Parent-Child-Agreement on Health-Related Quality of Life and its Determinants in Patients Born with Esophageal Atresia – A Swedish-German Cross-Sectional Study

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Abstract

Background The aim was to compare parent and child reported health-related quality of life (HRQOL) of children born with esophageal atresia (EA) and to determine factors that affect the level of parent-child agreement.

Methods We included 63 parent-child dyads of children born with EA aged 8-18 from Germany and Sweden. The generic PedsQL questionnaire and the condition-specific EA QOL questionnaire were used to assess children's HRQOL from parents- and child's perspective. The Peds QL Family Impact Module was used to assess parental HRQOL and Family Functioning.

Results On an individual level, intra-class correlation coefficients indicated strong levels of parent-child agreement (.61-.97). At the group level, the analyses showed no significant differences between the responses of parents and children. In cases where disagreement occurred, parents were more likely to rate generic HRQOL lower than the children (19-35%) and condition-specific HRQOL higher than the children (17-33%). Findings of the multiple regression analysis showed that the country (Germany vs. Sweden) was found to be a significant predictor of parent-child agreement in generic HRQOL and parental HRQOL to be a significant predictor of parent-child agreement in condition-specific HRQOL.

Conclusion The parent-child agreement is mostly good, suggesting that parent-reports are a reliable source of information. However, discrepancies may occur and cannot be explained by the child's age and gender, nor severity of the disease or Family Functioning, but solely by country respectively parental HRQOL. Clinicians should therefore not only observe the pediatric patient but also the parents.

Key Messages

- Agreement between child and parent reports when rating EA children's HRQOL is mainly good, suggesting that parent-reports are a reliable source of information.
- In EA children, the directional discrepancy in parent-report vs. child-report shows the tendency of parents underrating their children's generic HRQOL, while overrating their condition-specific HRQOL.
- In condition-specific HRQOL assessment of EA children, disagreement between child- and parent-report may be explained by parents' HRQOL and in generic HRQOL assessment disagreement may be explained by country origin.

Background

Esophageal atresia (EA) is a rare malformation occurring in 2.4/10 000 births. While survival rates today exceed 90%, long-term esophageal and respiratory sequelae remain common (Dellenmark-Blom et al., 2018). Although neonatal survival is no longer a problem, there is scarce data about the patients' long-term outcomes, including health-related quality of life (HRQOL) (Legrand et al., 2012).

As a multidimensional construct, HRQOL considers the impact of such rare chronic conditions on overall health and daily living (Bullinger & Quitmann, 2014), including physical, emotional, and social functioning. It can be measured using generic instruments and condition-specific instruments (Brütt et al., 2009).

In HRQOL related studies of pediatric patients, parents are often asked to report on their children's HRQOL (Dickinson et al., 2007; Silva, Crespo, Carona, Bullinger, & Canavarro, 2015). Parent proxy-reports are used, when the child is very young, or cannot complete the self-report due to illness or cognitive impairments (Silva, Crespo, Carona, Bullinger, & Canavarro, 2015; von Essen, 2004). A child self-report should be accompanied by a parent-report (Silva, Crespo, Carona, Bullinger, & Canavarro, 2015) because although parents often lack first-hand information, their reports provide important complementary information about children's HRQOL (Eiser & Varni, 2013).

Only one study examined differences in children's self-reports and parent proxy-reports in children with EA, which showed satisfactory agreement, but with varying results depending on the investigated HRQoL domain (Dellenmark-Blom et al., 2018; Flieder et al., 2018). However studies have shown that parents of children with chronic diseases tend to score their children's HRQOL lower than the children (Sheffler, Hanley, Bagley, Molitor, & James, 2009), the reliability of using parent proxy-reports has

been questioned (Panepinto, Hoffmann, & Pajewski, 2010). There remains a need for further research on the factors affecting the level of (dis-)agreement between parent proxy-reports and child self-reports (Quitmann, Rohenkohl, Sommer, Bullinger, & Silva, 2016). Identifying these factors will help to gain a deeper understanding of the difference in perspectives of HRQOL (White-Koning et al., 2007), which in turn may provide useful information for guiding clinical decision-making.

