Health-Related Quality of Life and Distress of Parents of Children With Avoidant Restrictive Food Intake Disorder

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ABSTRACT

Objectives: Health-related quality of life (HRQOL) of children with avoidant restrictive food intake disorder (ARFID) is impaired.

Aim: To measure HRQOL and distress of parents of children with ARFID.

Methods: Cross-sectional cohort study. Parents of children with ARFID, visiting our multidisciplinary feeding team, completed questionnaires on the online Quality of Life in Clinical Practice portal; the Questionnaire for Adult’s Health Related Quality of Life to assess parental HRQOL and the Distress Thermometer for Parents. Reference groups of parents of healthy (HC) and chronically ill children (CIC) were used.

Results: Eighty-five mothers and 62 fathers of 89 children with ARFID (58% female, median age 1.9 years) were included (response rate 68%). No differences were found regarding HRQOL in 11 of 12 domains between parents of children with ARFID and HC. Mothers of children with ARFID reported significantly higher HRQOL regarding pain and fathers a significantly lower HRQOL on depressive emotions compared to HC. No differences were found in overall and clinical distress scores between parents of children with ARFID and HC/CIC. Mothers of children with ARFID had significantly higher distress scores regarding cognitive problems compared to HC and parenting problems in children <2 years compared to HC/CIC. Significantly higher distress scores on parenting problems in children <2 years were found in fathers of children with ARFID compared to HC/CIC.

Conclusion: Most HRQOL and distress scores of parents of children with ARFID were comparable to reference groups. Since parents of children with ARFID perceive a lack of understanding and support from the environment, professionals should suggest peer support through patient’s organizations. Furthermore, it is important to offer professional support since parents indicated that they would like to talk to a professional about their situation.

Key Words: avoidant restrictive food intake disorder, distress, health-related quality of life, pediatrics

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METHODS

Participants and Procedure
In this cross-sectional cohort study conducted in our Diagnostic Center for Feeding Problems in the Amsterdam UMC, the Netherlands, every patient with feeding problems (0–10 years) is evaluated by a multidisciplinary team including a pediatrician/pediatric gastroenterologist, psychologist, dietician and speech language pathologist. Parents were contacted by a letter and a telephone call before consultation, and were asked to register for the online Quality of Life in Clinical Practice (Dutch: Kwaliteit van Leven in Kaart; English: Quality of Life in Clinical Practice) portal (www.hetklikt.nu) (15). As part of standard care, sociodemographics and distress of the parents were assessed online and an additional questionnaire regarding HRQOL was used. All parents of children with ARFID had one or more underlying disorders and their health-related quality of life (HRQOL) was decreased on several subscales compared to both healthy and chronically ill controls (7). In general, chronic pediatric conditions affect both the pediatric patient and their parents/caregivers. Additional parental care for children with extra needs may disturb the social activities and employment status of the parents. Several parental psychosocial consequences such as anxiety, burnout and mood problems can occur consequently (9–13). Although feeding problems may have consequences for all caregivers and all caregiver interactions may impact the growth and development of the child, fathers are mostly disregarded in the literature (14). The aim of our study was to assess HRQOL and distress of mothers and fathers of children with ARFID, which have not been studied before.

We hypothesized that mothers and fathers of children with ARFID have a lower HRQOL and more distress compared to parents of healthy children.

Measures

Patient Characteristics and Sociodemographics
Patient characteristics including age, sex, height, weight, tube feeding, and underlying diseases were obtained prospectively through standardized consultation. Height for age (SDS) and weight for height (SDS) were measured by TNO growth charts (16). If appropriate, SDS were retrieved from disease-specific growth charts for height and weight (16). The medical history was categorized by the International Statistical Classification of Diseases and Related Health Problems 11th Revision (ICD-11, version 4/2019) classification system. The research physician checked whether the children fulfilled the criteria for ARFID according to the DSM-5 retrospectively (1). Parents reported parental sociodemographics including age, sex, country of birth, marital status, educational level, and employment status.

Definitions being used for prematurity, small for gestational age, acute malnourishment, and chronic malnourishment are shown in the Supplemental Digital Content, http://links.lww.com/MPG/C330.

Parental Health-Related Quality of Life
To assess parental HRQOL, we used the Questionnaire for Adult’s Health Related Quality of Life (TAAQOL), which is a well-validated, reliable questionnaire, measuring the health status problems weighted by the impact of problems on well-being on 12 domains: gross motor functioning, fine motor functioning, cognitive functioning, sleep, pain, social functioning, daily activities, sexuality, vitality, positive emotions, depressive emotions, and aggressiveness (17). Most domains consist of two parts: determining the prevalence of a health problem or limitation in the past month, and assessing the emotional response to this health problem/limitation. Answers were scored on 4-point scales. A single score is attributed to each combination of a domain determining the prevalence of a health problem or limitation and the corresponding emotional response. The scores range from 0 to 100, in which higher scores indicate a better HRQOL (17). Insufficient Dutch language proficiency was an exclusion criterion. The reference group of mothers and fathers of healthy children recruited from one high school and two elementary schools, located in the same area as our hospital described by Hatzmann was used (12). We selected parents of the same age as our population.

The internal consistency for the TAAQOL scores in our cohort of mothers and fathers assessing Cronbach’s alpha values are reported as supplemental digital content, http://links.lww.com/MPG/C330. Domains with unacceptable Cronbach’s alpha values in mothers (gross motor functioning: 0.477) and fathers (fine motor functioning: −0.23), indicating the items do not measure the same construct, are not shown (18,19).

