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Health-related quality of life experiences among children and adolescents born with esophageal atresia: Development of a condition-specific questionnaire for pediatric patients

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ABSTRACT

Background/Purpose: The aims were to present the framework for the development a condition-specific health-related quality-of-life (HRQOL) questionnaire for children with esophageal atresia (EA) and to describe HRQOL experiences reported by children and by their parents.
Methods: Utilizing the well-established DISABKIDS methodology, standardized focus group discussions were held and transcribed. HRQOL experiences were identified, content analyzed and evaluated using descriptive statistics.
Results: 30 families (18 children 8–17 years, 32 parents of children 2–17 years) participated in ten focus group discussions. 1371 HRQOL experiences were identified referencing social, emotional and physical aspects of eating and drinking ($n = 368$), relationships with other people ($n = 283$), general life issues; physical activity, sleep and general health ($n = 202$), communicative/interactive processes of one's health condition ($n = 161$), body issues ($n = 109$), bothersome symptoms ($n = 81$), impact of health care use/medical treatment ($n = 78$), confidence in oneself and in the future ($n = 65$) and difficulties because of concomitant anomalies ($n = 24$). A basis of two age-related HRQOL questionnaires for children with EA (2–7 years, 8–17 years) was subsequently constructed.
Conclusions: EA interacts with various aspects of the child's life. In addition to HRQOL issues of eating and drinking, social dimensions like relationships and interactions with other people seem to be prominent condition-specific HRQOL parameters. The settings for the development of the first condition-specific HRQOL questionnaires for patients with EA are established.

Survival rates among children with esophageal atresia (EA) have improved to 95% [1]. However, chronic morbidity among children is common, ranging from dysphagia, gastroesophageal reflux and respiratory disorders [2,3] as well as growth retardation [4]. Children with EA may suffer from food impaction, oral aversion and vomiting [4] as well as from wheezing, dyspnea, barky or chronic cough and recurrent airway infections with subsequent need of medical care [3,5]. Moreover, additional morbidity may result from associated anomalies [6]. Although criteria to evaluate medical outcomes have become increasingly supplemented by patient-reported outcome standards [7], studies of health related quality of life (HRQOL) among children with EA are few ($n = 5$) [8–12]. Only two studies [8,9] have compared the overall HRQOL to healthy references, of which one demonstrated reduced overall HRQOL in pediatric patients with EA. Generic HRQOL measurements permit the advantage of comparison with healthy reference norms, condition-specific HRQOL questionnaires provide more sensitive information with regard to specific clinical characteristics [13]. Since no study using a condition-specific HRQOL questionnaire among patients with EA has been reported [14], knowledge of this field remains limited. The long-term aim is to advance knowledge of HRQOL among children with EA through the development of a condition-specific HRQOL questionnaire, in this study to describe the establishment of the conditions for such a questionnaire and the HRQOL experiences reported by children with EA and by their parents.

1. Materials and methods

Approval to conduct the study was obtained from the Regional Research Ethics Committee.

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1.1. Framework and assessment of condition-specific HRQOL experiences

This study was guided by the well-established methodology of the large European DISABKIDS project for children with chronic health conditions [15]. The methodology was preferred since the design comprises a distinction between generic, chronic-generic and condition-specific HRQOL measurements. Within the project, standardized pediatric condition-specific HRQOL measurements were developed. Moreover, a patient-derived nature of questionnaire construction is central [16] and in compliance with the Food and Drug Administration guidelines for construction of an HRQOL instrument [17]. The assessment of condition-specific HRQOL experiences is based on focus groups with children and their parents, composed according to the child's severity of disease, age and gender. The reported HRQOL experiences form the construction of questions for a preliminary questionnaire. This questionnaire undergoes a question reduction process and is evaluated through cognitive debriefing, a pilot test and ultimately a field test in a larger sample of the target population [15].

