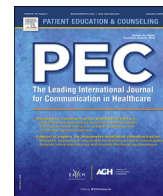




Contents lists available at ScienceDirect

Patient Education and Counseling

journal homepage: www.elsevier.com/locate/pateducou



Patient education for children with interstitial lung diseases and their caregivers: A pilot study

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ARTICLE INFO

Article history:

Received 26 April 2018

Received in revised form 15 January 2019

Accepted 18 January 2019

Keywords:

Children's interstitial lung disease (chILD)

Patient education

Self-management

Self-efficacy

ABSTRACT

Objective: Patient education in children with rare chronic diseases like children's interstitial lung disease (chILD) remains a challenge.

Aims: To develop and evaluate a component-based educational program for individual counselling and to improve patients' and caregivers' self-efficacy and treatment satisfaction. Furthermore, to create chILD-specific educational material and assess physicians' satisfaction with the intervention as well as patients' health-related quality of life (HrQoL).

Methods: The study was conducted in two German centers for pediatric pulmonology, as a single-group intervention with pre-post-follow-up design.

Results: Participants (N = 107, age: M = 7.67, SD = 5.90) showed significant improvement of self-efficacy (self-report: $t = 2.89$, $p < 0.01$; proxy-report: $t = 3.03$, $p < 0.01$), and satisfaction (patients: $t = 3.56$, $p = 0.001$; parents $t = 6.38$, $p < 0.001$) with the medical consultations. There were no pre-post differences in HrQoL. Participants were highly satisfied with the material and the physicians with the program.

Conclusions: The chILD education-program is a promising strategy to improve patients' and their parents' self-efficacy and treatment-satisfaction. Specific effects of the intervention need to be determined in a randomized controlled trial.

Practice implication: Healthcare providers managing pediatric patients with chILD, may choose to use a patient education-program specifically tailored to the needs of chILD patients and their families, such as the program described here, which is the first of its kind.

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1. Introduction

Children's interstitial lung disease (chILD) is a term for more than two hundred different mostly rare chronic lung diseases affecting the lung parenchyma. It is associated with varying morbidity. The estimated prevalence is 0.36 per 100,000, the incidence 1.32 per 1,000,000 [1–7]. Symptoms are nonspecific but patients typically present with tachypnoea, hypoxemia, failure to thrive, crackles on auscultation and diffuse infiltrates on chest radiography [8–10]. There is a wide range of disease severity [10] and a lack of knowledge about the pathogenesis and the clinical course in most of the specific chILD-diagnoses [4]. Hence, treating

patients with chILD constitutes a challenge for pediatric pulmonologists [11].

Patients and their families have to manage multicomponent treatment regimes, which can be complex, invasive, and time consuming, thus placing substantial burden on families who are already negotiating the typical activities and developmental challenges of childhood [12]. Individualized disease management requires a multifaceted approach including effective medical treatment and patient education to reduce disease related worries and support patients and families in their disease management (e.g. ventilation, inhalation, financial support) [13]. Patient education should help patients to acquire or maintain the skills they need to manage their life with this chronic disease in the best possible way (World Health Organization, 1998) [14]. Modi and colleague's conceptualization of self-management contains 3 interdependent components (self-management behaviors, contextual variables, and the processes).

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In Modi's Pediatric Self-management Model, self-management behaviors operate with individual, family, community, and health care system domains. Through underlying cognitive, emotional, and social processes, modifiable and nonmodifiable domain-specific influences impact self-management [12].

Patient education is a frequently used interventional approach for chronically ill patients, including chronic obstructive pulmonary disease (COPD), asthma or cystic fibrosis (CF). Patient education programs aim to enhance disease management, management of procedural distress [15–20] and improve perceived self-efficacy [21–23]. Self-efficacy is a construct based on the social cognitive theory, which refers to "people's beliefs about their capabilities to exercise control over their own level of functioning and over events that affect their lives" [38]. Different guidelines generated by national and international expert panels underline the importance of patient education in the field of chronic lung diseases [24–27]. The education material should be easy-to-read and strategically addressed to provide efficient, cost-effective, quality health education. Finally, quality health education aims to improve people's lives by facilitating a change in their attitudes, beliefs, knowledge, skill levels, and behavior [28].

Furthermore, improved disease management has been associated with better health-related quality of life (HrQoL) in children and their families with asthma [18,29–32] and CF [33,34]. Patient education therefore has the potential to improve patients' HrQoL and health [11,29], and interventions to improve self-management are recommended [9]. Nevertheless, findings regarding the impact of interventions that aim to improve self-management are not consistent regarding HrQoL. Some studies suggest significant positive outcomes [30,30,31,32], others a tendency towards improvements [33,34], while others show no effect [19,21,35].

Many families of children suffering from chILD reported knowing little generally about the disease, both at the time of diagnosis and during follow up [28]. In absence of evidence based treatment options, it appears extremely important to conduct shared medical decision making between healthcare professionals (HCPs), patients and their caregivers. This requires strategies to share the existing knowledge about chILD with affected families. Despite the physicians' awareness of the importance of a careful patient education program, implementation and sustainability cannot be taken for granted [35,36]. Friberg et al [37] concluded that patient education was not considered to be a part of routine care but was often omitted by HCPs due to other work demands and a high workload. Lelorain et al [14] found the key factor to motivate professionals was the integration of patient education into routine care within a multidisciplinary team. Additionally, healthcare managers should consider how they can promote patient education as a routine, expected, monitored and remunerated aspect of the provision of care [38–40].

Currently, there are no patient education-standards for patients with chILD and their families. Due to the high information needs about the disease and its treatment in affected families, the aim of the current study was to develop and evaluate a semi-structured patient education-intervention for children with chILD and their caregivers. We hypothesized a significant improvement:

- 1 of self-efficacy in patients with chILD and their parents after the educational program, compared to pre-intervention (primary outcome);
- 2 of satisfaction with the medical consultation after patient education-sessions;
- 3 in HrQoL after the educational program, compared to pre-intervention.
- 4 Finally, we explored feasibility of the semi-structured patient education within clinical routine care.