Parental Distress

The Distress Thermometer for Parents (DT-P; Dutch: Last Thermometer voor Ouders (LTO)), a well-validated self-report brief questionnaire was used to identify distress and everyday problems among parents of children visiting our Diagnostic Center for Feeding Problems (20). It consists of an overall distress score, which ranges from 0 (no distress) to 10 (extreme distress). Higher scores indicate higher levels of parental distress, and a score >4 indicates clinically important distress. Furthermore, it consists of a problem list involving six problem domains: practical, social, emotional, physical, cognitive, and parenting, addressing distress sources in the past week, which can be answered by “yes” or “no”. The score of domain parenting and included items were different for parents of children <2 years and children ≥2 years, because different parenting problems exist in different age groups (20). Additionally, five other questions are being asked, including perceived environmental support, lack of understanding from the environment, chronic illnesses of the parents, how parents go along with medical staff, and if the parent would like to discuss his/her situation with a professional (20). The questionnaire was available in Dutch and English.

DT-P scores of mothers and fathers of ARFID patients were compared to mothers and fathers of both a healthy as well as a chronically ill population (0–18 years) from the Dutch general population described by van Oers (21). We selected parents with children of the same age as our population.

Domains with unacceptable Cronbach’s alpha values in mothers (parenting problems ≥2 years of age: 0.415 and social
problems: 0.448) and fathers (parenting problems >2 years of age: 0.000 and social problems: 0.432) are not shown (18,19).

**Statistical Analyses**

The Statistical Package for Social Science (IBM SPSS Statistics) version 25 was used. Normality of the distribution of data was checked by QQ-plots, box plots, histograms, Kolmogorov-Smirnov and Shapiro-Wilk tests. Differences between groups were analyzed using Fischer Exact tests for categorical variables and Mann-Whitney U tests for not normally distributed variables. Domains with unacceptable Crohnbach’s alpha valables were excluded from the analyses. Due to multiple testing, we set the level of significance at $P < 0.01$. The formula $r = Z/\sqrt{N}$ was used to calculate effect sizes ($r$) for not normally distributed parameters, in which $Z$ is the $Z$-value and $N$ is the total number of samples. Effect sizes were considered to be small, medium, and large by standard values 0.1, 0.3, and 0.5, respectively (19).

**RESULTS**

**Sociodemographic and Medical Parameters**

In total, 183 new pediatric patients were seen at our outpatient clinic because of feeding problems between September 12, 2014 and May 10, 2019. The response rate was 67.8% ($n = 124/183$). Age ($P = 0.723$) and sex ($P = 0.875$) of the children did not differ significantly between participants and non-participants. In total, 71.8% ($n = 89/124$) of the participating patients fulfilled the DSM-5 criteria for ARIFD and were included in the present study. In total, 85 mothers and 62 fathers were included. Eighty-three mothers completed the TAAQOL and 39 the DT-P. Sociodemographics and characteristics of included ARIFD patients and parents are shown in Table 1. Fifty-seven percent of ARIFD patients (58.4% female, median age 1.9 years) required tube feeding and 91% had underlying diseases such as diseases of the digestive system (53.9%), respiratory system (48.3%), and prematurity (33.7%).

**Parental Health-Related Quality of Life**

The HRQOL of mothers and fathers of ARIFD patients compared to healthy controls is shown in Table 2. Age of parents did not differ between these groups ($P = 0.468$). Most of the HRQOL domains did not differ significantly. Only mothers of children with ARIFD had significantly higher HRQOL regarding depressive emotions ($P = 0.005$).

**Parental Distress**

Tables 3 and 4 show distress of mothers and fathers of ARIFD patients respectively compared to healthy and chronically ill controls. Age ($P = 0.972$) and sex ($P = 0.041$) of children with ARIFD did not differ from the selected controls ($\leq 6$ years), neither did the age of the parents ($P = 0.352$).

**Overall Distress Score**

The median overall distress scores and clinical distress scores of mothers (Table 3) and fathers (Table 4) of ARIFD patients did not differ significantly from those of healthy and chronically ill controls.

**Problem Domain Scores**

Mothers of ARIFD patients had significantly higher scores regarding the domains parenting problems in children <2 years of age ($P < 0.001$) and cognitive problems ($P = 0.003$) compared to mothers of healthy controls. Compared to mothers of chronically ill controls, mothers of ARIFD patients had a significantly higher score on the domain parenting problems in children <2 years of age ($P = 0.001$).

Fathers of ARIFD patients had a significantly higher score regarding the domain parenting problems in children <2 years of age compared to both fathers of healthy ($P < 0.001$) and chronically ill children ($P = 0.001$).

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**TABLE 1. Sociodemographics and characteristics of ARIFD patients ($n = 89$) and parents**

| Domain                                      | ARIFD patients | Parents of ARIFD patients |
|---------------------------------------------|----------------|---------------------------|
| Age                                         | Median (IQR)   | Median (IQR)              |
| Age                                         | 34.1 (30.7–39.1) | 37.1 (33.9–39.8)          |
| Country of birth, the Netherlands            | n (%)          | n (%)                     |
| Country of birth, the Netherlands            | 73 (91.3)      | 56 (93.3)                 |
| Married/living together                      | 79 (98.8)      | 60 (100)                  |
| Education                                    | Median (IQR)   | Median (IQR)              |
| Low§                                        | 6 (7.5)        | 1 (1.7)                   |
| Middle§                                      | 32 (40.0)      | 26 (43.3)                 |
| High§                                       | 42 (52.6)      | 33 (55.0)                 |
| Employment status                           | Median (IQR)   | Median (IQR)              |
| Median (IQR)                                | 58 (72.5)      | 55 (91.7)                 |

n: n missing data. ARIFD = avoidant restrictive food intake disorder; IQR = interquartile range; SD = standard deviation.