1.2. Focus group procedure

1.2.1. Selection of participants

Criteria were developed for two levels of severity of EA (mild to moderate and severe) including input from pediatric surgical expertise, QOL methodology expertise and from previous research of EA [5,12,18–22]. Medical records of 135 children born with EA in 1997–2013 were reviewed for clinical data. 73 children (54.0%) were categorized as cases of severe EA. Stratified for child gender, families of each five children with mild to moderate EA and five children with severe EA were selected in three age groups (0–7, 8–12, 13–17 years) and were invited to participate in the study. Children younger than eight years were represented by their parents (proxy-report). Patients were considered to have severe EA if one or more of four criteria were met (Table 1). Associated malformations were defined as severe if associated with patient disability [23].

1.2.2. Data collection

Informed consent to participate in the study was obtained. Separate focus group discussions with the children and their parents took place in 2014 and were digitally recorded. A questionnaire of child and family characteristics was completed. The DISABKIDS focus group manual, consisting of eight questions with a progressive focus on the main issues of HRQOL, formed a standard basis for discussions with both children and parents [15]. All participants were asked questions about the nature and extent to which the EA condition has continued to affect the child's daily life. Discussions were led and facilitated by a moderator who ensured that all participants had an opportunity to contribute. A research assistant was present during the child focus groups and was responsible for taking field notes of non-verbal communication and group interactions.

1.2.3. Data analysis

Focus group discussions were transcribed verbatim. HRQOL experiences were content analyzed as illustrated in Fig. 1. HRQOL experiences were extracted from the focus group text, merged with participant information into Excel 2010, formulated as a statement and card sorted into domains and overall HRQOL areas with a particular HRQOL content in common [15,16]. Descriptive statistical analysis of the clinical and sociodemographic data, the frequency and distribution of HRQOL statements according to domains, overall HRQOL areas, severity of EA, child gender, age group (0–7, 8–12, 13–17 years), child and proxy reports was performed using SPSS 22.0. The results were used to derive questions for the construction of a HRQOL questionnaire basis (adjusted for four weeks recall period and a five-point Likert scale from never to always). An unbiased categorization of HRQOL statements, as well as selection and reformulation into questions were ensured through consensus among several researchers.

Table 1
Inclusion criteria of patients in the severe esophageal group ($n = 15$).

	Number of patients (%)	Single inclusion criteria (%)
The primary anastomosis was delayed and/or EA replacement was accomplished	7 (47)[a]	3 (20)
Major surgical revision (open surgery) of the EA correction performed for causes as recurrent TEF or anastomotic leakage	5 (33)	1 (7)
Presence of a severe tracheomalacia or tracheobronchomalacia based on macroscopic estimation of an anteroposterior collapse documented as excessive, severe and/or of ≥75% without limitation of the child's age at the bronchoscopy [21][b]. If the child had been examined several times, the most recent bronchoscopy was considered the most valid for inclusion/exclusion	5 (33)	4 (27)
Presence of at least one other congenital health condition resulting in disability. The term disability was defined according to the ICF-CY. Disability is served as an umbrella term for impairments (problems in body function or structure as a significant deviation or loss), activity limitations or participation restrictions [23][b]	4 (27)[c]	3 (20)

EA, esophageal atresia; TEF, tracheoesophageal fistula; ICF-CY; International Classification of Functioning, Disability and Health-Child & Youth Version.
[a] Esophageal replacement was performed in three children.
[b] Reference number according to manuscript.
[c] The following associated malformations were considered as severe; anorectal malformation (two patients), severe urogenital malformations (one patient), central nervous system anomaly resulting in neurogenic bladder dysfunction (one patient), and congenital hypothyreos (one patient).

2. Results

2.1. Patient and parent characteristics

Ten focus group discussions were held (19.2 hours, mean 1.9 hours). All 30 families participated (100%) and were represented by 18 children (8–17 years) and 32 parents of children (2–17 years). As illustrated in Table 2, child gender and age were similar among patients with mild to moderate and severe EA. Morbidity and health care needs were present among children independent of severity of EA, but especially among patients with severe EA. In a majority of families, proxy-reports were provided by the maternal parent.

2.2. Condition-specific HRQOL experiences and the construction of a questionnaire basis for children with EA

1371 HRQOL statements/experiences were identified. A majority (66.9%) was generated from children with severe EA and their parents. Of the aggregate HRQOL experiences, 716 (52.2%) were child reports, and with respect to gender distribution, 730 (53.2%) and 641 (46.8%) were for males and females, respectively.