Studies have examined the level of agreement between parent proxy-reports and child self-reports of HRQOL for children with chronic diseases but showed inconsistent results (Quitmann et al., 2016; Silva, Crespo, Carona, Bullinger, & Canavarro, 2015; Upton, Lawford, & Eiser, 2008). It should be considered that the severity level of the child's disease may affect the level of parent-child agreement (Petsios et al., 2011). Parental factors of well-being (Witt et al., 2018) and stress (Bastiaansen, Koot, & Ferdinand, 2005) are associated with parental perceptions of their child's HRQOL. At the same time, the severity level of the child's disease is negatively associated with parental HRQOL (Witt et al., 2018), suggesting that the parental well-being may affect how parents rate their children's HRQOL and thus impacts on the level of parent-child-agreement.

Since there is currently no consistent knowledge of the direction of the discrepancies and its determinants, we aimed to investigate the directional disagreement and level of agreement between child and parent reports of generic and condition-specific HRQOL in 8 to 17 year-olds born with EA and the predictors (age, gender and EA severity level) of the level of parent-child-agreement. We hypothesize that higher parental HRQOL and better family functioning have positive effects on the parent-child-agreement.

Methods

Participants & Setting

In order to increase the knowledge on parent-child agreement on HRQOL of children born with EA, we conducted this multicenter study at the Department of Pediatric Surgery, Queen Silvia Children's Hospital in Gothenburg, Sweden, the Centre of Pediatric Surgery, Hannover Medical School and the Bult Children's Hospital, Hannover, and the Department for Medical Psychology, University Medical Center Hamburg-Eppendorf, Germany (Dellenmark-Blom et al., 2018). The study was approved by the Ethical Review Boards of Gothenburg, Sweden (DNR 958 – 13) and Hannover, Germany (2936 – 2015). Eligible patients with EA Gross type A-E had been treated at the clinics and were considered for participation when they met the following inclusion criteria: (1) children were aged 8–17 years at assessment, (2) sufficient German/Swedish language proficiency. The invited families gave informed consent before study participation. The current analyses considered data from the fieldtest phase (Dellenmark-Blom et al., 2018), including 63 parent-child-dyads.

Measures

The PedsQL 4.0 ™ Generic Core Module

The PedsQL 4.0 ™ questionnaire is a generic instrument to assess children's HRQOL via child self-report and parent proxy-report (J. W. Varni, Seid, & Kurtin, 2001). The questionnaire includes 23 items, assessing Physical, Emotional, Social, and School Functioning, and can be calculated in the three summary scores (Total Scores, Physical Health Summary Score, and Psychosocial Health Summary Score) with higher scores indicating better HRQOL.

The EA-QOL© questionnaire

The condition-specific EA-QOL© questionnaire consists of a child- and parent-reported 24-item questionnaire for children aged 8–17 years (domains; Eating, Social Relationships, Body Perception, and Health & Well-being; Total Score) with higher scores representing better HRQOL (Dellenmark-Blom et al., 2017).

PedsQL™ Family Impact Module

The PedsQL™ Family Impact Module (PedsQL™ FIM) was used to assess parental HRQOL and Family Functioning (J.W. Varni, Seid, & Rode, 1999). Parental HRQOL (20 items) was calculated as the mean of items from the domains Physical, Emotional,

Social, and Cognitive Functioning. Family Functioning (8 items) was calculated from the subscales Daily Activities and Family Relationships. Higher scores represent higher HRQOL respectively better Family Functioning.

Statistical analysis

Parent-child-agreement on children's HRQOL was examined at individual and group level (Sneeuw, Sprangers, & Aaronson, 2002); using intra-class correlation coefficients [ICC] (Landis & Koch, 1977), respectively analyses of covariance for repeated measures (ANCOVA). We entered the perspectives (parent vs. child) as the within-subjects factors and children's age, children's gender and the severity level of EA (mild to moderate vs. severe) (Dellenmark-Blom et al., 2016) as covariates into the model.