§ In case of prematurity (gestational age was <37 wk), age was corrected up to the corrected age of 24 mo.

Upper secondary education.

Primary education, lower vocational education, lower or middle general secondary education.

Higher vocational education.
### TABLE 2. HRQOL (measured by the TAAQOL) of mothers and fathers of children with ARFID compared to mothers and fathers of healthy children

| HRQOL                        | Mothers of ARFID patients | Mothers of healthy children | P  | r   |
|------------------------------|----------------------------|-----------------------------|----|-----|
|                              | n  | Median (IQR)          | n  | Median (IQR)          |     |     |
| Daily activities             | 83 | 100 (75–100)           | 66 | 100 (79.7–100)         | 0.713| –0.04|
| Aggressive emotions          | 83 | 88.9 (77.8–100)        | 65 | 88.9 (77.8–100)        | 0.232| –0.13|
| Cognitive functioning        | 83 | 93.8 (62.5–100)        | 67 | 87.5 (62.5–100)        | 0.422| –0.09|
| Fine motor functioning       | 83 | 100 (100–100)          | 67 | 100 (100–100)          | 0.036| –0.22|
| Gross motor functioning *    | 83 | 100 (68.8–100)         | 67 | 100 (68.8–100)         |     |     |
| Pain                         | 83 | 87.5 (75–100)          | 67 | 75 (62.5–87.5)         | <0.001| –0.41|
| Positive emotions            | 83 | 66.7 (50–83.3)         | 67 | 66.7 (66.7–83.3)       | 0.398| –0.09|
| Sexuality                    | 83 | 100 (62.5–100)         | 60 | 100 (75–100)           | 0.866| –0.02|
| Sleep                        | 83 | 75 (50–100)            | 67 | 81.3 (56.3–93.8)       | 0.376| –0.10|
| Social functioning           | 83 | 100 (75–100)           | 66 | 87.5 (75–100)          | 0.516| –0.07|
| Depressive emotions          | 83 | 75 (50–91.7)           | 67 | 83.3 (75–91.7)         | 0.020| –0.25|
| Vitality                     | 83 | 58.3 (33.3–75)         | 67 | 66.7 (50–83.3)         | 0.038| –0.23|

Fathers of ARFID patients

| HRQOL                        | Mothers of ARFID patients | Mothers of healthy children | P  | r   |
|------------------------------|----------------------------|-----------------------------|----|-----|
|                              | n  | Median (IQR)          | n  | Median (IQR)          |     |     |
| Daily activities             | 61 | 100 (75–100)           | 17 | 93.8 (87.5–100)       | 0.610| –0.06|
| Aggressive emotions          | 61 | 100 (88.9–100)         | 17 | 100 (88.9–100)        | 0.514| –0.08|
| Cognitive functioning        | 61 | 100 (81.3–100)         | 16 | 100 (89.1–100)        | 0.261| –0.15|
| Fine motor functioning       | 61 | 100 (100–100)          | 17 | 100 (100–100)         |     |     |
| Gross motor functioning *    | 61 | 100 (100–100)          | 17 | 100 (100–100)         | 0.689| –0.05|
| Pain                         | 61 | 87.5 (75–100)          | 17 | 81.3 (65.6–100)       | 0.378| –0.11|
| Positive emotions            | 61 | 66.7 (66.7–83.3)       | 17 | 66.7 (66.7–79.2)      | 0.731| –0.05|
| Sexuality                    | 61 | 100 (75–100)           | 17 | 100 (25–100)          | 0.579| –0.07|
| Sleep                        | 61 | 81.3 (53.1–100)        | 17 | 81.3 (62.5–100)       | 0.367| –0.12|
| Social functioning           | 61 | 87.5 (75–100)          | 17 | 81.3 (75–100)         | 0.963| –0.01|
| Depressive emotions          | 61 | 83.3 (66.7–91.7)       | 17 | 91.7 (83.3–100)       | 0.005| –0.36|
| Vitality                     | 61 | 66.7 (50–83.3)         | 17 | 75 (62.5–83.3)        | 0.192| –0.17|

Higher scores indicate a higher HRQOL. P-values were measured by Mann-Whitney U tests. Significance level was set at P < 0.01. ARFID = avoidant restrictive food intake disorder; HRQOL = health-related quality of life; IQR = interquartile range; TAAQOL = TAAQOL Questionnaire for Adult’s HRQOL.

*Since Cronbach’s alpha value in this problem domain was unacceptable, the domain score is not shown.

### Everyday Problem Scores

Mothers of children with ARFID more often reported problems with memory (P = 0.004), feeding the child <2 years of age (P = 0.000), development of the child <2 years of age (P = 0.002) and following advice about treatment/medication for the child <2 years of age (P = 0.001) compared to mothers of healthy controls. Less often did they show problems with finance/insurances (P = 0.005). Compared to mothers of chronically ill patients, mothers of ARFID patients experienced problems in feeding the child (P = 0.000) more often. Less often they did have problems in dealing with (ex)partner (P = 0.007).