2.2.1. Characteristics and distribution of HRQOL domains

The HRQOL statements were categorized into 30 domains referencing emotional, social and physical perspectives of HRQOL experiences. Table 3 provides a description of each domain in descending order of statement frequency (116 to three). In Supplemental Material 1, each domain description is accompanied by a representative focus group quote as expressed by children and their parents.

2.2.2. Characteristics and distribution of overall HRQOL areas/domains

The allocation of 30 domains into nine overall HRQOL areas and their distribution according to severity of EA, child gender and child age

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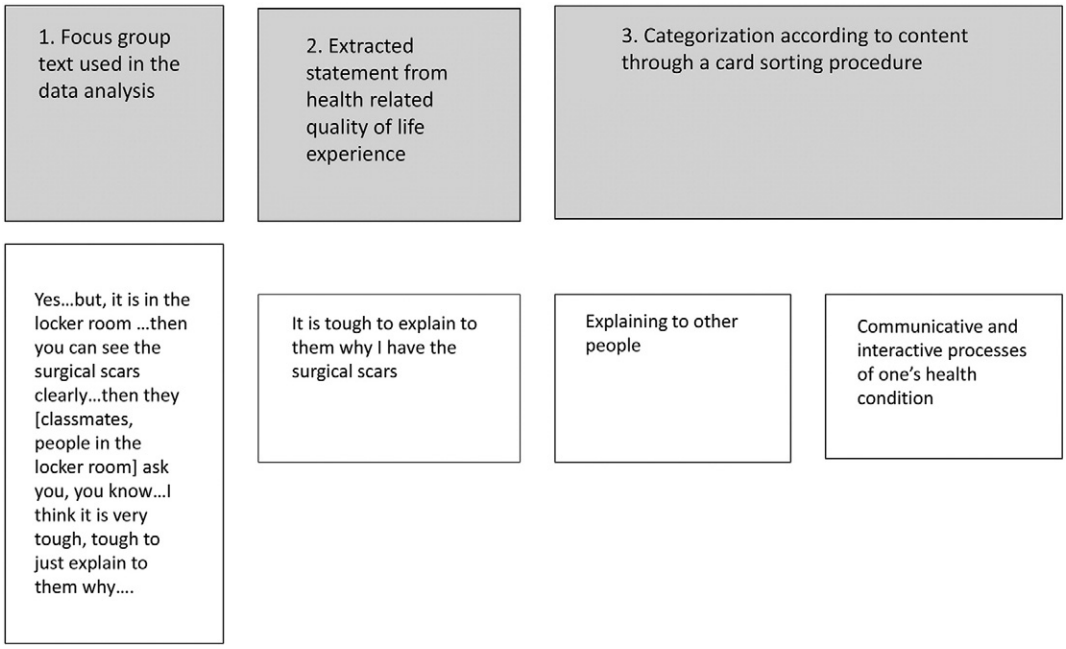

Fig. 1. Illustration of the content analysis procedure. Health-related quality of life experiences reported by children and by parents were identified in the transcribed focus group discussions. These were merged into Excel 2010 with participant information, formulated as a statement in a reduced form and each of them were printed on a card with participant information. During a card sorting procedure performed by three researchers, the statements were sorted into categories of health-related quality of life experiences that shared a common feature.

Table 2
Characteristic of focus group participants.