2. Methods

2.1. Study design

This study was conducted between June 2014 and December 2016, as a single-arm study with a pre-post 1-month follow-up design in two German tertiary pediatric pulmonology centers, specialized in chILD in Hannover and Munich. Information material was developed based on the current literature and results of a self-developed questionnaire, which assessed current information needs of affected families in Germany. The latter were identified interviewing preliminary focus groups at the two centers. For details of the development of the patient information-brochure see online supplemental materials E1. The chILD-specific patient education-program focused on the communication of disease-specific knowledge and treatment related skills, in order to improve the patients' and caregivers' adaptation to the medical challenges of these rare lung diseases. We developed instructions for clinicians as a guideline to conduct patient education continuously, based on the didactic principles "keep it short and simple" and patient-centered care of "shared-decision making" [41]. A checklist to assess the individual information needs was filled out prior to each consultation by patients and their parents. Based on this checklist the physician conducted the patient education-session. Contents were based on the patient information brochure previously developed by us. They were delivered as a combination of printed material and verbal communication. Subsequently, the printed material was delivered to patients and their parents with the opportunity to read it carefully at home. The study protocol was approved by the Institutional Review Board (IRB) at the University of Munich, Germany (EK 111-13), which was the coordinating site of the chILD-EU consortium and by the participating centers in Ulm and Hannover. The chILD-EU consortium was conceptualized for clinical scientists and pediatric pulmonologists to assemble cohorts of children with well-defined disease entities. The disease entities are verified by international panels of clinicians, radiologists, geneticists and pathologists. Follow-up of children with a confirmed diagnosis is monitored in a web-based database to allow international collaboration on these rare disease entities. The aim was to develop evidence-based and consensus-agreed clinical guidelines to achieve better care of patients afflicted by chILD and to improve their HrQoL (<http://www.childeu.net>) [10].

2.2. Participants

Inclusion criteria were (1) being a child/caregiver of a child with proven or suspected chILD, (2) child's age is between 0 months and 22 years, and (3) sufficient German language skills to understand educational contents. Parents were informed about the study and all participating patients, if old enough, gave their informed assent and all caregivers gave their informed consent to participate in the study before the baseline assessment.

2.3. Intervention

2.3.1. Patient information-brochure

The chILD-specific patient education-program consisted of one generic and one disease specific part. The generic part was standardized for all patients with chILD and their parents. This component offers basic education material about the structure and function of airways, chILD-diagnosis, diagnostic procedures, therapy, nutrition, complications, school, leisure time, holidays and traveling, social and financial support, research, self-help groups and a short medical dictionary that should be useful for all patients/parents to be able to better cope with the chronic

condition. The disease specific part contained information that could be chosen by the HCP depending on the patient's particular individual medical or psychosocial needs. All components could be combined and were independent from each other. Patients and caregivers could collect specific educational leaflets from the information booklet which was available as a web-based resource (<http://www.childeu.net>). A detailed overview of the content of the patient education-program is given in E-Table 2.

2.3.2. Instructions for clinicians

Instructions were developed as a guideline for physicians to conduct the patient education-consultation. It contains helpful hints for a developmentally adapted procedure and practical suggestions to transfer disease-specific and treatment related information in a way that non-professionals and children are able to understand. The instructions for the clinicians are given in the online supplement (E2).

2.3.3. Checklist

To assess the patients' and caregivers' individual information needs, we developed and implemented a checklist analogue to the patient information-brochure. The checklist for patients and caregivers can be seen in the online supplement (E3, E4). For each education session, patients and parents could prioritize three topics they wanted to learn more about.

A detailed protocol of the manualized program is available for free at the chILD-EU web site <http://www.childeu.net>.

2.3.4. Implementation

The implementation of the patient education program followed six steps: (i) HCPs were trained in a one-day train-the-trainer inhouse session by experienced study staff based on the patient information-brochure and instructions for clinicians. (ii) patients' and/or caregivers' information needs were assessed with the checklist right before the appointment with the HCP, (iii) educational goals were agreed upon with the patient and/or the caregiver using the completed checklist, (iv) educational sessions were conducted in addition to the regular clinical visits using an additional timeslot of approximately 30 min, based on the prioritized educational goals, with the education booklet for illustration as needed, (v) the patient education booklet with further information material was given for homework, (vi) patients' and/or caregivers' skills were evaluated at least 4 weeks after the patient education-program.

2.4. Instruments

The questionnaire to assess satisfaction with consultation was used at baseline (t0) which refers to previously conducted consultations and immediately afterwards (t1) in order to avoid a bias regarding the remembered counselling session. Questionnaires to evaluate self-efficacy and HrQoL were administered at baseline (t0) and one month after the visit (t2) to allow contents of the counseling to be implemented in the daily routine. The questionnaire to assess satisfaction with the information booklet was only used at t2. Satisfaction of the HCPs was assessed at t1. Socio-demographic data of the participants and medical information of the patients were collected at baseline.

2.4.1. Primary outcome

2.4.1.1. Self-efficacy. The self-efficacy-questionnaire for children with chILD assessed parents' perceptions of their self-efficacy regarding their child's disease as well as patients' own perception of their self-efficacy. It was adapted from the Inflammatory Bowel Disease questionnaire (IBD-yourself) [42] and was modified for

patients with chILD. The questionnaire comprises 12 items (e.g. I know how to protect myself/my child against infections.). A self-report version for children ≥ 8 years and a parent version for children ≥ 1 month are available. In both versions 4-point response scales are utilized (4 = Yes; 3 = Yes, probably; 2 = No, probably not; 1 = No). Hence, the possible range is between 0 and 48 points. Higher scores indicate perception of higher self-efficacy. The items can be added to a total score. The self-report (*Cronbachs'* $\alpha = 0.81$) and the parent-proxy report (*Cronbachs'* $\alpha = 0.90$) of the German version showed high internal consistency.

2.4.1.2. Secondary outcomes

2.4.1.2.1. Satisfaction with the educational program. This questionnaire assessed the child's and parents' satisfaction with the patient education-session. Using 9 items (e.g. HCPs answered my questions), a 4-point response scale is utilized (4 = Yes; 3 = Yes, probably; 2 = No, probably not; 1 = No). In the last item participants were asked to evaluate the patient education program by using a scale from 6 (excellent) to 1 (insufficient). The possible range is between 0 and 42. The questionnaire was constructed specifically for this study as a self-report for children > 8 years and as a parent-report version. Higher scores indicate greater satisfaction. In our sample, the self-report (*Cronbachs'* $\alpha = 0.69$) showed sufficient and the parent-proxy report (*Cronbachs'* $\alpha = 0.84$) showed high internal consistencies.

2.4.1.2.2. Health-related quality of life (chILD-QL). The chILD-QL is a disease-specific HrQoL questionnaire applicable to infants, toddlers, children and adolescents between 1 month and 18 years. Convergent validity was demonstrated and internal consistency reliability was high [10]. The chILD-QL self-report is available for children aged 8 years and above and comprises 5–8 items (e.g. 'I get scared when I have to have treatments or medical examinations'). The chILD-QL parent-proxy report is available in developmentally adapted versions for children 1 month and above and comprises 5–11 items. For self- and parent-proxy reports a 5-point response scale is utilized (0 = never a problem; 1 = almost never a problem; 2 = sometimes a problem; 3 = often a problem; 4 = almost always a problem). Items are reverse-scored and linearly transformed to a 0–100 scale. Higher scores indicate a better HrQoL. The individual responses are summed up to form a total score, indicating overall QoL. In the current study *Cronbachs'* α ranged between 0.89 and 0.94 for the self-report and between 0.74 and 0.93 for the parent-proxy report.