Fathers of ARFID patients reported problems with feeding the child <2 years of age (P = 0.000), development of the child <2 years of age (P = 0.003) more often compared to fathers of healthy controls. Compared to fathers of chronically ill patients, fathers of ARFID patients reported problems with feeding the child <2 years of age (P = 0.000) more often.

### Additional Questions

Both mothers and fathers reported a lack of understanding from their environment (P < 0.001 and P = 0.003, respectively) and wanted to talk to a professional about their situation (P < 0.001 and P < 0.001, respectively) more often compared to mothers and fathers of healthy controls. Mothers of ARFID patients less often experienced enough support from the environment (P < 0.001) compared to mothers of healthy controls. Compared to mothers of chronically ill children, less often did they have a chronic disease that they would like to talk to a professional about their situation more often compared to parents of healthy children.

### Health-Related Quality of Life

We expected a lower HRQOL in parents of children with ARFID and healthy controls regarding parental HRQOL on 11 of 12 domains; however, mothers of children with ARFID reported a significantly higher HRQOL regarding pain and fathers of children with ARFID reported a significantly lower HRQOL regarding depressive emotions. No differences were found in total and clinical distress scores, but several domain scores and everyday problems differed significantly between parents of children with ARFID and healthy/chronically ill children. Furthermore, parents of children with ARFID reported that people react with a lack of understanding and that they would like to talk to a professional about their situation more often compared to parents of healthy children.

