 of the 35 items are used for the scoring except for item 28 and 32 which are open-ended. Thus, total scores range from 33 to 99 reflecting the severity of HA symptoms with high scores indicating more HA symptoms. The CIAS has shown good psychometric properties including a high test–retest (10–14 days) reliability ($r = 0.86$), a high internal consistency ($\alpha = 88$) and concurrent validity with similar constructs such as the Childhood Anxiety Sensitivity Index [10, 37]. In the current study, the measure demonstrated good internal consistency ($\alpha = 0.86$).

*The Children's Somatization Inventory* (CSI-24, child version) (see above) scoring of the questionnaire is identical to the parent version. The internal consistency was satisfactory in the present study ($\alpha = 0.85$).

*Kidscreen-27* (child version) (see above) [38], the physical and psychological well-being dimensions. Scoring is identical to the parent version. The Cronbach's alpha in the present study was ($\alpha = 0.86$).

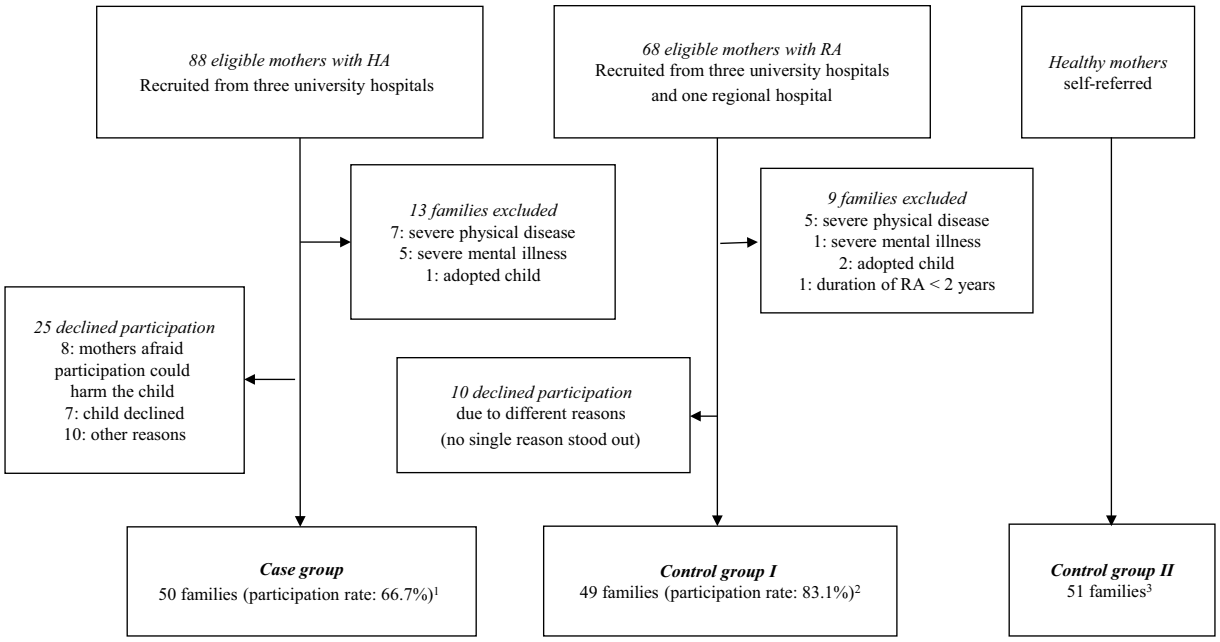
*The Spence Children's Anxiety Scale* (SCAS, child version) [39] (see above) consists of 44 items where six are added to reduce negative response bias and are not included in the scoring. The total scores range from 0 to 114 similar to the parent version. In the current study, the measure demonstrated good internal consistency ($\alpha = 0.83$).

## Data analysis plan

Descriptive statistics were used to characterise the three groups and to provide raw data on primary and secondary outcomes. All scores were summarised using a percentage, the mean and standard deviation (SD), or the median and inter quartile range (IQR).