2.4.1.2.3. Satisfaction with the information booklet. A satisfaction questionnaire was constructed specifically for this study to assess the child's and parent's satisfaction with the information booklet as a self-report for children > 8 years and as a parent-report version. Both versions comprise 6 items (e.g. 'The information material is helpful for me.') on a utilized 4-point response scale (4 = Yes; 3 = Yes, probably; 2 = No, probably not; 1 = No) with a range of the scores between 0 and 26. In the last item participants were asked to evaluate the printed information material by using a scale from 6 (excellent) to 1 (insufficient). Higher scores indicate greater satisfaction with the information booklet. Items were added to a total score. In our study the self-report (*Cronbachs'* $\alpha = 0.73$) and the parent-proxy report (*Cronbachs'* $\alpha = 0.76$) showed sufficient internal consistency.

2.4.1.2.4. Satisfaction of the healthcare professionals with the educational program. This questionnaire was developed for this study by the research team and assessed satisfaction of HCPs with conducting a structured patient education-session and consists of 6 items (e.g. Structured PE is feasible in the daily routine care in clinics.) using 4-point response scale (4 = Yes; 3 = Yes, probably; 2 = No, probably not; 1 = No). Hence, the possible range is between 0 and 30. In the last item healthcare professionals were asked to evaluate the

patient education program by using a scale from 6 (excellent) to 1 (insufficient). Higher scores indicate higher satisfaction. Each item was analyzed separately due to the low internal consistencies. In our sample $Cronbachs' \alpha = 0.37$ showed low internal consistencies.

2.4.1.2.5. Medical and socio-demographic information. Medical variables were collected through a medical case report form filled out by the responsible physician, based on the patient's medical charts. Due to the heterogeneous group of rare chILD-diagnoses, initial diagnoses underwent a peer-review process by specialized clinicians within the study and were finally categorized according to the chILD-EU Classification Scheme [5,9,43,44]. Socio-demographic and socio-economic information were collected by the caregivers with a self-constructed questionnaire.

2.5. Statistical analyses

2.5.1. Sample size determination

In accordance with previous studies of patient education-programs for children and families with chronic conditions of the airways, the sample size necessary was calculated with the G*Power program 3.1 based on the following assumptions regarding the primary outcomes [17,18]. We expected at least pre-post effect sizes, Cohen's *d*, of 0.4 in the primary outcomes, based on two Cochrane review articles regarding educational intervention studies for children with asthma and CF [17,18]. 84 participants were sufficient to detect a controlled effect of $d \geq 0.4$ at a significance level of .05 (2-tailed) with a statistical power of 95%, using a Student's *t*-test for paired samples.

2.5.2. Statistical analyses

Statistical analyses were performed with the software program IBM Statistical Package for the Social Sciences (SPSS) for Windows Version 21.0. Single missing values in questionnaire raw item scores were replaced by the respondent's mean value of the questionnaire, if the proportion of missing data was less than 25%, otherwise the entire questionnaire was excluded from analysis. The imputation method last-observation-carried-forward (LOCF) was employed to replace missing data. This procedure assumed that the outcomes remain constant at the last observed value after drop out [45] and defined the last observation (prior to drop-out for patients dropping out) as last time point for patients completing the questionnaires [46].

Primary outcome analysis was based on the 1-month follow-up assessment. Our primary hypothesis was tested by *t*-tests for paired samples applying a significance level of $p < 0.05$ (two-sided). Levene's test was conducted and depending on assumptions of equality of variances *t*-scores were adjusted accordingly. All analyses regarding secondary outcomes were conducted in an exploratory manner. To reduce the risk of type I errors due to multiple tests, the significance level for the secondary outcome analyses were corrected using a Bonferroni-corrected α -level of 0.025 (two-sided).

Descriptive analyses (means and standard-deviations) were carried out to examine satisfaction with the patient information booklet, and satisfaction of the HCPs with patient education.

Additionally, effect sizes (Cohen's *d*) were calculated for within-group pre-post comparisons using the following formula: $d = (M_1 - M_2) / SD_{pooled}$ [47,48]

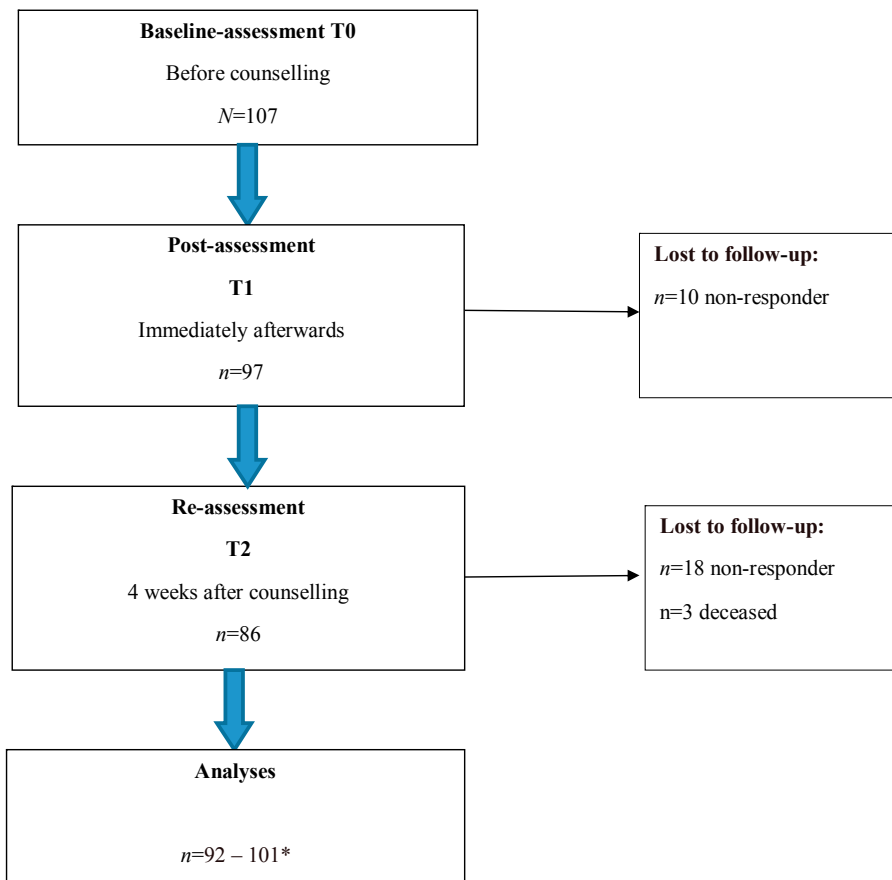


Fig. 1. CONSORT flowchart of the study.