DISCUSSION

This study found no differences between parents of children with ARFID and healthy controls regarding parental HRQOL on 11 of 12 domains; however, mothers of children with ARFID reported a significantly higher HRQOL regarding pain and fathers of children with ARFID reported a significantly lower HRQOL regarding depressive emotions. No differences were found in total and clinical distress scores, but several domain scores and everyday problems differed significantly between parents of children with ARFID and healthy/chronically ill children. Furthermore, parents of children with ARFID reported that people react with a lack of understanding and that they would like to talk to a professional about their situation more often compared to parents of healthy children.
|                          | Mothers of ARFID patients | Mothers of healthy children | Mothers of chronically ill children |
|--------------------------|----------------------------|-----------------------------|-----------------------------------|
|                          | (n = 71)                   | (n = 329)                   | (n = 56)                           |
| Overall distress score   | 4 (2–6)                    | 3 (1–6)                     | 4 (2–7)                            |
| Clinical                 | 39 (54.9)                  | 139 (42.2%)                 | 30 (53.6)                         |
| Total problem scores     | 5 (2–10)                   | 5 (2–9)                     | 6 (2–12)                           |
| Total with < 2 y parenting | 37 7 (4.5–12)              | 107 6 (3–11)                | 18 9 (2–11.5)                     |
| Total with > 2 y parenting | 34 4 (2–10)               | 222 5 (2–9)                 | 38 6 (2–13.5)                     |
| Practical problems       | 1 (0–2)                    | 1 (0–2)                     | 2 (0–3)                            |
| Housing                  | 2 (2.8)                    | 21 (6.4)                    | 0 (0–1)                            |
| Work/study               | 14 (19.7)                  | 80 (24.3)                   | 13 (23.2)                         |
| Finances/insurance       | 3 (4.2)                    | 56 (17.0)                   | 10 (17.9)                         |
| Housekeeping             | 17 (25.9)                  | 86 (26.1)                   | 25 (44.6)                         |
| Transport                | 2 (2.8)                    | 13 (4.0)                    | 8 (14.3)                           |
| Child care/child supervision | 14 (19.7)               | 48 (14.6)                   | 18 (32.1)                         |
| Leisure activities/relaxing | 26 (36.6)               | 83 (25.2)                   | 25 (44.6)                         |
| Social problems*         | 4 (5.6)                    | 42 (12.8)                   | 13 (23.2)                         |
| Dealing with (ex)partner | 6 (8.5)                    | 38 (11.6)                   | 10 (17.9)                         |
| Dealing with friends     | 2 (2.8)                    | 15 (4.6)                    | 4 (7.1)                            |
| Interacting with your child (ren) | 12 (16.9) | 32 (9.7)                    | 12 (21.4)                         |
| Emotional problems       | 1 (0–3)                    | 0.874–0.01                  | 1.5 (0–4)                         |
| Controlling emotions     | 16 (22.5)                  | 88 (26.7)                   | 15 (26.8)                         |
| Self-confidence          | 15 (21.1)                  | 77 (23.4)                   | 15 (26.8)                         |
| Fears                    | 13 (18.3)                  | 37 (11.2)                   | 8 (14.3)                           |
| Depression               | 21 (29.6)                  | 119 (36.2)                  | 20 (35.7)                         |
| Feeling tense or nervous | 34 (47.9)                  | 127 (38.6)                  | 24 (42.9)                         |
| Loneliness               | 9 (12.7)                   | 29 (8.8)                    | 12 (21.4)                         |
| Feelings of guilt        | 11 (15.5)                  | 67 (20.4)                   | 10 (17.9)                         |
| Use of substances (eg, alcohol, drugs and/or medication) | 1 (1.4) | 8 (2.4) | 3 (5.4) | 0.320 0.25 0.03–2.50 |
| Intrusive/recurrent thoughts about a specific event | 17 (23.9) | 61 (18.5) | 14 (25.0) | 1.000 0.94 0.42–2.13 |
| Physical problems        | 2 (1–3)                    | 0.410–0.04                  | 2 (1–4)                           |
| Eating                   | 10 (14.1)                  | 36 (10.9)                   | 9 (16.1)                           |
| Weight                   | 15 (21.1)                  | 84 (25.5)                   | 17 (30.4)                         |
| Sleep                    | 27 (38.0)                  | 100 (30.4)                  | 17 (30.4)                         |
| Fatigue                  | 51 (71.8)                  | 213 (64.7)                  | 37 (66.1)                         |
| Out of shape/condition   | 19 (26.8)                  | 72 (21.9)                   | 21 (37.5)                         |
| Pain                     | 12 (16.9)                  | 82 (24.9)                   | 19 (33.9)                         |
| Sexuality                | 12 (16.9)                  | 38 (11.6)                   | 9 (16.1)                           |
| Cognitive problems       | 0 (0–2)                    | 0 (0–1)                     | 0 (0–2)                           |
| Concentration            | 24 (33.8)                  | 62 (18.8)                   | 19 (33.9)                         |
| Memory                   | 31 (43.7)                  | 85 (25.8)                   | 22 (39.3)                         |
| Parenting problems < 2 y | n = 37                     | 1 (1–3)                     | n = 18                            |
| Making contact with our child | 3 (8.1) | 2 (1.9) | 1 (5.6) | 1.000 1.50 0.15–15.52 |
| Problem Area                                      | Mothers of ARFID patients | Mothers of healthy children | Mothers of chronically ill children |
|--------------------------------------------------|---------------------------|-----------------------------|-------------------------------------|
| n (%) Median (IQR)                               | n (%) Median (IQR)        | n (%) Median (IQR)          | n (%) Median (IQR)                  |
| Nursing your child                               | 3 (8.1) 2 (1.9)           | 0.107 4.63 0.74–28.89       | 1 (5.6) 1.000 1.50–15.52            |
| Feeding your child                               | 34 (91.1) 18 (16.8)       | 0.000 56.04 15.51–202.46    | 1 (5.6) 0.000 192.67 18.62–1993.84  |
| Development of your child                        | 10 (27.0) 7 (6.5)         | 0.002 5.29 1.84–15.20       | 2 (11.1) 0.298 2.96 0.58–15.26      |
| Following advice about treatment/giving medication| 8 (21.6) 3 (2.8)          | 0.001 9.56 2.38–38.37       | 1 (5.6) 0.244 4.69 0.54–40.80       |
| Sleep of the child                               | 8 (21.6) 26 (24.3)        | 0.825 0.86 0.35–2.11        | 6 (33.3) 0.510 0.55 0.16–1.93       |
| Behaviour/crying of the child                    | 8 (21.6) 18 (16.8)        | 0.620 1.36 0.54–3.47        | 3 (16.7) 1.000 1.38 0.32–5.98       |
| Parenting problems >2 years                      | n = 34                    | n = 222 0 (0–0)             | n = 38 0 (0–1)                       |
| Dealing with your child                          | 9 (26.5) 30 (13.5)        | 0.070 2.30 0.98–5.41        | 8 (21.1) 0.782 1.35 0.45–4.02       |
| Dealing with the feelings of your child           | 7 (20.6) 17 (7.7)         | 0.025 3.17 1.19–8.23        | 5 (13.2) 0.530 1.71 0.49–6.00       |
| Talking about the disease/ consequences with your child | 1 (2.9) 4 (1.8)       | 0.513 1.65 0.18–15.23       | 3 (7.9) 0.617 0.35 0.04–3.57       |
| Independence of your child                       | 3 (8.8) 11 (5.0)          | 0.408 1.86 0.49–7.03        | 8 (21.1) 0.197 0.36 0.09–1.50       |
| Following advice about treatment/giving medication| 4 (11.8) 7 (3.2)         | 0.044 4.10 1.13–14.83       | 2 (5.3) 0.412 2.40 0.41–14.02       |
| Additional questions                             |                          |                            |                                     |
| Enough support from environment                  | 54 (76.1) 307 (93.3)      | 0.000 0.23 0.11–0.46        | 44 (78.6) 0.833 0.87 0.37–2.01      |
| People react with a lack of understanding         | 27 (38.0) 41 (12.5)       | 0.000 4.31 2.41–7.70        | 12 (21.4) 0.054 2.25 1.01–5.00      |
| How do you go along with medical stuff           | 54 (76.1) n/a             | n/a                         | n/a                                 |
| Do you have a (chronic) illness yourself         | 10 (14.1) 63 (19.1)       | 0.397 0.69 0.34–1.43        | 21 (37.5) 0.003 0.27 0.12–0.65      |
| Would like to talk to a professional about situation | 0.000                   |                             | 0.013                               |
| Yes                                              | 14 (19.7) 14 (4.3)        |                             |                                     |
| Maybe                                            | 15 (21.1) 40 (12.2)       |                             |                                     |

*P*-values were measured by Mann-Whitney *U* tests. Significance level was set at *P* = 0.01. ARFID = avoidant restrictive food intake disorder; CI = confidence interval; DT-P = distress thermometer for parents; IQR = interquartile range; OR = odds ratio.