We calculated parent-child-disagreement as absolute and directional discrepancies. Directional discrepancies we categorized into three groups ("parent-report < child-report", "agreement" and "parent-report > child-report") based on the threshold for important differences in quality of life (Norman, Sloan, & Wyrwich, 2003).

We performed multiple linear regression analyses in order to identify variables that predict the level of parent-child-agreement (absolute discrepancies). Children's gender, children's age, severity level of EA, parental HRQOL and family functioning as well as the country (Germany vs. Sweden) we entered as predictors.

Statistical analyses we performed using SPSS (version 23.0). Except for sociodemographic and clinical variables, we replaced missing values by the individual mean score for each variable, if missing data were random and less than 25% of the values (*EA-QOL©*) respectively random and less than 50% of the values (PedsQL 4.0 ™). We tested differences of sociodemographic and clinical variables between the both samples from Germany and Sweden using Student's t-Test or chi-square test when appropriate.

Results

Chi-square respectively student's t-test showed that sociodemographic and clinical characteristics of the children were similarly distributed between German and Swedish participants, except for associated anomalies and anorectal malformations. Parent characteristics showed differences between both countries (Table 1).

Table 1
Children's and parents sociodemographic characteristics of the German and Swedish sample

		German sample	Swedish sample			
		n = 23	n = 40			
Children's characteristics		**No. (%) or M (± SD)**	**No. (%) or M (± SD)**	**t or χ^2**	**df**	**p**
Child age (in years)		12.77 (2.98)	12.24 (3.46)	.62	61	.54
Child gender	Male	13 (56.5%)	22 (55%)	.78	1	.38
	Female	10 (43.5%)	18 (45%)			
EA severity level[a]	Mild/moderate	14 (60.9%)	18 (45%)	.02	1	.90
	Severe	9 (39.1%)	22 (55%)			
Primary anastomosis	Yes	21 (95.7%)	34 (85%)	1.68	1	.20
	No	2 (4.3%)	6 (15%)			
Associated anomalies	Yes	15 (65.2%)	29 (72.5%)	9.92	1	.01
	No	8 (34.8%)	11 (27.5%)			
Cardiac malformation	Yes	7 (30.4%)	18 (45%)	2.68	1	.10
	No	16 (69.6%)	22 (55%)			
Anorectal malformation	Yes	5 (21.7%)	5 (12.5%)	29.35	1	≤ .01
	No	18 (78.3%)	35 (87.5%)			
Birthweight (in grams)	≤ 2,500 g	7 (30.4%)[b]	15 (37.5%)[c]	1.85	1	.17
	> 2,500 g	9 (39.1%)	23 (57.5%)			
Gestational age (in weeks)		35.65 (3.71)[d]	36.44 (3.61)[e]	− .75	54	46
Parents characteristics		**No. (%) or Mean (± SD)**	**No. (%) or Mean (± SD)**	**t or χ^2**	**df**	**p**
Parent age		46.04 (7.11)	43.52 (6.33)[e]	1.39	60	.17
Parent gender	Male	1 (4.3%)	5 (12.5%)[e]	40.32	1	≤ .01
	Female	22 (95.7%)	34 (85%)			
Partnership	Single parent	1 (4.3%)[b]	9 (22.5%)[e]	27.56	1	≤ .01
	Co-habitant parent	21 (91.3%)	30 (75%)			
Employment status	Full/part time worker	18 (78.3%)[e]	36 (90%)[c]	111.97	6	≤ .01
	Parental/sick leave / unemployed	4 (17.4%)	2 (5%)			
Health status	Healthy	18 (78.3%)[e]	33 (82.5%)[e]	27.56	1	≤ .01
	Doctor diagnosis	4 (17.4%)	6 (15%)			

Abbreviations: M = Mean, SD = Standard deviation, t = t-value, χ^2 = chi-square, df = degrees of freedeom, p = p-value, EA = Esophageal atresia