First, a characteristic of the three groups was made with regard to (1) maternal age and educational level; (2) maternal and paternal HA symptoms (WI-7), and health-related quality of life (PCS and MCS scores), and (3) the children's gender and age. Second, in our main analysis, the child's self-reported HA and physical symptoms as well as maternal-reported physical symptoms in the child were compared in the three groups. We also analysed difference between the groups in illness behaviour, i.e. illness-related school absence, recent use of painkillers, and number of contacts to the GP during the past year. In addition, we compared self-reported and maternal proxy-reported anxiety symptoms in the children in all three groups and further the distributions of Kidscreen scores (physical and psychological health-related quality of life) and maternal proxy-reported SDQ scores (emotional problems and total sumscore) were recorded. Finally, in order to categorise the number of children with high- and low self-reported HA symptoms, respectively, in the three groups we used a cut-off value of 62 on the CIAS, based on the 90th percentile in a normal population sample of 11-to 12-year-old Danish children [13] to define a "probable case of HA".

For variables that were non-normally distributed or did not have equal SDs within the three groups, the Kruskal–Wallis and Wilcoxon's ranksum tests were used to compare the distribution of the groups. For all other continuous variables, the between-group comparison was done using a one-way ANOVA, whereas in cases where the dependent variable was dichotomous, a $\chi^2$-test was used. We adjusted for multiple testing using Bonferroni-correction




HA = health anxiety; RA = rheumatoid artritis
[1]Paternal self-reports: N=39 (95%), [2]Paternal self-reports: N=35 (87%), [3]Paternal self-reports: N=44 (90%)

**Fig. 1** Flowchart of participants

in all analyses (a total of 20 analyses), except for the ones concerning the main analysis which reflected our primary hypothesis. All statistical analyses were done using STATA version 13 for Windows.

## Results

### Background characteristics

The final study sample was composed of 150 families: an HA group of 50 families with a mother diagnosed with severe HA, an RA control group of 49 families with a mother diagnosed with RA, and a healthy control group of 51 families with a healthy mother (Fig. 1).

Demographic characteristics were similar across the three groups of children: the HA group 11.8 years (SD 2.40), the RA group 12.3 years (SD 2.50), and the healthy group 11.6 years (SD 2.32), $F(2,147) = 1.17$, $p = 0.314$. The ratio of girls/boys were: HA group: 58% (29/50), RA group: 45% (22/49), healthy group: 55% (28/51), $\chi^2(2) = 1.859$, $p = 0.395$.

Among the three groups, mothers with RA were the oldest and reported the lowest physical health- related quality of life, whereas mothers with HA reported the highest level of HA symptoms as well as the lowest mental health-related quality of life (Table 1). For fathers, no statistically significant differences were found between the groups with respect to degree of HA symptoms or physical and mental health-related quality of life (all $p$ values >0.05).

### Child self-reports

Case children reported significantly more HA symptoms compared to children of mothers with RA, but not compared to children of healthy mothers (Table 2). No significant differences in the report on physical symptoms (Table 2), anxiety symptoms measured by the overall SCAS score (Table 3), and health-related quality of life (Table 4) were found between the three groups. Twenty-five children in total had a CIAS score of ≥62 indicating that they were probable cases with clinically significant HA (HA group: 24.0% (12/50), RA group: 4.0% (2/49), healthy group: 21.6% (11/51). The number of probable cases was significantly higher in the HA group compared to the RA group ($\chi^2(1)$: 8.09, $p = 0.004$), whereas no statistical difference was seen between the HA and healthy groups ($\chi^2(1)$: 0.08, $p = 0.771$).

### Maternal proxy reports

Compared with either one or both control groups, mothers with HA reported significantly higher levels for their child on physical symptoms, contacts to the GP (Table 2), total anxiety score, and panic anxiety subscore (not significant after adjustment for multiple testing)