*Since Cronbach’s alpha value in this problem domain was unacceptable, the domain score is not shown. Mann-Whitney *U* tests with effect sizes *r* for continuous variables, Fischer Exact tests with OR and 95% CI.*
| Overall distress score | Fathers of ARFID patients | Fathers of healthy children | Fathers of chronically ill children |
|------------------------|---------------------------|----------------------------|----------------------------------|
| (n = 39) | Median (IQR) | Median (IQR) | P | OR | 95% CI |
| n (%) | 3 (0–6) | 2 (1–4) | 0.254 | 0.07 | 2.5 (1–6.75) | 0.572 | 0.07 |
| Clinical | 17 (43.6) | 65 (29.3) | 0.874 | 0.49–1.71 | 9 (45.0) | 0.172 | 0.46 |
| Total problem scores | 2 (0–6) | 65 (29.3) | 0.418 | 0.05 | 4.5 (1–11.5) | 0.047 | 0.26 |
| Total with < 2 y parenting | 21 | 4 (1.5–12.5) | 87 | 3 (1–7) | 0.171 | 0.13 | 10 | 8 (2–15.5) | 0.289 | 0.19 |
| Total with > 2 y parenting | 18 | 0.5 (0–5.25) | 135 | 2 (1–6) | 0.104 | 0.13 | 10 | 2.5 (1–7.25) | 0.217 | 0.24 |
| Practical problems | 0 (0–1) | 0 (0–2) | 0.472 | 0.04 | 3 (15.0) | 0.398 | 0.47 |
| Housing | 3 (7.7) | 9 (4.1) | 0.397 | 0.19 | 0.51–1.74 | 3 (15.0) | 0.398 | 0.47 |
| Work/study | 5 (12.8) | 64 (28.8) | 0.048 | 0.36 | 0.14–0.97 | 4 (20.0) | 0.471 | 0.59 |
| Finances/insurance | 1 (2.6) | 37 (16.7) | 0.024 | 0.13 | 0.02–0.99 | 5 (25.0) | 0.014 | 0.08 |
| Housekeeping | 7 (17.9) | 31 (14.0) | 0.470 | 1.35 | 0.55–3.32 | 7 (35.0) | 0.198 | 0.41 |
| Transport | 1 (2.6) | 9 (4.1) | 1.000 | 0.62 | 0.08–5.06 | 3 (15.0) | 0.108 | 0.15 |
| Child care/child supervision | 7 (17.9) | 18 (8.1) | 0.073 | 2.48 | 0.96–6.61 | 7 (35.0) | 0.198 | 0.41 |
| Leisure activities/relaxing | 10 (25.6) | 43 (19.4) | 0.390 | 1.44 | 0.65–3.17 | 8 (40.0) | 0.371 | 0.52 |
| Social problems | 0 (0–0) | 0 (0–0) | 0.304 | 0.06 | 1.5 (0–3.75) | 0.240 | 0.15 |
| Dealing with (ex)partner | 3 (7.7) | 29 (13.1) | 0.437 | 0.56 | 0.16–1.92 | 4 (20.0) | 0.213 | 0.33 |
| Dealing with family | 2 (5.1) | 14 (6.3) | 1.000 | 0.80 | 0.18–3.68 | 3 (15.0) | 0.325 | 0.31 |
| Dealing with friends | 3 (7.7) | 2 (0.9) | 0.025 | 9.17 | 1.48–56.78 | 1 (5.0) | 1.000 | 1.58 |
| Emotional problems | 1 (0–2) | 0 (0–2) | 0.304 | 0.06 | 1.5 (0–3.75) | 0.240 | 0.15 |
| Controlling emotions | 5 (12.8) | 20 (9.0) | 0.553 | 1.49 | 0.52–4.22 | 7 (35.0) | 0.084 | 0.27 |
| Self-confidence | 6 (15.4) | 25 (11.3) | 0.429 | 1.43 | 0.55–3.76 | 3 (15.0) | 1.000 | 1.03 |
| Fears | 4 (10.3) | 15 (6.8) | 0.500 | 1.56 | 0.50–5.03 | 1 (5.0) | 0.653 | 2.17 |
| Depression | 11 (28.2) | 52 (23.4) | 0.545 | 1.28 | 0.60–2.76 | 9 (45.0) | 0.780 | 0.76 |
| Feeling tense or nervous | 15 (38.5) | 62 (27.9) | 0.188 | 1.61 | 0.79–3.28 | 9 (45.0) | 0.780 | 0.76 |
| Loneliness | 1 (2.6) | 6 (2.7) | 1.000 | 0.95 | 0.11–8.09 | 1 (5.0) | 1.000 | 0.50 |
| Feelings of guilt | 5 (12.8) | 14 (6.3) | 0.176 | 2.19 | 0.74–6.46 | 5 (15.0) | 0.283 | 0.44 |
| Use of substances (eg, alcohol, drugs and/or medication) | 4 (10.3) | 9 (4.1) | 0.111 | 2.71 | 0.79–2.96 | 1 (5.0) | 0.653 | 2.17 |
| Intrusive/reoccurring thoughts about a specific event | 8 (20.5) | 24 (10.8) | 0.110 | 2.13 | 0.88–5.16 | 4 (20.0) | 1.000 | 1.03 |
| Physical problems | 1 (0–2) | 1 (0–2) | 0.304 | 0.13 | 0.030 | 0.13 | 1 (0–2) | 0.330 | 0.13 |
| Eating | 2 (5.1) | 16 (7.2) | 1.000 | 0.70 | 0.15–3.15 | 0 (0) | 0.544 | n/a |
| Weight | 1 (2.6) | 34 (15.3) | 0.038 | 0.15 | 0.02–1.10 | 4 (20.0) | 0.041 | 0.11 |
| Sleep | 10 (25.6) | 47 (21.2) | 0.532 | 1.28 | 0.58–2.82 | 4 (20.0) | 0.753 | 0.38 |
| Fatigue | 17 (43.6) | 111 (50.0) | 0.491 | 0.77 | 0.39–1.53 | 10 (50.0) | 0.423 | 0.77 |
| Out of shape/condition | 8 (20.5) | 41 (18.5) | 0.824 | 1.14 | 0.49–2.66 | 4 (20.0) | 1.000 | 0.37 |
| Pain | 5 (12.8) | 35 (15.8) | 0.811 | 0.79 | 0.29–2.15 | 6 (30.0) | 0.159 | 0.34 |
| Sexuality | 3 (7.7) | 20 (9.0) | 1.000 | 0.84 | 0.24–2.98 | 4 (20.0) | 0.213 | 0.33 |
| Cognitive problems | 0 (0–0) | 0 (0–0) | 0.775 | 0.02 | 0.300 | 0.15 |
| Concentration | 6 (15.4) | 24 (10.8) | 0.416 | 1.50 | 0.57–3.95 | 4 (20.0) | 0.721 | 0.73 |
| Memory | 4 (10.3) | 27 (12.2) | 1.000 | 0.83 | 0.27–2.51 | 4 (20.0) | 0.424 | 0.46 |
| Parenting problems < 2 y | n = 21 | 1 (1–2.5) | n = 87 | 0 (0–1) | 0.000 | 0.48 | 0.001 | 0.35 |
| Making contact with our child | 0 (0) | 1 (1.1) | 1.000 | n/a | n/a | 0 (0) | n/a | n/a |