Variable	Mild–moderate esophageal atresia (*n*, children = 15; *n*, parents = 16)			Severe esophageal atresia (*n*, children = 15; *n*, parents = 16)		
	Frequency (%)	Mean/Median (SD)	Min/Max	Frequency (%)	Mean/Median (SD)	Min/Max
Child information						
Gender						
Female	7 (47)			7 (47)		
Male	8 (53)			8 (53)		
Age at focus group follow-up (year)		10/9 (5)	2/17		10/11 (5)	3/15
Gestational age at birth (week)		37/38 (3)	32/41		35/34 (4)	30/41
Birth weight (gram)		2581/2700 (1)	1660/3340		2193/2120 (1)	1080/3420
Associated anomalies	5 (33)			11 (73)		
Dilatation of esophagus	6 (40)	0.9/0 (1)	0/5	12 (80)	7/5 (9)	0/30
Medication	8 (53)			12 (80)		
Antireflux medication	2 (13)			8 (53)		
Bronchodilators or Inhaled steroids	5 (33)			9 (60)		
Other	2 (13)			7 (47)		
Growth retardation (weight and/or height < 2SD)	0 (0)			7 (47)		
Gastrostomy	0 (0)			2 (13)		
Additional school support	1 (7)			4 (27)		
Siblings	14 (93)			13 (87)		
Area of residence						
Rural residence (outside metropolitan and town)	3 (20)			4 (27)		
Town (<100000)	4 (27)			7 (46)		
Metropolitan (>100000)	8 (53)			4 (27)		
Parental information						
Proxy-representatives						
Mother	12 (75)			14 (87)		
Father	4 (25)			2 (13)		
Age (year)		44/43 (7)	28/59		39/39 (5)	33/47
Civil state (families)						
Married	9 (60)			9 (60)		
Divorced	2 (13)			1 (7)		
Cohabitant partner	3 (20)			4 (27)		
Non-cohabitant partner	0 (0)			1 (7)		
Widow	1 (7)			0 (0)		
Healthy	14 (88)			15 (94)		
National descent Swedish	13 (81)			13 (81)		

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Table 3
Presentation of HRQOL statements/experiences ($n = 1371$) in descending order according to domain.

Domain	Frequency (%)	
Physical activity like sport and play	116 (8.5)	Experiences of problems with regard to performing or participating physical activities like sport and play
Bullying and social exclusion	88 (6.4)	Experiences of being teased, bullied, called things, getting comments for being small/short/eating differently/having a special cough
Food issues	82 (6.0)	Experiences of food limitation, food restriction, special food preferences, the need of adjusted food. Problems with feeling hunger, taste and insufficient nutrition
Impact of choking	79 (5.8)	Experiences of problems and strains (such as worry, fear, panic) with choking/food getting stuck in the throat while eating. Efforts and need of chewing, dividing into pieces, peeling, tearing, avoiding food to eliminate choking
Nutritional intake experiences	78 (5.7)	Experiences of eating slower, different or similar food as peers/siblings, wishes to eat like others and strains of having difficulties swallowing
Communication with other people about EA	78 (5.7)	Experiences of being open, concealing, confident or careful when communicating with others about EA
School cafeteria experiences	73 (5.3)	Experiences of problems or stress when eating in the school cafeteria, e.g., lack of time to eat and spend time with peers during lunch break, being (left) alone in the school cafeteria because of eating slow, dissatisfaction with receiving special food. The need of a special place and of support from teachers
Friends in school and free time	70 (5.1)	Experiences of having or not having friends to spend time with during breaks and free time, their understanding and support regarding problems as a result from EA
Body issues and surgical scar experiences	70 (5.1)	Experiences of feeling different because of surgical scars, scoliosis and/or winged scapula. Inconvenience/dis/satisfaction, difficulties or pride because of the look of their body or the appearance of scar(s) to other people. Problems of clothing because of body issues such as trying to conceal the surgery scar(s) or a winged-scapula
Sleep	64 (4.6)	Experiences of sleeping disorders caused by acid reflux or respiratory problems. Need of adjustment and planning of medications, food intake and/or of having your head high/using extra pillows in order to sleep well
Self-perception because of EA	58 (4.2)	Experiences, thoughts and emotions of being born with EA; perceptions of being different, unique, special, sad, ashamed, cool, abnormal or of being lucky to have survived
Medical treatment	53 (3.9)	Experiences of dissatisfaction, stress, boredom or sadness of medication intake or dilatation of the esophagus. Experiences of reduced compliance, hiding or throwing medications because of despise of medication intake
Family relationship and communication	46 (3.4)	Experience of parental support, child–parent communication, of the presence or lack of confidence and security in the family related to problems that could arise because of EA morbidity
Fluid intake experiences	45 (3.3)	Experiences of the need to be aware and to plan increased fluid intake when eating, to need more fluid intake when eating compared to peers and to be bothered and to feel different because of this
Explaining to other people	42 (3.1)	Experiences of having to explain to other people about your cough, difficulties with swallowing, vomiting, surgical scar(s), breathing difficulties, wheezing, physical shape
Loneliness	41 (3.0)	Experiences of any difficulties gaining friends and/or having reduced social competence, isolated emotions of loneliness, of being the only one born with EA, reduced knowledge of other people with EA and wishes to meet children with EA
Other people's questions and wonderings	41 (3.0)	Experiences of other people's questions and wonderings because of surgical scar(s), the gastrostomy button, choking problems, special/barky cough, respiratory problems or special food
Being short and small for age	39 (2.8)	Experiences of being shorter and smaller than peers, worry, stress, dissatisfaction, sadness of being short and small and of seeing younger siblings becoming taller. Being bothered by the need to look up at your friends. Problems with finding clothes that fit your body
Respiratory symptoms	39 (2.8)	Being bothered and having problems because of mucus production, cough, recurrent respiratory infections, breathing problems, having a cough that sounds barky, louder or more special compared to other people
Acid reflux experiences	24 (1.8)	Perceived problems with acid reflux symptoms like pain, or disgusting taste
Additional difficulties because of concomitant anomalies	24 (1.8)	Experiences of problems as a result of other concomitant anomalies like complexity of food intake when having esophageal atresia and anorectal malformation or difficulties with urinary leakage, urinary catheter or troubles with constipation
Perceived general health	22 (1.6)	Experiences of health and life and factors important to perceive health (friends, school, sports)
Vomiting experiences	18 (1.3)	Experiences of vomiting at school, among peers and during play
Understanding and support from school	18 (1.3)	Perceptions of the teachers' available or non-available support and understanding of the child's health condition and health care needs and experiences of child safety in school
Expression of empathy to others	17 (1.2)	Empathy, warmth, sense of justice to people that are different, weak or have other difficulties
School absence	13 (1.0)	Experiences of school absence because of illness and hospital visits, e.g., problems of being away from friends or of the need for careful study planning to keep the same study pace as classmates
Gastrostomy button	12 (0.9)	Experiences of having a gastrostomy button
Children's parties	11 (0.8)	Experiences at children's parties, e.g., stress while eating, requirement of special food or vomiting problems at or after eating
Confidence in future	7 (0.5)	Thoughts and emotions of worry, confidence or insecurity about the future
Confidence in finding a partner	3 (0.2)	EA impact of the confidence in finding a partner