*Note: LOCF was performed only if the proportion of missing data was less than 25%, otherwise the entire questionnaire was excluded from analysis.

3. Results

3.1. Sample description

One hundred seven patients (54 of them males) were enrolled in the study, whereof $n = 97$ (90.1%) filled in follow-up assessments immediately after consulting and 86 (80.4%) completed the 4-weeks follow up (see Fig. 1). Participants were on average 7.67 ($SD = 5.90$) years. Educational sessions had a mean duration of $M = 28.00$ ($SD = 8.52$, $Min = 20$, $Max = 40$) for the patient and $M = 29.65$ ($SD = 10.84$, $Min = 11$, $Max = 60$) minutes for parents. Details of the socio-demographic information are presented in Table 1. Medical characteristics of all participating patients are shown in Table 2. A total of 19 physicians conducted patient education-sessions. Demographic characteristics of the participating physicians are shown in Table 3.

3.2. Primary outcomes

3.2.1. Self-efficacy

The paired t -tests calculated on the sample ($n_{\text{parents}} = 101$ and $n_{\text{patients}} = 38$) demonstrated significant improvements for both self-reported ($t = 2.89$; $p = 0.006$) and parent reported self-efficacy ($t = 3.03$; $p = 0.003$). Pre-post effect sizes indicated by Cohen's d

were small for parent proxy reports and moderate for self-reports (Table 4, E-Figure 6).

3.3. Secondary outcomes

3.3.1. Satisfaction with patient education

The paired t -test of satisfaction with patient education, calculated on the sample ($n_{\text{parents}} = 98$ and $n_{\text{patients}} = 38$) showed a significant improvement from pre- to post-treatment for both self-reports ($t = 3.56$; $p = 0.001$) and parent-proxy reports ($t = 6.38$; $p < 0.001$). In both groups, pre-post effect sizes indicated by Cohen's d were moderate (Table 4, E-Figure 6).

3.3.2. Health-related quality of life

The parent proxy report for the childrens' HrQoL, calculated on the sample ($n_{\text{parents}} = 92$), demonstrated no significant improvement for the QoL total score ($t = 0.47$; $p = 0.64$), nor did the self-reported HrQoL ($n_{\text{patients}} = 38$; $t = 0.34$; $p = 0.74$) (Table 4, E-Figure 6).

3.3.3. Satisfaction with the information booklet

Self-reports were on average 17.77 ($SD = 4.98$) with a range of the scores between 2 and 22. Parent-proxy reports were on average 19.41 ($S = 3.14$). The range was between 4 and 22.

3.3.4. Satisfaction of the healthcare professionals with the structured patient education

Overall the HCPs were satisfied with conducting the patient education-program. Mean scores ranged from 2.38 to 3.64. HCPs evaluation of the program was very good ($M = 5.64$, $SD = 0.43$). Details of the analyses of the HCPs reports are presented in Table 5.

4. Discussion and conclusion

4.1. Discussion

This study investigated the efficacy of a structured patient education-intervention for children with cILD and their parental caregivers. Most outcome scores evaluated improved significantly between the pre- and post-intervention assessments. In detail, we found a significant increase in the primary outcome self-efficacy, which is in line with other studies performed in patients with CF [15,17] and asthma [18,49–51], as well as for the secondary outcome satisfaction with patient education. We could not find significant incremental effects of the additional psychoeducational intervention program regarding HrQoL [17,49,52,53].

Similar to Kennedy et al we found that our patient information brochure combined with HCPs training in patient-centered care improved overall patient satisfaction [54]. Patients and parental caregivers evaluated our information material very positively. The information material was utilized as a tool during the consultation to achieve a partnership in planning a system of shared medical decision making between HCPs, patients and/or their caregivers. As recommended in the literature, participating HCPs of our study worked with patients and their parental caregivers to clarify which components were of relevance to their current situation [54]. This additional support of HCPs seems to be a crucial element in implementing advice [55].

Furthermore, HCPs in our study reported to be overall satisfied with conducting the patient education-program. Although a higher score was obtained for future usage of the program, feasibility to conduct it in clinical practice appeared challenging. An additional 20–30 minute time slot to conduct one patient-education session during the visit seemed sufficient. A specific and continuing

Table 1
Socio-demographic information of the study participants.

Characteristics	N (107)	(%)
Age (years)		
mean	7.67	
SD	5.90	
Span	0-22	
Age groups		
1-12 months	12	11.2
13-24 months	10	9.3
2-4 years	22	20.6
5-7 years	15	14.0
8-12 years	23	21.5
13-18 years	21	19.6
> 18 years	4	3.7
Gender		
female	48	50.5
male	54	44.9
not reported	5	4.7
Participating caregivers		
mother	74	69.2
father	18	16.8
mother and father together	7	6.5
other (e.g. legal guardian)	3	2.8
not reported	5	4.7
Caregivers' level of education		
< 10 years of school	54	50.4
> 10 years of school	48	44.9
not reported	5	4.7
Mothers' employment		
full-time	19	17.8
part-time	46	43.0
not employed	31	29.0
not reported	11	10.3
Fathers' employment		
full-time	92	86.0
part-time	1	0.9
not employed	3	2.8
not reported	11	10.3
Child's care or educational institution		
not in any institution	27	25.2
daycare/nursery/preschool	24	22.4
school	45	42.1
vocational training	2	1.9
other	2	1.9
not reported	7	6.5

Table 2
Medical characteristics of the study participants.