**TABLE 4.** Distress (measured by the DT-P thermometer) in fathers of children with ARFID compared to fathers of healthy and chronically ill children.
| Topic                                      | Fathers of ARFID patients | Fathers of healthy children | Fathers of chronically ill children |
|--------------------------------------------|---------------------------|----------------------------|-----------------------------------|
| (n = 39)                                   | (n = 222)                 | (n = 20)                   |
| n (%) Median (IQR)                         | Median (IQR) P rOR 95% CI | Median (IQR) P rOR 95% CI  |
| Nursing your child                         | 1 (4.8) 3 (3.4)           | 1.000 1.40 0.14–14.18      | 0 (0.0) 1.000 n/a n/a            |
| Feeding your child                         | 19 (90.5) 8 (9.2)         | 0.000 93.81 18.41–478.05   | 1 (10.0) 0.000 85.50 6.82–1071.27|
| Development of your child                  | 6 (28.6) 4 (4.6)          | 0.003 8.30 2.09–32.98      | 1 (10.0) 0.379 3.60 0.37–34.94   |
| Following advice about treatment/         | 3 (14.3) 1 (1.1)          | 0.023 14.33 1.41–145.78    | 0 (0.0) 0.533 n/a n/a            |
| giving medication                          |                          |                            |                                   |
| Sleep of the child                         | 6 (28.6) 19 (21.8)        | 0.567 1.43 0.49–4.19       | 1 (10.0) 0.379 3.60 0.37–34.94   |
| Behaviour/crying of the child             | 3 (14.3) 15 (17.2)        | 1.000 0.80 0.21–3.06       | 2 (20.0) 1.000 0.67 0.09–4.80    |
| Parenting problems > 2 y                   | n = 18                    | n = 135                    | n = 10                            |
| Dealing with your child                    | 0 (0)                     | 12 (8.9) 0.362 n/a n/a    | 1 (10.0) 0.357 n/a n/a           |
| Dealing with the feelings of your child    | 3 (16.7) 10 (7.4)         | 0.183 2.50 0.62–10.11      | 0 (0.0) 0.533 n/a n/a            |
| Talking about the disease/                | n (0)                     | 2 (1.5) 1.000 n/a n/a     | 0 (0.0) n/a n/a n/a              |
| consequences with your child              |                          |                            |                                   |
| Independence of your child                | 0 (0)                     | 8 (5.9) 0.597 n/a n/a     | 0 (0.0) n/a n/a n/a              |
| Following advice about treatment/         | 0 (0)                     | 5 (3.7) 1.000 n/a n/a     | 0 (0.0) n/a n/a n/a              |
| giving medication                          |                          |                            |                                   |
| Additional questions                      |                          |                            |                                   |
| Enough support from environment           | 34 (87.2) 206 (92.8)      | 0.216 0.53 0.18–1.54       | 18 (90.0) 1.000 0.76 0.13–4.29   |
| People react with a lack of understanding  | 11 (28.2) 21 (9.5)        | 0.003 3.76 1.64–6.82       | 1 (5.0) 0.044 7.46 0.89–62.71    |
| How do you go along with medical          | n/a                       | n/a n/a n/a n/a           | n/a n/a n/a n/a                   |
| staff                                    |                          |                            |                                   |
| Do you have a (chronic) illness yourself  | 23 (59.0) 24 (10.8)       | 0.389 0.45 0.10–1.97       | 5 (25.0) 0.038 0.16 0.03–0.93    |
| Would like to talk to a professional      | 0.000                     | n/a n/a n/a n/a           | 0.000 n/a n/a n/a                 |
| about situation                           |                          |                            |                                   |
| Yes                                       | 7 (17.9) 5 (2.3)          |                            | 1 (5.0)                            |
| Maybe                                     | 7 (17.9) 22 (9.9)         |                            | 3 (15.0)                           |

P-values were measured by Mann-Whitney U tests. Significance level was set at P = 0.01. ARFID = avoidant restrictive food intake disorder; CI = confidence interval; DT-P = distress thermometer for parents; IQR = interquartile range; OR = odds ratio.