	German sample	Swedish sample
[a] The severity of EA was divided into mild/moderate and severe according to predefined clinical criteria published elsewhere (Dellenmark-Blom et al., 2016)		
[b] 7 missings; [c] 2 missings; [d] 6 missings; [e] 1 missing		

At individual level, the ICCs indicated strong levels of agreement between parent- and child-reported children's HRQOL for both generic and condition-specific HRQOL (Table 2). At group level, the analyses of covariance for repeated measures showed no significant differences between the responses of parents and children concerning sociodemographic and clinical variables (Table 2).

Table 2
Inter-rater reliability: analyses of covariance for repeated measures (ANCOVA); country-specific

			parent-report		child-report		ICC [CI] [a]	ANCOVA for repeated measures [b]		
		Dyads n	M (SD)	α	M (SD)	α		F	p	N_p^2
GERMANY Peds QL	Physical	23	80.84 (20.63)	.86	84.65 (18.89)	.87	.87 [.69-.95]**	1.07	.39	.17
	Emotional	21	79.76 (23.05)	.90	86.43 (17.90)	.84	.82 [.55-.93]**	.09	.97	.02
	Social	23	80.87 (21.57)	.82	83.62 (16.84)	.78	.73 [.37-.89]**	1.28	.32	.19
	School	22	69.77 (19.67)	.78	75.68 (19.60)	.84	.83 [.60-.93]**	1.31	.31	.21
	Psychosocial	21	77.94 (17.46)	.90	82.46 (14.28)	.88	.79 [.47-.91]**	.86	.48	.16
	Total	21	79.32 (16.93)	.93	83.46 (14.08)	.92	.79 [.49-.92]**	.95	.44	.17
SWEDEN Peds QL	Physical	32	82.32 (23.04)	.91	84.67 (21.64)	.92	.95 [.90-.98]**	.87	.47	.10
	Emotional	31	81.67 (22.34)	.90	84.03 (20.47)	.87	.90 [.80-.95]**	.35	.79	.04
	Social	31	88.06 (17.64)	.85	88.55 (18.45)	.88	.90 [.79-.95]**	.21	.89	.03
	School	32	77.93 (20.66)	.85	80.31 (19.30)	.83	.95 [.90-.98]**	.14	.94	.02
	Psychosocial	31	82.92 (17.56)	.92	84.62 (16.79)	.93	.95 [.90-.98]**	.10	.96	.01
	Total	31	82.93 (18.55)	.95	84.91 (17.14)	.96	.97 [.93-.98]**	.15	.93	.02
GERMANY EA QOL	Eating	21	75.85 (20.36)	.81	72.64 (19.63)	.72	.89 [.72-.95]**	.55	.66	.11
	Body	20	84.50 (20.38)	.87	85.00 (21.64)	.86	.61 [-.01-.85]*	.07	.97	.02
	Social	21	79.65 (19.80)	.84	76.70 (21.21)	.79	.89 [.74-.96]**	.48	.70	.10
	Health	20	84.38 (16.78)	.64	80.94 (23.51)	.86	.78 [.46-.91]**	1.75	.21	.29
	Total	20	80.65 (16.69)	.92	78.25 (17.73)	.88	.90 [.76-.96]**	.63	.61	.13
SWEDEN EA QOL	Eating	36	73.61 (21.36)	.84	71.44 (19.75)	.76	.93 [.97-.97]**	.18	.91	.02
	Body	38	80.00 (19.59)	.82	80.39 (20.94)	.81	.95 [.91-.98]**	1.55	.22	.13
	Social	38	78.38 (16.71)	.74	74.53 (17.30)	.74	.86 [.73-.93]**	.07	.98	.01

		parent-report		child-report		ICC [CI] [a]	ANCOVA for repeated measures [b]		
	Dyads n	M (SD)	α	M (SD)	α		F	p	N_p^2
Health	38	81.41 (17.28)	.70	80.26 (19.25)	.76	.92 [.85–96]**	.61	.62	.06
Total	36	78.18 (15.32)	.91	76.09 (15.55)	.89	.93 [.86-.97]**	.23	.88	.02