Characteristics	N (107)	(%)
Treated in clinic		
Munich	47	43.9
Hannover	60	56.1
Classification Scheme ^{A)}		
DPLD/ILD not confirmed, entity ambiguous	32	29.9
More frequent in infants		
A1 - DPLD-Diffuse developmental disorders	2	1.9
A2 - DPLD-Growth abnormalities deficient alveolarisation	8	7.5
A3 - DPLD-Infant conditions of undefined ethology	13	12.1
A4 - DPLD-related to alveolar surfactant region	17	15.9
All age groups affected		
B1 - DPLD-related to systemic disease processes	20	18.7
B2 - DPLD-in the presumed immune intact host, related to exposures (infectious/non-infectious)	6	5.6
B3 - DPLD-in the immunocompromised host or transplanted	2	1.9
B4 - DPLD-related to lung vessels structural processes	3	2.8
By - DPLD-unclear NON-neonate	1	0.9
Non-DPLD		
C1 - Localized, congenital gross structural abnormalities of the lungs	2	1.9
D - Airway disorders	1	0.9
Period of time between start of symptoms and diagnosis (months)		
Mean	23.72	
SD	42.97	
Range	0–197	
Not reported	78	
Disease severity		
Asymptomatic without oxygen	40	37.4
Symptomatic, without oxygen (e.g. tachypnoe, dyspnoe)	31	29.0
Symptomatic with oxygen	26	24.3
Symptomatic continuous	14	13.1
Symptomatic only at physical strain	9	8.4
Symptomatic only at sleep	7	6.5
Symptomatic intermittent (e.g. in the context of infections)	9	8.4
Symptomatic with oxygen and intermittent no-invasive ventilation	2	1.9
Symptomatic with continuous no-invasive ventilation	0	0.0
Symptomatic with invasive ventilation	5	4.6
Symptomatic at ECMO	0	0.0
Not reported	7	6.5
Listed for lung transplantation		
Yes	10	9.3
No	90	84.1
Not reported	7	6.5
Clinical course of lung disease (since last visit)		
Improvement	15	14.0
Stable in condition	40	37.4
Deterioration	16	15.0
Not reported	36	33.6
Disease-specific treatment (last six weeks)		
Yes	44	41.1
No	55	51.4
Not reported	8	7.5
Type of medication		
Steroid pulse (systemic)	16	15.0
Steroids (chronic, systemic)	9	8.4
Acithromycin	11	10.3
Hydroxychloroquine	9	8.4
Antibiotics	23	21.5
Other	13	12.1

^{A)}Note: Subjects' medical information is based on physicians' reports. ^{A)} Application of a Novel Classification Scheme.

training may help to ensure that HCPs have the appropriate education stance and ability to tailor patient education to patients' and/or caregivers' needs [14,38].

Pre-post effect sizes were found to be between low and moderate. Assuming a high emotional burden due to the uncertain prognosis and consequent high information needs of patients and parents, this seems satisfactory. In line with Smith et al. patients and their parents who were included in our study about 24 months after the diagnosis with a confirmed disease specific diagnosis seem to have learned to manage the disease [56]. Additionally, as found by other studies [57,58] it is possible that family adaptation and resilience within our study population

allows positive adaptation to adverse health states. This might lead to acceptable HrQoL for most children and adolescents with chronic conditions.

The drop-out rate of 9.4% (n=10) immediately after the consultation, and of 19.6% (n=21) at the four weeks follow-up was low and indicated a high commitment of the participating families to fill-in follow-up assessments. This might be due to a strong relationship between the families and the clinic staff at both centers. In agreement with Carpenter et al. [32] our findings suggest that a good child-provider communication has the potential to positively affect outcomes of children with chILD and their caregivers.

Table 3
Socio-demographic information of the educating physician.

Characteristics	N (19)	(%)
Age (years)		
mean	36.79	
SD	6.85	
Span	26-57	
Gender		
female	10	52.6
male	9	47.4
Level of education (medical qualification)		
pediatric fellow	2	10.5
pediatrician	14	73.7
Pediatrician training in pediatric pneumology	11	57.9
Pediatrician, certified as pediatric pneumologist	5	26.3
Additional medical certification (Allergology)	1	5.3
Number of educational sessions per physician		
1 patient	11	57.9
2-10 patients	6	31.6
11-20 patients	1	5.3
> 20 patients	1	5.3

4.2. Limitations

Results from this pilot study need to be interpreted cautiously given its many limitations. First, the single group pre-post study design makes it difficult to identify specific effects of the patient education-program. Second, in the area of rare diseases it was not possible to recruit a sufficient sample of patients with chILD for a randomized controlled trial study design which included a control and an intervention group. Consequently, in absence of a control group it is usually difficult to identify the specific effects which are

solely due to our intervention. Third, we found a large standard deviation in all variables. Fourth, the stability of our effects over time was not evaluated. Fifth, we included both cases with confirmed, and also with suspected chILD-diagnoses. Hence, we cannot exclude the possibility that cases with confirmed diagnoses at study admission might have had more disease specific knowledge than cases without a confirmed diagnosis. Sixth, there was no evaluation of whether clinicians benefited from the train-the-trainer inhouse session. Seventh, the questionnaire to assess our secondary outcome satisfaction of HCPs with the patient education-program showed low internal consistencies. Finally, there were also ceiling effects in all of our outcome measures, which prevented our pilot study from detecting higher improvements. On average, participants showed relatively high scores of self-efficacy, satisfaction with consultations, and HrQoL already at baseline, leaving only limited room for improvement by participating in the patient education-program. This might be due to the selection of our study population and the involvement of specialized clinics with high levels of expertise regarding rare diseases, which might not be representative of other centers.

4.3. Conclusion

The patient-education program was feasible to conduct for a physician in a clinical context by adding 20 to 30 min to a routine clinic encounter to provide education. Furthermore, the program was successfully implemented in two German tertiary centers for pediatric pulmonology and provided preliminary evidence that a structured patient-education program for children/adolescents with chILD and their parents improves self-efficacy and satisfaction with consulting. Contribution of additional factors, like

Table 4
Means, standard deviations and results of paired t-tests of primary and secondary outcomes, as well as effect sizes.

Outcome	N	Range	Baseline		Follow-up		t	p	Cohens d (pre-post)
			Mean	SD	Mean	SD			
Primary outcomes ^{b)}									
Self-efficacy									
caregiver report	101	16.00-48.00	40.20	7.41	41.68	6.38	3.03	0.003	0.21
self-report	38	14.00-48.00	34.82	7.60	38.32	6.77	2.89	0.006	0.49
Secondary outcomes									
Satisfaction with patient education ^{a)}									
caregiver report	98	11.00-38.00	33.61	4.67	35.88	3.84	6.38	<0.001	0.53
self-report	38	20.00-38.00	31.45	3.96	34.05	4.69	3.56	0.001	0.60
Health-related Quality of Life ^{b)}									
chILD-QoL (caregiver report)	92	25.00-100.00	80.40	15.64	80.98	14.46	0.47	0.64	0.04
chILD-QoL (self-report)	38	56.82-100.00	82.41	12.09	82.82	12.46	0.34	0.74	0.03

Abbreviations: T1 = baseline assessment before counselling ^{a)} T2=follow-Up 1 immediately after counselling ^{b)} T3=follow-Up 4 weeks after counselling M = mean SD = standard deviation t = standard score of the paired t-statistic p = p value of the statistical significance test d = : pre-post effect size Cohen's d (formula: d = M1-M2/SD_{pooled})
*Note: pre-post effect size Cohen's d >0.8: large 0.5 - 0.8: moderate <0.2 small.