Since Cronbach’s alpha value in this problem domain was unacceptable (indicating the items do not measure the same construct), the domain score is not shown. Mann-Whitney U tests with effect sizes r for continuous variables, Fischer Exact tests with OR and 95% CI.
it may be difficult to leave feeding moments to others and parents may feel hounded consequently (22). Pediatric feeding disorders have been associated with parental psychosocial problems before (23,24). Many studies found associations between maternal depressive symptoms and pediatric feeding disorders (23,25–28). Maternal depressive symptoms are associated with nonresponsive and controlling feeding styles (such as persuading the child to eat by promising incentives, verbal pressure or physical force) (24,29,30), which can lead to negative associations, avoidance and severe pediatric feeding problems (31–33). Surprisingly, we only found a trend towards lower HRQOL of depressive emotions in mothers of children with ARFID. HRQOL regarding depressive emotions of fathers of children with ARFID was lower compared to fathers of healthy children. Other studies also found that parental depressive symptoms are associated with feeding problems in children (34,35). We hypothesize this could be due to the same mechanism described in mothers.

Distress

Surprisingly, total and clinical distress scores in parents of children with ARFID did not differ between those of healthy and chronically ill children. In a recently published large study including children with pediatric feeding disorders ages 2.9 years, parental stress was assessed by the self-report Parent Stress Index-Short Form. The total parental stress scale in parents of children with feeding disorders was significantly higher (36). Mothers of children with ARFID reported problems with finance/insurances less often compared to mothers of healthy children. Since financial problems are associated with parental emotional distress, this could be an explanation why no differences were found in total and clinical distress scores (37).

We found higher distress scores in the cognitive domain and the contributing everyday problem with memory in mothers of children with ARFID. Chronic stress is known to be related to problems with memory (38). Maternal memory problems may be part of maternal psychological/psychiatric disorders such as attention deficit disorder and posttraumatic stress disorder, which are known risk factors for pediatric feeding disorders (39,40).

Distress scores of parents of ARFID patients in the domain parenting <2 years were higher compared to parents of healthy and chronically ill patients. Everyday problems “feeding your child,” “development of your child,” and “following advice about treatment/medication” from this domain were more common in parents of ARFID patients compared to healthy/chronically ill patients. Explanations for problems with the development of the child is that feeding problems can lead to malnutrition and micronutrient deficiency, which can lead to impaired cognitive development in children and that children with developmental delays are at risk for feeding disorders (41,42).

Strikingly, parents of ARFID patients reported a lack of understanding and experienced less support from other people compared to parents of healthy children. This corresponds with our experience from clinical practice that they feel misunderstood and receive unhelpful advice from others, which can feel like an attack on their quality as a caregiver.

In general, we expected a lower HRQOL and more distress in parents of children with ARFID. It could be that their HRQOL is not so much affected and that they do not experience distress besides the feeding moments. In our experience, parents often visit our Center with medical concerns. Therefore, they might as well underreport parental distress factors to avoid thrifting away the focus from the medical concerns. Furthermore, the questionnaires might not be sensitive enough in detecting all relevant psychosocial problems in parents of children with ARFID.

Pediatric feeding comprises a complex interaction of a myriad of factors, including biological, sensorimotor skills, psychosocial, sociocultural, family and environmental factors (43–45). Some authors even suggest to approach feeding disorders as a multidimensional relational disorder (46). Although caregivers play such an important role, paternal context has been mostly disregarded (22). To our knowledge, this is the first study to assess HRQOL and distress in both parents of children with ARFID systematically. Although we explicitly invited all parents to participate, more mothers completed questionnaires, which might imply that mothers are still more involved in the primary care for children (36).

Limitations

The main limitation of our study is that the age and sex of the healthy children of the HRQOL norm group were unavailable. Furthermore, chronic disease in the distress norm group was parent-reported and might have been interpreted differently by a clinician. Parents completed the DT-P non-anonymously (enabling to discuss results during psychological consultation), whereas the normative group completed the questionnaires anonymously. Due to the small sample size, the study might have been underpowered to detect associations with small effect sizes. Selection bias may have occurred if completing the questionnaires took too much effort for the more distressed parents with lower HRQOL, leading to underrepresentation of distress and overestimation of HRQOL in the results.

Future Studies

It is important to note that correlation does not imply causation. Pediatric feeding problems may impact the psychosocial status of the parents, but the psychosocial status of the parents might as well influence pediatric feeding. Future prospective studies, therefore, should focus on the causal relationship between parental distress/HIQOL and ARFID and should include fathers as well. Further assessment regarding questionnaires detecting relevant psychosocial problems in parents of children with ARFID could be done. Additionally, the relationship between parental psychopathology and ARFID would be interesting to investigate. Furthermore, a core outcome set for ARFID developed by clinicians as well as (parents of) patients resulting in a standardized set of relevant outcomes and measures that should be used in clinical studies, would allow to compare results of different studies.

CONCLUSION

The HRQOL and distress of parents of children with ARFID were comparable to reference groups. Since parents of children with ARFID, however, perceive a lack of understanding and support from the environment professionals can assess interest in peer support and refer to parents to national patient’s organizations. Furthermore, it is important to offer professional support, since parents indicated that they would like to talk to a professional about their situation. Screening for parental psychosocial problems could be beneficial to detect problems in specific domains, such as depressive emotions, cognitive problems and parenting problems.

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