Abbreviations: M = Mean, SD = Standard deviation, α = Cronbach's alpha, F = F-value, p = P-value, Ŋp2 = partial eta square

[a] Intraclass correlation coefficients reference values: ICC < .40 = poor agreement, ICC between .41 and .60 = moderate agreement, ICC between .61 and .80 = good agreement, ICC > .81 = excellent agreement (Landis & Koch, 1997). *≤.05; **≤.01

[b] analyses of covariance for repeated measures (ANCOVA), entering the informant (parent vs. child) as the within-subject factor and the sociodemographic and clinical variables (age, gender, EA severity level) as covariates

The examination of the directional discrepancies between parent- and child-reports demonstrated 47–69% agreement between children and parents in rating generic HRQOL and 42–71% in rating condition-specific HRQOL. In the remaining cases, disagreement occurred. In cases where disagreement occurred, parents were more likely to rate generic HRQOL lower than the children (19 vs. 35%). At the same time, parents were more likely to rate children's HRQOL higher than the children when using condition-specific measures (17 vs. 33%) (Fig. 1).

Findings of the multiple regression analysis using the absolute discrepancies for parent-child-agreement assessed with the generic instrument showed that the country (Germany vs. Sweden) was found to be a significant predictor for the level of parent-child-agreement. Parent-child dyads from Sweden showed better agreement than dyads from Germany. Using the condition-specific *EA-QOL©* questionnaire, results showed that parental HRQOL significantly predicted parent-child-agreement with higher parental HRQOL leading to increased parent-child-agreement on children's HRQOL. Neither child's age, child's gender nor EA severity level or Family Functioning showed any effect on the parent-child-agreement (Table 3). However, EA severity level was highly correlated with parental HRQOL (r=-.45, p = ≤ .01) and Family Functioning (r=-.39, p = ≤ .01).

Table 3
Regression analysis in EA patients and their parents

	Peds QL Total Score Difference				EA QOL Total Score Difference			
	β	t	p	[CI]	β	t	p	[CI]
Child gender[a]	− .12	− .91	.37	[.26–1.28]	.25	1.89	.06	[.01 − .39]
Child age	.02	.17	.87	[-.23 − .09]	.05	.44	.67	[-.02 − .02]
EA severity level[b]	.01	.10	.92	[-.02 − .03]	− .14	-1.09	.28	[-.21 − .06]
Parental HRQoL	.07	.34	.73	[-.01 - -01]	**− .73**	**-4.04**	**≤ .01**	**[-.02 - − .01]**
Family Functioning	− .31	-1.65	.10	[-.01 − .01]	.39	1.92	.07	[.01 − .40]
Country[c]	**− .43**	**-3.47**	**≤ .01**	**[-.43 - − .12]**	− .01	− .04	.97	[-.12 − .12]
Model Summary	**R^2=.20**				**R^2=.24**			
	F(6,58) = 2.44, p = .04				**F(6,61) = 3.16, p ≤ .01**			
Abbreviations: **β** = Beta, t = t-value, p = p-value, CI = confidence interval, R^2= R-square, F = F-value, EA = Esophageal atresia, HRQoL = Helath-related quality of life								
[a] Child gender: 0 = male, 1 = female								
[b] EA severity level: 0 = mild moderate, 1 = severe [23]								
[c] Country: 0 = Germany; 1 = Sweden								
*p ≤ .05 **p ≤ .01								

Discussion

This study investigated the parent-child-agreement of HRQOL in a sample of children and adolescents born with EA and determinants of discrepancies. Inconsistent with previous findings from the literature, which advocate moderate levels of agreement in pediatric HRQOL assessment, we have found strong levels of agreement between child- and parent-reported children's HRQOL on the individual level. While Quitmann et al. (2016) also report at least moderate to good ICC levels for children and adolescents with short stature and their parents, other studies reported only moderate levels of parent-child agreement in chronic diseases (Eiser & Varni, 2013; Silva, Crespo, Carona, Bullinger, & Canvarro, 2015).