EA, esophageal atresia; HRQOL, Health-related quality of life.

group are shown in Table 4. The HRQOL areas and the reported frequency were eating and drinking ($n = 368$ [26.8%]), relationships to other people ($n = 283$ [20.6%]), general life issues such as physical activity, sleep and perceived health ($n = 202$ [14.7%]), communicative/interactive processes of one's health condition ($n = 161$ [11.7%]), body image issues ($n = 109$ [8.0%]), bothersome symptoms ($n = 81$ [5.9%]), impact of health care use and medical treatment ($n = 78$ [5.7%]), confidence in oneself and future ($n = 65$ [4.7%]) and additional difficulties because of concomitant anomalies ($n = 24$ [1.8%]). Some HRQOL areas were rarely or not at all represented among each age group. In all domains, the larger part of HRQOL experiences was described by and for children with severe EA.

2.2.3. The basis of two condition-specific HRQOL questionnaires for children with EA

Two age-related versions of HRQOL questionnaires for children with EA aged 2–7 years (parent report only) and children aged 8–17 years (child and parent report) were developed (Fig. 2).

3. Discussion

This study investigated HRQOL experiences among children with EA and showed that condition-specific HRQOL parameters concerned various aspects ranging from participation in play and sport, nutritional intake, social and emotional concerns and body image issues. Several

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Table 4
Allocation of HRQOL experiences ($n = 1371$) into nine HRQOL areas and their distribution according to severity of esophageal atresia, child gender and child age group.