**Table 1** Maternal sociodemographics and maternal and paternal health anxiety and health-related quality of life

| Maternal | HA group ($N = 50$) | RA group ($N = 49$) | Healthy group ($N = 51$) | $p$ value (overall) |
|---|---|---|---|---|
| Mean age (SD) | 41.1 (4.2) | 45.1 (5.5) | 42.3 (5.2) | **<0.001**[a] |
| Medium or long education, $N$ (%) | 37.0 (74.0) | 29.0 (59.2) | 41.0 (80.4) | 0.062[b] |
| Health anxiety (WI-7), median (IQR) | 46.4 (28.6–64.3) | 7.1 (0.0–14.3) | 0.0 (0.0–7.1) | **<0.001**[c] |
| Physical health (PCS, SF-36), mean (SD) | 50.3 (6.2) | 42.3 (9.8) | 52.5 (5.9) | **<0.001**[c] |
| Mental health (MCS, SF-36), mean (SD) | 40.3 (12.8) | 54.8 (8.5) | 54.8 (5.5) | **<0.001**[c] |
| Paternal | HA group ($N = 39$) | RA group ($N = 35$) | Healthy group ($N = 44$) | $p$ value (overall) |
| Health anxiety (WI-7), median (IQR) | 0.0 (0–7.1) | 0.0 (0–7.1) | 0.0 (0–7.1) | 0.823[c] |
| Physical health (PCS, SF-36), mean (SD) | 54.2 (3.6) | 53.6 (5.7) | 54.7 (4.5) | 0.566[c] |
| Mental health (MCS, SF-36), mean (SD) | 53.2 (8.0) | 55.0 (6.7) | 54.2 (8.6) | 0.431[c] |

Bold values indicate $p < 0.05$

*HA* health anxiety, *RA* rheumatoid arthritis, *SD* standard deviation, *WI-7* Whiteley-7, *IQR* inter-quartile range, *PCS* physical component summary of the SF-36, *MCS* mental component summary of the SF-36

[a] ANOVA

[b] Chi-square

[c] Kruskal–Wallis

**Table 2** Child health anxiety, physical symptoms and illness-related behaviour

| | HA group ($N = 50$) | RA group ($N = 49$) | Healthy group ($N = 51$) | HA vs. RA $p$ value | HA vs. healthy $p$ value |
|---|---|---|---|---|---|
| Child report | | | | | |
| Health anxiety (CIAS), mean (SD) | 57.1 (9.2) | 53.6 (6.9) | 54.8 (8.7) | **0.041**[a] | 0.167[a] |
| Physical symptoms (CSI-24), mean (SD) | 9.7 (8.0) | 10.8 (8.3) | 8.7 (8.5) | 0.495[a] | 0.562[a] |
| Maternal proxy report | | | | | |
| Physical symptoms (CSI-24), mean (SD) | 8.0 (10.3) | 4.3 (3.7) | 4.1 (3.4) | **<0.006**[a] | **<0.004**[a] |
| Contacts to the GP past year, ≥3 times, $N$ (%) | 13 (26.0) | 3 (6.1) | 6 (11.8) | **0.007**[b] | 0.067[b] |
| Painkiller past 2 weeks (yes), $N$ (%) | 7 (14.0) | 6 (12.2) | 9 (17.6) | 0.796[b] | 0.616[b] |
| School absence past year due to illness, $N$ (%) | | | | 0.296[b] | 0.390[b] |
| No | 9/50 (18.0) | 14/48 (29.2) | 12/51 (23.5) | | |
| Yes (<1 week) | 29/50 (58.0) | 27/48 (56.3) | 32/51 (62.7) | | |
| Yes (≥1 week) | 12/50 (24.0) | 7/48 (14.6) | 7/51 (13.7) | | |

Bold values indicate $p < 0.05$

*HA* health anxiety, *RA* rheumatoid arthritis, *CIAS* the Childhood Illness Attitude Scales, *SD* standard deviation, *CSI-24* the Children's Somatization Inventory, *GP* general practitioner

[a] ANOVA

[b] Chi-square

(Table 3). In addition, they also reported significantly more overall mental problems as well as emotional symptoms in their children (Table 4). Finally, mothers with HA reported lower psychological well-being in their children compared to mothers with RA and lower physical well-being in their children compared to healthy mothers (not significant after adjustment for multiple testing) (Table 4).

## Data from schools

Information regarding illness-related school absence in the past year obtained directly from the schools did not show a difference between the three groups of children consistent with the mothers' reporting in the three groups (Table 2). Data obtained from the schools regarding absence corresponding to 0 days during a year in the three groups was HA = 17.8%