Contrary to the results from previous studies (Quitmann et al., 2016; Silva et al., 2013), we found no differences in the agreement between generic and condition-specific instruments. According to our results, the agreement was good between children and parents (Dellenmark-Blom et al., 2018). However, a proportion of child-parent dyads also demonstrated directional differences in the rating children's generic and condition-specific HRQOL. Interestingly, the direction of the differences between parent- and child-reported children's HRQOL differed depending on the instrument used. Parents tended to underrate children's HRQOL using generic HRQOL measures. At the same time, the parents were more likely to score their children's HRQOL higher than the children when using the condition-specific tool, with the exception of the domain Body Perception. A possible explanation relates to the nature of the questions of the different measurement levels. A condition-specific instrument is more sensitive to clinical characteristics and raises issues of relevance for the patient group. The EA-QOL questionnaire was developed using the child experiences of primary importance, and parents' as complementary importance. Therefore, it might be easier for children to answer those questions (Dellenmark-Blom et al., 2017; Dellenmark-Blom et al., 2016).

Approximately half of the dyads showed an agreement – defined as differences of equal or less than half of the standard deviation of the score - between parent's perspective and children's perspective. When disagreement occurred, it was likely to be in the direction of parents underrating children's HRQOL using the generic HRQOL measurement. In all domains, with exception of the domain Social Functioning, the underrating was present in approximately one-third of parents. The same pattern was found in

other studies of children with chronic health conditions (Silva, Crespo, Carona, Bullinger, & Canavarro, 2015). This is consistent with previous research in children with chronic diseases, which found that parents tend to underestimate their child's HRQOL (Eiser & Varni, 2013; Levi & Drotar, 1999; Quitmann et al., 2016; Rohenkohl et al., 2015; Sheffler et al., 2009; Silva, Crespo, Carona, Bullinger, & Canavarro, 2015). Inconsistent with these findings, the direction of disagreement we found in our sample for the condition-specific HRQOL measurement was opposed. One-third of parents overrated their children's HRQOL, especially in the domains Eating, Social Relationships, and the Total Score. Here again, it might be that the questionnaire was more sensitive to the children's perspective than the parents' since it was developed primarily according to the children's experiences (Dellenmark-Blom et al., 2017; Dellenmark-Blom et al., 2016; Dellenmark-Blom et al., 2018).

On the domain level, our findings showed that the lowest rates of agreement were present in the generic domain Physical Functioning as well as in the condition-specific domain Eating. Both domains can be regarded as observable HRQOL dimensions for the parents that are not related to the internal experiences of the child. Using the PedsQL, the level of agreement was found to be highest in the domain Social Functioning, while using the EA-QOL, agreement was highest in the domain Health & Wellbeing." Thus, these results contrast with previous research, which describes better agreement for observable dimensions (Eiser & Varni, 2013; Patel, Lai, Goldfield, Sananes, & Longmuir, 2017; Rajmil, Rodriguez López, López-Aguilà, & Alonso, 2013). This underlines the importance of capturing the child-report using the EA-QOL questionnaire in clinical practice when monitoring and providing supportive interventions to the child's HRQOL.