Overall domain	Severity of EA		Child gender		Child age group		
	Frequency (%)		Frequency (%)		Frequency (%)		
	Mild–moderate	Severe	Male	Female	2–7	8–12	13–17
Eating and drinking; food issues, impact of choking, nutritional intake, school cafeteria, fluid intake, children's parties	100 (7.3)	268 (19.5)	201 (14.7)	167 (12.2)	79 (5.8)	165 (12.0)	124 (9.0)
Relationships with other people; bullying and social exclusion, friends, family, loneliness, understanding and support from school, expression of empathy to others, confidence in finding a partner	94 (6.9)	189 (13.8)	140 (10.2)	143 (10.4)	44 (3.2)	148 (10.8)	91 (6.6)
General life issues; physical activity like sport and play, sleep, perceived general health	67 (4.9)	135 (9.9)	127 (9.3)	75 (5.5)	45 (3.3)	108 (7.9)	49 (3.6)
Communicative/interactive processes of one's health condition; communication with other people about EA, explaining to other people, other people's questions and wonderings	65 (4.7)	96 (7.0)	72 (5.2)	89 (6.5)	16 (1.2)	95 (6.9)	50 (3.6)
Body issues; body issues and surgical scar(s), being short and small for age	52 (3.8)	57 (4.2)	52 (3.8)	57 (4.2)	13 (0.9)	66 (4.8)	30 (2.2)
Bothersome symptoms; respiratory symptoms, acid reflux, vomiting	30 (2.2)	51 (3.7)	39 (2.8)	42 (3.1)	17 (1.2)	48 (3.5)	16 (1.2)
Impact of health care use and medical treatment; medical treatment, school absence, gastrostomy button	18 (1.3)	60 (4.4)	55 (4.0)	23 (1.7)	11 (0.8)	39 (2.8)	28 (2.0)
Confidence in oneself and in future; self-perception because of EA, confidence in future	25 (1.8)	40 (2.9)	33 (2.4)	32 (2.3)	6 (0.4)	29 (2.1)	30 (2.2)
Additional difficulties because of concomitant anomalies	3 (0.2)	21 (1.5)	11 (0.8)	13 (0.9)	16 (1.2)	8 (0.6)	0
Total	454 (33.1)	947 (66.9)	730 (53.2)	641 (46.8)	247 (18.0)	706 (51.5)	418 (30.5)

EA, esophageal atresia.

HRQOL experiences like those of stigma, isolated emotions and impact of medical treatment are similar to those of children with other chronic health conditions [16,24]. In this study, HRQOL experiences of eating and drinking may be understood as particularly important. As previously known, children with EA may suffer from feeding difficulties and growth retardation, especially at early child age [4]. Additionally, this study provides information that among EA children up to 17 years, feeding difficulties and growth retardation can give rise to social and emotional strains such as being different to peers or being teased. Also, EA may lead to negative HRQOL experiences in the school cafeteria and during children's parties. Moreover, EA seem to possibly affect the child's relationships and interaction with other people. In agreement, Dingemann et al. [12] in 2014 showed that children with complicated EA had reduced social function, and in 2012 Legrand et al. [8] demonstrated that children with EA type III, with associated anomalies had reduced school functioning. However, Lepeytre et al. [9] in 2013 identified improved social function among children with EA type III ages 8–13 years. Altogether, this implies that HRQOL of children with EA may depend on the quality of functioning with peers, family and teachers. Moreover, the reported HRQOL experiences revealed that the child's physical state could limit participation in play and sport and that symptoms were bothersome. In comparison, previous studies have shown an impaired general health and/or physical health [8,11] and that the presence of symptoms could negatively influence overall HRQOL [10]. In this study, children with EA independent of surgical scar(s), scoliosis or a winged-scapula experienced concerns that were related to discomfort among others, to the sense of being different or to dissatisfaction because of their look. Similar to our findings, Koivusalo et al. [25] demonstrated that about half of adults with EA had complaints of surgical scar(s) and 11% were disturbed by a disfigured or winged scapula, Deurloo et al. [26] demonstrated that 9% of adults felt negatively affected by their surgical scars and Lima et al. [27] demonstrated that after esophageal replacement (mean age of patients was 34.5 years), esthetic results were a main problem, especially during adolescence.