**Table 3** Child anxiety symptoms

| | HA group ($N = 50$) | RA group ($N = 49$) | Healthy group ($N = 51$) | HA vs. RA $p$ value[a] | HA vs. healthy $p$ value[a] |
|---|---|---|---|---|---|
| Child report (SCAS), median (IPQ) | | | | | |
| Total score | 20 (14–32) | 19 (9–27) | 19 (11–27) | 0.223 | 0.141 |
| Separation anxiety | 3 (1–5) | 3 (1–4) | 2 (1–4) | 0.621 | 0.134 |
| Social anxiety | 5 (3–6) | 4 (3–6) | 4 (3–6) | 0.432 | 0.406 |
| Obsessive–compulsive disorder | 3 (1–6) | 3 (1–5) | 2 (1–4) | 0.958 | 0.267 |
| Panic | 1 (0–3) | 1 (0–2) | 1 (0–2) | 0.180 | 0.325 |
| Physical injury fears | 3 (1–5) | 3 (1–4) | 3 (1–5) | 0.308 | 0.648 |
| Generalised anxiety disorder | 5 (3–7) | 4 (2–5) | 4 (3–6) | 0.016* | 0.169 |
| Maternal proxy report (SCAS), median (IQR) | | | | | |
| Total score | 11 (6–23) | 8 (5–14) | 8 (4–14) | 0.029* | 0.011* |
| Separation anxiety | 2 (1–5) | 1 (0–3) | 1 (0–3) | 0.106 | 0.046* |
| Social anxiety | 3 (1–5) | 3 (1–4) | 2 (1–4) | 0.404 | 0.252 |
| Obsessive–compulsive disorder | 0 (0–2) | 0 (0–1) | 0 (0–1) | 0.105 | 0.064 |
| Panic | 0 (0–2) | 0 (0–0) | 0 (0–0) | 0.025* | 0.009* |
| Physical injury fears | 2 (1–3) | 1 (0–3) | 1 (0–3) | 0.067 | 0.056 |
| Generalised anxiety disorder | 3 (2–5) | 2 (1–3) | 2 (1–3) | 0.013* | 0.004* |

*HA* health anxiety, *RA* rheumatoid arthritis, *SCAS* the Spence Children's Anxiety Scale, *IQR* inter-quartile range

* Adjustment for multiple testing (Bonferroni): $p$ values ≤0.0025 (0.05/20) are considered statistically significant

[a] Kruskal–Wallis

**Table 4** Child health-related quality of life and emotional symptoms

| | HA group ($N = 50$) | RA group ($N = 49$) | Healthy group ($N = 51$) | HA vs. RA $p$ value | HA vs. healthy $p$ value |
|---|---|---|---|---|---|
| Child report | | | | | |
| Kidscreen, mean (SD) | | | | | |
| HRQoL, physical well-being | 50.0 (7.3) | 49.8 (10.1) | 52.7 (8.3) | 0.906[a] | 0.129[a] |
| HRQoL, psychological well-being | 51.4 (7.5) | 52.8 (7.8) | 51.9 (7.5) | 0.353[a] | 0.727[a] |
| Maternal proxy report | | | | | |
| Kidscreen, mean (SD) | | | | | |
| HRQoL, physical well-being | 48.9 (10.4) | 51.4 (10.4) | 53.7 (8.3) | 0.201[a] | 0.016[a]* |
| HRQoL, psychological well-being | 46.4 (8.6) | 50.6 (8.3) | 49.5 (6.8) | 0.009[a]* | 0.058[a] |
| SDQ, median (IQR) | | | | | |
| Total score | 6.5 (4–12) | 5 (3–8) | 4 (2–7) | 0.014[b]* | **<0.001**[b] |
| Emotional problems (subscale) | 2.0 (1–6) | 1 (0–2) | 1 (0–2) | 0.014[b]* | **<0.001**[b] |

Bold values indicate $p < 0.05$

*HA* health anxiety, *RA* rheumatoid arthritis, *SD* standard deviation, *HRQoL* health related quality of life, *SDQ* the Strength and Difficulties Questionnaire, *IQR* inter-quartile range

* When adjusted for multiple testing (Bonferroni) only $p$ values ≤0.0025 (0.05/20) are considered statistically significant

[a] ANOVA

[b] Kruskal–wallis

(8/45), RA = 15.4% (6/39), healthy = 16.3% (7/43). School absence <1 week during a year was HA = 60.0% (27/45), RA = 64.1% (25/39), and healthy = 65.1% (28/43), and finally school absence ≥1 week during a year was HA = 22.2% (10/45); RA = 20.5% (8/39), healthy group = 18.6% (8/43). No significant statistical differences were found; HA vs. RA group; $\chi^2(2) = 1.157$, $p = 0.953$ and HA vs. healthy group; $\chi^2(2) = 0.262$, $p = 0.910$.