In our analyses, sociodemographic (age, gender) and clinical variables (EA severity level) did not contribute to explaining a significant amount of variation of the extent of parent-child discrepancies. The level of agreement between parents and children instead seemed more strongly associated with familial and social factors (Quitmann et al., 2016; Silva, Crespo, Carona, Bullinger, & Canvarro, 2015; Van Roy, Groholt, Heyerdahl, & Clench-Aas, 2010). The current literature reported diverse findings on variables explaining parent-child-agreement. While some studies found higher levels of agreement for older children and explaining this by growing cognitive and communication skills (Annett, Bender, DuHamel, & Lapidus, 2003; Peetsold, Heij, Deurloo, & Gemke, 2010), other studies described higher agreement in younger children supporting the hypothesis that increased independence during puberty may limit the exchange between parents and children (April, Feldman, Platt, & Duffy, 2006; Rajmil et al., 2013). However, there were also studies, which did not find a significant influence of child age on the level of parent-child agreement (Patel et al., 2017). So far, there are no consistent results on the effect of child gender on the parent-child agreement, yet there are only a few studies considering this variable in statistical analysis (Eiser & Morse, 2001; Upton et al., 2008). The country of residence only explained the parent-child-agreement in our study when using the generic tool. It is difficult to identify a definite explanation for these results, but there might be different norms or traditions of how parents and children in different countries communicate about the child's health condition and its consequences in daily life (Matza et al., 2013). No other variables in the model contributed significantly to the extent of parent-child-agreement on generic HRQOL. The extent of disagreement of the condition-specific measurement was explained by the parent-reported parental HRQOL. The lower the parental HRQOL, the higher the level of parent-child discrepancies. In other studies parents with higher parental stress, especially depressive symptomatic reported significantly more limitations of their children than parents without these stressors (Kobayashi & Kamibeppu, 2011). This correlation has been more strongly demonstrated for mothers than for fathers (Davis, Davies, Waters, & Priest, 2008), and most parent-reports in this study were maternal perspective. However, Eiser and Varni (2013) add that the majority of research has taken into account primarily the maternal perspective, meaning that ratings of fathers or differences between mothers and fathers are only permitted to a limited extent. Indeed, it would be relevant for everyday clinical practice whether there are systematic differences in the assessment of HRQOL of children between mothers and fathers, which they have to be considered by the clinicians.

Limitations

Some limitations should be taken into account when interpreting the results of the present study. With regard to the sample, it should first be mentioned that due to the low response rate of fathers, the parents' perspective mostly consisted of mothers' reports. Even though this is not uncommon in pediatric health care studies and clinical practice (Upton et al., 2008), the results are therefore clearly biased towards the mothers' perspective and should be replicated in future studies using larger samples of fathers to determine whether there are differences between father and mother report. Although this sample size is relatively large

for a study in a rare disease such as EA the small sample size of 63 parent-child-dyads and the different sample composition in Germany and Sweden does not allow to draw meaningful conclusions. Moreover, our study sample seems fairly representative regarding gestational age at birth, primary esophageal repair and child gender, but the prevalence of associated anomalies seems slightly higher than in previous reports, especially cardio-vascular anomalies (Stoll, Alembik, Dott, & Roth, 2009).

Another limitation concerns the methods of data collection. Since families have filled out the questionnaires at home, a parental influence of the children's answers cannot be excluded even though parents have explicitly been asked not to disturbed their children's answers. The results of this study can be used as an indication towards future studies investigating parent-child agreement in pediatric patients with rare malformations such as EA.

Conclusion

Of importance to clinical practice and future research, the parent-child-agreement when rating EA children's HRQOL is mostly good, suggesting that parent reports are a reliable source of information. However, discrepancies may occur which may differ in the direction to child-report depending on generic or condition-specific measurement level. The parent-child-agreement on HRQOL cannot be explained by child's age and gender, nor severity of the disease or Family Functioning, but solely by country (generic HRQOL) respectively parental HRQOL (condition-specific HRQOL), a fact possibly due to the different health care systems in Germany and Sweden.

List Of Abbreviations

HRQOL - health-related quality of life

EA - Esophageal atresia

PedsQL - Pediatric Quality of Life Inventory

EA-QOL- The Esophageal-Atresia-Quality-of-life

Declarations

Ethics approval and consent to participate

The study was approved by the Ethical Review Boards of Gothenburg, Sweden (DNR 958-13) and Hannover, Germany (2936-2015).

Consent for publication

Not applicable.

Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Competing interests

No competing interests are declared.

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All authors have identified they have no financial relationships relevant to this study and its results to disclose.