Table 5
Physician's satisfaction with patient education.

Item	N	Mean	SD	Min	Max
A structured patient education is very helpful.	19	3.56	0.45	3.00	4.00
This structured patient education program is feasible to conduct in clinical practice.	19	2.90	0.51	2.00	4.00
The timeslot between 20 to 30 minutes is sufficient.	19	3.38	0.53	2.40	4.00
Using the patient information booklet is helpful.	19	3.56	0.57	2.00	4.00
The patient information booklet comprised information I did not know before.	19	2.38	1.19	1.00	4.00
I would use the structured patient education program with pediatric patients and their parental caregivers in the future.	19	3.64	0.43	3.00	4.00
My assessment for the "chILD-EU patient education program" reads as follows . . .	19	5.64	0.43	5.00	6.00

spontaneous improvement, learning from other patients and families during participation in self-help groups or internet forums, cannot be excluded. Further studies should investigate the effectiveness of the educational program within a randomized controlled trial including more incident cases and cost-effectiveness analyses as well as using more sensitive instruments. Moreover, more studies are needed to investigate moderators and mediators of the effects of our chILD-specific patient education-program. The implementation of the intervention in further European countries as a part of the chILD-EU consortium is pending. Factors that might have an impact on dissemination and further implantation of patient education-programs should be also be addressed in future studies.

4.4. Practice implications

A standardized approach of HCPs should use written information material in combination with information given during a consultation to arrive at improved shared medical decision making. Information material should be given for homework and contents should be discussed if needed once again as an ongoing process. More feedback of patient-reported outcomes (PROs) and results of patient education given to HCPs seems to keep them motivated [14]. Besides training workshops on patient education skills it's of high importance to make it possible for HCPs to consolidate their skills in clinical routine care [39]. We developed a new patient education-program for a very heterogeneous group of rare diseases. Based on the preliminary results of our pilot study, chILD patient education can be considered as a promising approach to support children and adolescents with chILD as well as their parental caregivers. As a next step, chILD patient education should be further investigated in randomized controlled trials.

Funding source

The research leading to these results has received funding from the [European Union's] Seventh Framework Programme ([FP7/2007–2013]) under grant agreement no [305653].

Financial disclosure statement

The authors declare that they have no competing financial interests in relation to this article.

Conflict of interest statement

The authors declare that they have no conflicts of interests.

Acknowledgments

The authors would like to thank all members of the medical health care team in the two German centers for pediatric pulmonology for their cooperation. In addition, we thank patients and families for participating in the study at the two clinical sites.

Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:<https://doi.org/10.1016/j.pec.2019.01.016>.

References

- [1] B. Bradley, H.M. Branley, J.J. Egan, M.S. Greaves, D.M. Hansell, N.K. Harrison, N. Hirani, R. Hubbard, F. Lake, A.B. Millar, W.A. Wallace, A.U. Wells, M.K. Whyte, M. L. Wilsner, Interstitial lung disease guideline: the british thoracic society in collaboration with the thoracic society of Australia and New Zealand and the irish thoracic society, *Thorax* 63 (Suppl 5) (2008) v1–v58, doi:<http://dx.doi.org/10.1136/thx.2008.101691>.
- [2] H. Thomas, K.A. Risma, T.B. Graham, A.S. Brody, G.H. Deutsch, L.R. Young, P.M. Joseph, A kindred of children with interstitial lung disease, *Chest* 132 (2007) 221–230, doi:<http://dx.doi.org/10.1378/chest.06-2476>.
- [3] A. Bush, S. Cunningham, J. de Blic, A. Barbato, A. Clement, R. Epaud, M. Hengst, N. Kiper, A.G. Nicholson, M. Wetzke, D. Snijders, N. Schwerk, M. Griesse, European protocols for the diagnosis and initial treatment of interstitial lung disease in children, *Thorax* 70 (2015) 1078–1084, doi:<http://dx.doi.org/10.1136/thoraxjnl-2015-207349>.
- [4] R. Dinwiddie, N. Sharief, O. Crawford, Idiopathic interstitial pneumonitis in children: a national survey in the United Kingdom and Ireland, *Pediatr. Pulmonol.* 34 (2002) 23–29, doi:<http://dx.doi.org/10.1002/ppul.10125>.
- [5] M. Griesse, M. Haug, F. Brasch, A. Freihorst, P. Lohse, R. von Kries, T. Zimmermann, D. Hartl, Incidence and classification of pediatric diffuse parenchymal lung diseases in Germany, *Orphanet J. Rare Dis.* 4 (2009) 26, doi:<http://dx.doi.org/10.1186/1750-1172-4-26>.