## Discussion

Overall, this family case–control study found that children of mothers with HA reported statistically significantly, although weakly, more HA symptoms than children of mothers with RA, but not more than children of healthy mothers. Furthermore, a significantly higher proportion, i.e. 24.0%, of children of HA mothers reported HA symptoms at a level defined as "being a probable case of HA" compared to only 4.0% of the children of mothers with RA, whereas there was only a trend towards a difference in comparison with children with healthy mothers (21.6%). When using maternal proxy reports, mothers with HA in general reported more health-related problems in their children, i.e. more physical and emotional symptoms as well as more frequent attendance to the GP with their child. Taken together, the results indicate that maternal HA only weakly affects children's own report of HA, suggesting that it is not a strong risk factor for the development of HA symptoms in children aged 8-17 years. However, the results may on the other hand suggest that mothers with severe HA conceive their children as more ill and also present their children more often in the health care system.

Different study designs and methods could be the likely explanation for the somewhat divergent findings in the present study compared to previous investigations on intergenerational transmission of HA [8, 11, 18]. Two of the prior studies used a cross-sectional population-based sample without control groups. The study by Wright et al. [11] included children with a mean age of 10.6 years (females) and 11.1 years (males) and found that only when combined with depressive symptoms, parental/guardian HA was associated with child HA. Koteles et al. [8] included adolescents with a mean age of 16.2 years and found a positive association between parental and adolescent HA. This could indicate that transmission of HA in younger children presupposes larger parental influence and that mechanisms such as social learning may require a quite long exposure before intergenerational transmission of HA becomes evident. Also a potential genetic vulnerability may first be significant later in life. In the present study, 77.7% of the participating children were under the age of 14, and even though prospective studies suggest that smaller children might display HA symptoms [9, 13] and related anxiety disorders are common at this age [40], some anxiety problems become more prevalent in adolescence [41]. Finally, the third study using a case–control design [18] was limited by a small sample size and by the case group being parents with any somatoform disorder, not specifically HA. As somatoform disorders are a broad group of different disorders characterised by various clinical features, their case group is not directly comparable to our more homogeneous group solely consisting of mothers with HA. Furthermore, none of the three studies took into account the health status of the other parent.

In contrast to studies using a retrospective design thus being prone to recall bias [42–45], we did not find an association between HA and the early exposure to severe physical illness in the family (e.g. parents). On the contrary, children of mothers with RA reported the lowest level of HA symptoms which could suggest that early exposure to illness experiences could actually learn the child to master illness and thereby serve as a protective factor for the development of child HA if the early experience is in the presence of promotive factors [46]. With regard to other related concepts (self-reports on other anxiety symptoms, physical complaints, and health-related quality of life), the three groups of children did not differ significantly indicating that having a mother with either HA or a chronic physical disease does not affect the child's self-experienced well-being. However, it should be emphasised that we only investigated one risk factor, namely maternal health status, rather than the cumulative influence of various risk factors, which has previously been suggested as an important strategy [47].

The higher maternal proxy reports of health-related symptoms and lower health-related quality of life in case children could suggest biased perception and reporting by mothers with HA, which corroborates previous findings showing maternal anxiety to be a predictor for higher levels of maternal-reported child anxiety [48].

On the other hand, it is interesting that maternal and child scores on the child's physical and emotional symptoms were similar in the HA group, whereas in the other two groups they were at variance, with lower scores from maternal proxy-reports. It could, therefore, also be argued that the findings may actually reflect enhanced maternal vigilance and better appreciation of the child's actual symptoms in the HA group. Future prospective studies could explore in more detail the possible effect of maternal estimation of their children's symptoms on child health-related outcomes. In addition, the case children also had more visits to their GP during the past year compared to children of mothers with RA, which is in agreement with previous studies reporting maternal anxiety and worries regarding the child's health to significantly predict a higher health care use of the child [49, 50]. All in all, the findings could indicate that mothers with HA experience their child's health, both when it comes to emotional and physical symptoms, in a more worried and negative way leading to more health-seeking behaviour on behalf of their children.