Authors' contribution

MDB is the principal investigator of the study. MDB, JEC, KA, JD, MB, BU, SW and JHQ developed the study concept and the design. MDB developed the study materials and MDB, SW, JHQ, SK, JD, CD, LJ, VG, JEC and KA acquired the data. SW, JHQ and SK analysed and interpreted the data. SW wrote the first draft of the manuscript. MDB, SK, JD, KA, CD, JEC, BU, MB, VG, LJ and JHQ revised the first draft critically for important intellectual content. All authors have revised the subsequent drafts critically, approved the final manuscript to be published and agreed to be accountable for all aspects of the work.

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Figures

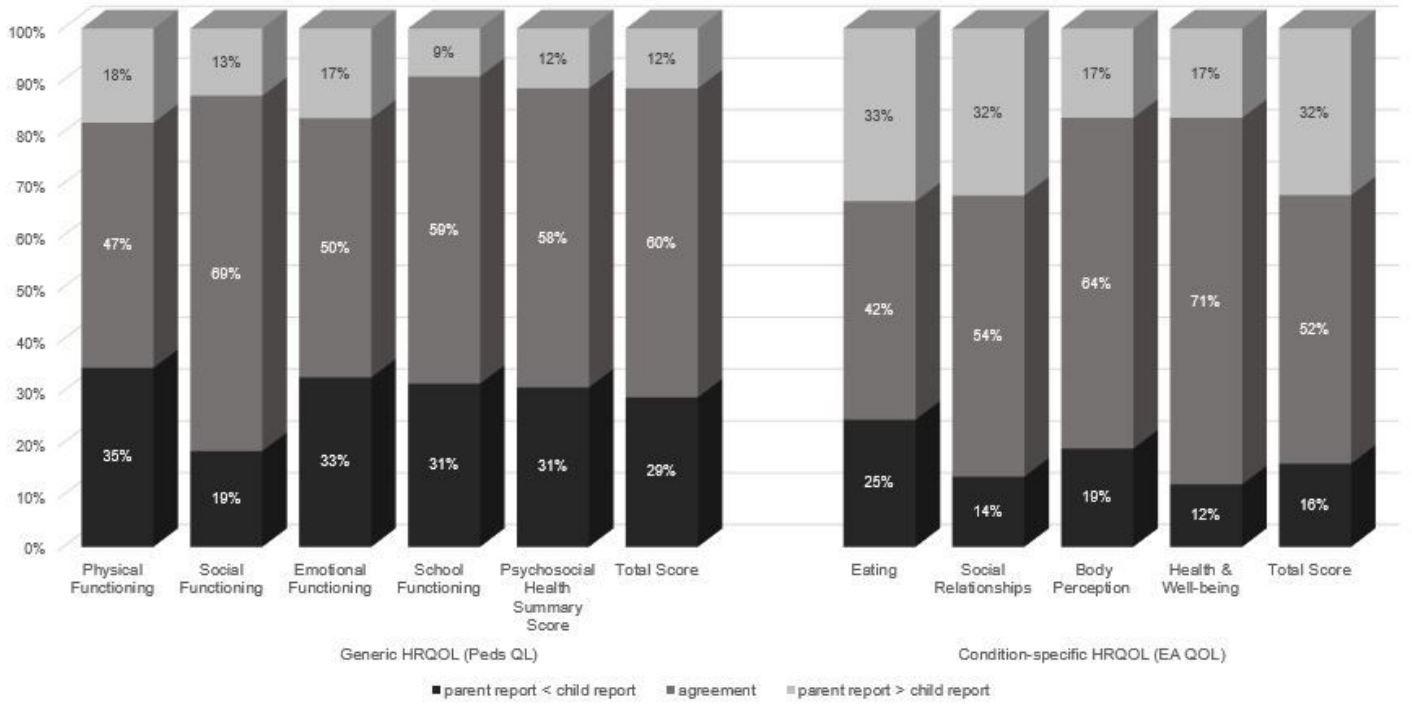

Figure 1

Distribution of parent-child directional discrepancies on reports of generic and condition-specific HRQOL