- [6] A. Bush, G. Anthony, A. Barbato, S. Cunningham, A. Clement, R. Epaud, C. Gilbert, L. Goldbeck, K. Kronfeld, A.G. Nicholson, N. Schwerk, M. Griesse, Research in progress: put the orphanage out of business, *Thorax* 68 (2013) 971–973, doi:<http://dx.doi.org/10.1136/thoraxjnl-2012-203201>.
- [7] K.C. Meyer, Diagnosis and management of interstitial lung disease, *Transl. Respir. Med.* 2 (2014) 4, doi:<http://dx.doi.org/10.1186/2213-0802-2-4>.
- [8] S. Das, C. Langston, L.L. Fan, Interstitial lung disease in children, *Curr. Opin. Pediatr.* 23 (2011) 325–331, doi:<http://dx.doi.org/10.1097/MOP.0b013e3283464a37>.
- [9] G.H. Deutsch, L.R. Young, R.R. Deterding, L.L. Fan, S.D. Dell, J.A. Bean, et al., Diffuse lung disease in young children: application of a novel classification scheme, *Am. J. Respir. Crit. Care Med.* 176 (2007) 1120–1128, doi:<http://dx.doi.org/10.1164/rccm.200703-3930C>.
- [10] M. Niemitz, N. Schwerk, L. Goldbeck, M. Griesse, Development and validation of a health-related quality of life questionnaire for pediatric patients with interstitial lung disease, *Pediatr. Pulmonol.* 53 (2018) 954–963, doi:<http://dx.doi.org/10.1002/ppul.24018>.
- [11] A.C. Coelho, M.M. Knorst, M.B. Gazzana, S.S.M. Barreto, Predictors of physical and mental health-related quality of life in patients with interstitial lung disease: a multifactorial analysis, *J. Bras. Pneumol.* 36 (2010) 562–570, doi:<http://dx.doi.org/10.1590/S1806-37132010000500007>.
- [12] A.C. Modi, A.L. Pai, K.A. Hommel, K.K. Hood, S. Cortina, M.E. Hilliard, S.M. Guilfoyle, W.N. Gray, D. Drotar, Pediatric self-management: a framework for research, practice, and policy, *Pediatrics* 129 (2012) e473–e485, doi:<http://dx.doi.org/10.1542/peds.2011-1635>.
- [13] M.J. McCarthy, R. Herbert, M. Brimacombe, J. Hansen, D. Wong, M. Zelman, Empowering parents through asthma education, *Pediatr. Nurs.* 28 (2002) 465–473.
- [14] S. Lelorain, A. Bachelet, N. Bertin, M. Bourgoin, French healthcare professionals' perceived barriers to and motivation for therapeutic patient education: a qualitative study, *Nurs. Health Sci.* 19 (2017) 331–339, doi:<http://dx.doi.org/10.1111/nhs.12350>.
- [15] L.K. Bartholomew, D.I. Czyzewski, G.S. Parcel, P.R. Swank, M.M. Sockrider, M.J. Mariotto, D.V. Schidlow, R.J. Fink, D.K. Seilheimer, Self-management of cystic fibrosis: short-term outcomes of the cystic fibrosis family education program, *Health Educ. Behav.* 24 (1997) 652–666, doi:<http://dx.doi.org/10.1177/109019819702400511>.
- [16] M. Zwerink, M. Brusse-Keizer, P.D. van der Valk, G.A. Zielhuis, E.M. Monnikhof, J. van der Palen, P.A. Frith, T. Effing, Self management for patients with chronic obstructive pulmonary disease, *Cochrane Database Syst. Rev.* (2014), doi:<http://dx.doi.org/10.1002/14651858.CD002990.pub3> CD002990.
- [17] E. Savage, P.V. Beirne, M.N. Chroinin, A. Duff, T. Fitzgerald, Self-management education for cystic fibrosis, *Cochrane Database Syst. Rev.* (2009), doi:<http://dx.doi.org/10.1002/14651858.CD007641.pub3>.
- [18] F. Wolf, J. Guevara, C. Grum, N. Clark, C. Cates, Educational interventions for asthma in children, *Cochrane Database Syst. Rev.* (2008) 1–79, doi:<http://dx.doi.org/10.1002/14651858.CD000326>.
- [19] F. Gallefoss, P.S. Bakke, Impact of patient education and self-management on morbidity in asthmatics and patients with chronic obstructive pulmonary disease, *Respir. Med.* 94 (2000) 279–2887, doi:<http://dx.doi.org/10.1053/rmed.1999.0749>.
- [20] E.C. Devine, J. Pearcy, Meta-analysis of the effects of psychoeducational care in adults with chronic obstructive pulmonary disease, *Patient Educ. Couns.* 29 (1996) 167–178, doi:[http://dx.doi.org/10.1016/0738-3991\(96\)00862-2](http://dx.doi.org/10.1016/0738-3991(96)00862-2).
- [21] T. Jonsson, H.E. Ekvall, C.A. Thorstensson, F. Eek, P. Bergman, L.E. Dahlberg, The effect of education and supervised exercise on physical activity, pain, quality of life and self-efficacy - an intervention study with a reference group, *BMC Musculoskelet. Disord.* 19 (2018) 198, doi:<http://dx.doi.org/10.1186/s12891-018-2098-3>.
- [22] S.Y. Chen, S. Sheu, C.S. Chang, T.H. Wang, M.S. Huang, The effects of the self-efficacy method on adult asthmatic patient self-care behavior, *J. Nurs. Res.* 18 (2010) 266–274, doi:<http://dx.doi.org/10.1097/NRj.0b013e3181f3e33f>.
- [23] L. Sharifi, Z. Pourpak, H. Heidarnazhad, S. Bokaie, M. Moin, Asthma knowledge, attitude, and self-efficacy in Iranian asthmatic patients, *Arch. Iran. Med.* 14 (2011) 315–320.
- [24] F. Blackstock, K. Webster, Disease-specific health education for COPD: a systematic review of changes in health outcomes, *Health Educ. Res.* 22 (2007) 703–717, doi:<http://dx.doi.org/10.1093/her/cyl150>.