### Strengths and limitations

The present study has several strengths. It is the first to use a family case–control design with two control groups to examine the possible transmission of HA and related constructs. Mothers with HA and RA were diagnosed at a hospital department and only included if their disorder had a duration of minimum 2 years to ensure that the children were exposed to illness for a long period of time. Furthermore, both the children and fathers were not to have any diagnosed severe physical or mental disorder to obtain three comparable groups of children only differing with regard to maternal health status exposure. Information was obtained from different sources, i.e. self-reports, maternal proxy reports, and from the schools, which strengthens the data validity. Finally, the data were collected independently for the mother and the child to exclude mutual influences on their responses.

The study has several limitations. First, there is a possibility of selection bias, particularly given the moderate participation rate in the HA group of 66.7%. Many of the mothers with HA declining participation gave as a reason that they feared that study participation could mentally harm or affect their child when he or she was being exposed to questions regarding death and illness. Furthermore, the mothers with HA were recruited from departments specialised in treating somatoform disorders. Thus they had very likely accepted their condition as HA and a considerable proportion had already received psychological treatment for HA. Another potential limitation was the self-referral of healthy mothers, which may have attracted women with a particular interest in this study, e.g. mothers of children displaying symptoms of HA. Thus, the children in this control group had a higher mean score on the CIAS (54.8) compared to the mean score (51.9) reported in a recent Danish cohort study on 11- to 12-year-old children [13]. In summary, these factors might have blurred any differences among the case group and the controls, especially with regard to the control group of children of healthy mothers. Second, the use of questionnaires to assess complex concepts such as health beliefs and attitudes in younger children may be of limited reliability [51]. The younger children (8–9 years) in general had difficulties completing the questionnaires themselves, and in most cases the research assistant had to read the questionnaires aloud, which might have influenced the answers. However, we used the CIAS, which currently is the only available measure to assess HA in young individuals [10]. Third, in accordance with the existing research on child anxiety the current study focused on the role of mothers in child HA. However, the study design precluded the possibility of examining differences between parental dyads within the groups. Thus, there is accumulating evidence that fathers are likely to have a significant impact on children's development of internalising symptoms [52]. It is, therefore, possible that having a non-HA father may modulate the possible effect of having a mother with HA, which could be the reason for the mixed findings with regard to differences in probable child HA cases between the case group and the two control groups. Fourth, a full history and duration of all types of psychological problems were not obtained in either children or parents. Therefore, some parents or children scoring higher on emotional distress may have had pre-existing psychological problems, including anxiety disorders, which may have preceded onset of maternal HA symptoms. We did not control for pre-existing health conditions in other family members, including siblings, which could also have affected the findings. Finally, the cross-sectional design means that assumptions that any of the found associations are causal cannot be made.

## Conclusion

This family case–control study found only weak evidence of increased risk of self-reported HA and related constructs in children being exposed to maternal HA. However, the findings based on the maternal reports do raise the possibility of the existence of parental HA by proxy. This is in line with descriptions of clinical experiences and observations of parents repeatedly presenting their child in the health care system because of a persistent fear that doctors are missing something in their child (not to be confused with Münchhausen by proxy) and thereby risking that the child is exposed to unnecessary and potential harmful investigations [53]. It could, therefore, be considered to specifically target parental worries about their child's health in the treatment and management of adults with HA. This may also prevent that maladaptive familial transactional cycles and communication regarding health-related issues over time induce HA vulnerability in the child, i.e. prevent that he or she might in adulthood turn to similar maladaptive illness behaviour as exposed to in the childhood [54]. However, further research is needed to fully determine the effect of parental negative health beliefs and HA on child outcome and will demand testing of multi-risk factor models, including the possible cumulative effect of being exposed to HA in both parents as well as prospective studies looking at the development and maintenance of child HA over time.

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**Compliance with ethical standards**

**Ethical standards** Prior to study start, the Science Ethics Committee of the Central Denmark Region (206/2011) was consulted to ensure that the study complied with the Helsinki Declaration II. The study was approved by the Data Protection Agency (1-16-02-285-12) and the Danish Health Authority (3-3013-201/1). Eligible families received detailed oral and written information about the project including the voluntary nature and anonymity of their participation. Parents gave oral and written informed consent and for children verbal assent was obtained in all cases prior to study participation.

**Conflict of interest** On behalf of all authors, the corresponding author states that there is no conflict of interest.

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