From the perspective of the pediatric surgeon, knowledge of HRQOL issues of significance to the EA children may improve the decision-making process of surgical technique and the patient–surgeon communication during the postoperative follow-up. It may permit identification of reduced HRQOL, presentation of well-directed patient and parent information, provision of relevant family support services and importantly, optimization of the postoperative medical management. A follow-up in accordance with the patient's needs might improve long-term health and HRQOL outcomes. International guidelines for the construction of patient reported outcome measurements [15,17] were so far applicable; a questionnaire basis was formed by HRQOL experiences identified in this study. It is anticipated that a condition-specific questionnaire [13] could help improve the evaluation of surgical techniques of EA.

Considering the rate of associated anomalies and of the medical treatment because of respiratory or esophageal morbidity, children in this study were representative [3,8,28]. Nevertheless, generalization of the findings is limited because of a small patient sample and the single-center study design. Although an increased number of HRQOL statements may indicate that an issue discussed in the focus groups is important, it can neither be used to generalize the finding nor determine the extent to which HRQOL is affected. Moreover, the categorization of severity of EA is complex. Previous research did not employ a uniform definition of severe EA, and risk factors for development of complications are only partly known [18,19]. Accordingly, inclusion criteria for severe EA were carefully constructed. As an indication of group homogeneity, an increased prevalence of HRQOL experiences was provided by or on behalf of children with severe EA. Because of age-group variations, two age-related versions of HRQOL questionnaires were constructed. Focus groups data are not applicable for statistical testing; future investigations are needed to not only determine the extent to which HRQOL is affected, but also identify factors that may influence condition-specific HRQOL.

It strengthens the study that all participants have contributed information and that the number of child reports is high, however the selection and categorization of HRQOL experiences risk interpretation bias. Therefore, efforts were made to maintain objectivity; consensus among several researchers and utilization of field notes in order to better understand the reported findings minimized interpretation of the participants' reports [16]. Presentation of a categorization process and focus group quotes exemplified completion of objectivity.

4. Conclusion

Following international guidelines for the construction of a patient-reported outcome measure, the perspectives of children with EA and their parents have been incorporated into the development of the first condition-specific HRQOL questionnaire for patients with EA. According to the reported HRQOL experiences, EA interacts with various aspects of the child's life, especially among children with severe EA. In addition to HRQOL issues of eating and drinking, social dimensions of relationships

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Quality of Life Questionnaire for children with Esophageal Atresia

Domain	Questionnaire for children 2-7 years (parent report)	Questionnaire for children 8-17 years (child and parent report)	Example of question
Eating and drinking	22	32	*Are you stressed by having to rush to eat your food in the school cafeteria?*
Relationships with other people	11	21	*Do you get teased about things in school?*
General life issues	9	15	*Do you have the strength to play sports (e.g. running, playing football) and play as your friends do?*
Communicative/ interactive processes of one's health condition	3	9	*Does it feel awkward when others ask you about esophageal atresia/your health condition?*
Body issues	5	12	*Are you careful about what you wear because of your scar /scars?*
Bothersome symptoms	3	10	*Are you bothered by that you have a different kind of cough?*
Impact of health care use and medical treatment	3	8	*Is it hard having to take medications?*
Confidence in oneself and future	1	10	*Do you think about how your future will be because of your health condition?*
Additional difficulties due to concomitant anomalies	1	1	*Do you feel that more than one medical condition cause you difficulties in daily life?*
Total number of questions	58	118	

Fig. 2. Selected questions to be used as a basis for two age-related condition specific health-related quality of life questionnaires for children and adolescences with esophageal atresia. The basis of two condition-specific health-related quality of life questionnaires, for children aged 2–7 years (parent report only) and children aged 8–17 years (child and parent report), were subsequently formed by the results of the focus groups. The number of questions is presented in relation to overall domain and an example of a question is given. The questions are selected for further evaluation through cognitive debriefing, a pilot study and a field test and will undergo a reduction process.

and interactions with other people appear to be prominent condition-specific parameters. Body image concerns seem to be present. As complements to clinical endpoints such as survival or morbidity rates, such condition-specific parameters provide important information that increases our understanding of the possible long-term impact of EA and/or surgical techniques from the patient's perspective. This information can be used to improve the evaluation of pediatric surgery and it also sheds light on issues that should be integrated into follow-up routine care. In the end, the identified parameters may serve as possible benchmarks between centers, enable comparison of subgroups of EA and treatments, but also enhance the evaluation of pediatric surgical care in research.

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