- [25] E.D. Bateman, S. Hurd, P.J. Barnes, J. Bousquet, J.M. Darzen, M. Fitzgerald, P. Gibson, K. Ohta, P. O'Byrne, E. Pedersen, E. Pizzichini, S.D. Sullivan, S.E. Wenzel, H.J. Zar, Global strategy for asthma management and prevention: GINA executive summary, *Eur. Respir. J.* 31 (2008) 143–178, doi:http://dx.doi.org/10.1183/09031936.00138707.
- [26] P.G. Gibson, H. Powell, A. Wilson, M.J. Abramson, P. Haywood, A. Bauman, M.J. Hensley, E.H. Walters, J.J.L. Roberts, Self-management education and regular practitioner review for adults with asthma, *Cochrane Database Syst. Rev.* (2009), doi:http://dx.doi.org/10.1002/14651858.CD001117 CD001117.
- [27] J.J. Klein, J. van der Palen, S.M. Uil, G.A. Zielhuis, E.R. Seydel, C.L.A. van Herwaarden, Benefit from the inclusion of self-treatment guidelines to a self-management programme for adults with asthma, *Eur. Respir. J.* 17 (2001) 386–394.
- [28] J. McDougal, A. Gettys, J.S. Hagood, Child family education, *Pediatr. Allergy Immunol. Pulmonol.* 23 (2010) 87–90, doi:http://dx.doi.org/10.1089/ped.2010.0005.
- [29] K.T. Petsios, K.N. Priftis, E. Hatzigiorgou, J.N. Tsanakas, G. Antonogeorgos, V.N. Matziou, Determinants of quality of life in children with asthma, *Pediatr. Pulmonol.* 48 (2013) 1171–1180, doi:http://dx.doi.org/10.1002/ppul.22768.
- [30] P.K. Gandhi, K.M. Kenzik, L.A. Thompson, D.A. DeWalt, D.A. Revicki, E.A. Shenkman, L.C. Huang, Exploring factors influencing asthma control and asthma-specific health-related quality of life among children, *Respir. Res.* 14 (2013) 26, doi:http://dx.doi.org/10.1186/1465-9921-14-26.
- [31] E. Ahmad, D.E. Grimes, The effects of self-management education for school-age children on asthma morbidity: a systematic review, *J. Sch. Nurs.* 27 (2011) 282–292, doi:http://dx.doi.org/10.1177/1059840511403003.
- [32] D.M. Carpenter, G.X. Ayala, D.M. Williams, K.B. Yeatts, S. Davis, B. Sleath, The relationship between patient-provider communication and quality of life for children with asthma and their caregivers, *J. Asthma* 50 (2013) 791–798, doi:http://dx.doi.org/10.3109/02770903.2013.808347.
- [33] L. Goldbeck, A. Fidika, M. Herle, A.L. Quittner, Psychological interventions for individuals with cystic fibrosis and their families, *Cochrane Database Syst. Rev.* (2014), doi:http://dx.doi.org/10.1002/14651858.CD003148.pub3 CD003148.
- [34] L. Goldbeck, C. Babka, Development and evaluation of a multi-family psychoeducational program for cystic fibrosis, *Patient Educ. Couns.* 44 (2001) 187–192, doi:http://dx.doi.org/10.1016/S0738-3991(00)00184-1.
- [35] C. Fournier, A. Gautier, H. Mosnier-Pudat, C. Druet, A. Fagot-Campagna, I. Aujoulat, ENTRED 2007: results of a french national survey on self-management education to people with diabetes . . . still a long way to go!, *Ther. Patient Educ.* 6 (2014).
- [36] C. Rey, E. Verdier, P. Fontaine, S. Lelorain, Fostering hospital physicians' involvement in therapeutic patient education: avenues for continuing training and team support, *Ther. Patient Educ.* 8 (2016), doi:http://dx.doi.org/10.1111/nhs.12350.
- [37] F. Friberg, V. Granum, A.L. Bergh, Nurses' patient-education work: conditional factors - an integrative review, *J. Nurs. Manage.* 20 (2012) 170–186, doi:http://dx.doi.org/10.1111/j.1365-2834.2011.01367.x.
- [38] H. Pinnock, E. Epiphaniou, G. Pearce, H. Parke, T. Greenhalgh, A. Sheikh, C.J. Griffiths, S.J. Taylor, Implementing supported self-management for asthma: a systematic review and suggested hierarchy of evidence of implementation studies, *BMC Med.* 13 (2015) 127, doi:http://dx.doi.org/10.1186/s12916-015-0361-0.
- [39] H. Pinnock, Supported self-management for asthma, *Breathe Sheff. (Sheff)* 11 (2015) 98–109, doi:http://dx.doi.org/10.1183/20734735.015614.
- [40] H. Pinnock, H.L. Parke, M. Panagioti, L. Daines, G. Pearce, E. Epiphaniou, P. Bower, A. Sheikh, C.J. Griffiths, S.J. Taylor, Systematic meta-review of supported self-management for asthma: a healthcare perspective, *BMC Med.* 15 (2017) 64, doi:http://dx.doi.org/10.1186/s12916-017-0823-7.
- [41] K. Rivera-Spoljaric, M. Halley, S.R. Wilson, Shared clinician-patient decision-making about treatment of pediatric asthma: what do we know and how can we use it? *Curr. Opin. Allergy Clin. Immunol.* 14 (2014) 161–167, doi:http://dx.doi.org/10.1097/ACI.000000000000046.
- [42] M. Zijlstra, C. de Bie, L. Breij, M. van Pieterse, A. van Staa, L. de Ridder, J. van der Woude, J. Escher, Self-efficacy in adolescents with inflammatory bowel disease: a pilot study of the "IBD-yourself", a disease-specific questionnaire, *J. Crohns Colitis* 7 (2013) e375–e385, doi:http://dx.doi.org/10.1016/j.crohns.2013.02.017.
- [43] M. Griesse, E. Seidl, M. Hengst, S. Reu, H. Rock, G. Anthony, N. Kiper, N. Emiralioglu, D. Snijders, L. Goldbeck, R. Leidl, J. Ley-Zaporozhan, I. Kruger-Stollfuss, B. Kammer, T. Wesselak, C. Eismann, A. Schams, D. Neuner, M. MacLean, A.G. Nicholson, M. Lauren, A. Clement, R. Epaud, J. de Blinc, M. Ashworth, P. Aurora, A. Calder, M. Wetzke, M. Kappler, S. Cunningham, N. Schwerk, A. Bush, International management platform for children's interstitial lung disease (chILD-EU), *Thorax* 73 (2018) 231–239, doi:http://dx.doi.org/10.1136/thoraxjnl-2017-210519.
- [44] M. Griesse, A. Irnstetter, M. Hengst, H. Burmester, F. Nagel, J. Ripper, M. Feilcke, I. Pawlita, F. Gothe, M. Kappler, A. Schams, T. Wesselak, D. Rauch, T. Wittmann, P. Lohse, F. Brasch, C. Kroner, Categorizing diffuse parenchymal lung disease in children, *Orphanet J. Rare Dis.* 10 (2015) 122, doi:http://dx.doi.org/10.1186/s13023-015-0339-1.
- [45] O. Siddiqui, M.W. Ali, A comparison of the random-effects pattern mixture model with last-observation-carried-forward (LOCF) analysis in longitudinal clinical trials with dropouts, *J. Biopharm. Stat.* 8 (1998) 545–563, doi:http://dx.doi.org/10.1080/10543409808835259.
- [46] J. Shao, B. Zhong, Last observation carry-forward and last observation analysis, *Stat. Med.* 22 (2003) 2429–2441, doi:http://dx.doi.org/10.1002/sim.1519.
- [47] J. Cohen, *Statistical Power Analysis for the Behavioral Sciences*, second ed, Erlbaum, Hillsdale NJ, 1988.
- [48] K.J. Klauer, *Guide of Cognitive Training [Handbuch Kognitives Training]*, Hogrefe, Göttingen, 2001.
- [49] A. Butz, L. Pham, L. Lewis, C. Lewis, K. Hill, J. Walker, M. Winkelstein, Rural children with asthma: impact of a parent and child asthma education program, *J. Asthma* 42 (2005) 813–821, doi:http://dx.doi.org/10.1080/02770900500369850.
- [50] L. Cicutto, S. Murphy, D. Coutts, J. O'Rourke, G. Lang, C. Chapman, P. Coates, Breaking the access barrier: evaluating an asthma center's efforts to provide education to children with asthma in schools, *Chest* 128 (2005) 1928–1935, doi:http://dx.doi.org/10.1378/chest.128.4.1928.
- [51] B. Velsor-Friedrich, T. Pigott, B. Srof, A practitioner-based asthma intervention program with African American inner-city school children, *J. Pediatr. Health Care* 19 (2005) 163–171, doi:http://dx.doi.org/10.1016/j.pedhc.2004.12.002.
- [52] E.E. Patterson, M.P. Brennan, K.M. Linskey, D.C. Webb, M.D. Shields, C.C. Patterson, A cluster randomised intervention trial of asthma clubs to improve quality of life in primary school children: the School Care and Asthma Management Project (SCAMP), *Arch. Dis. Child.* 90 (2005) 786–791, doi:http://dx.doi.org/10.1136/adc.2004.062612.
- [53] N.L. Young, A.M. Foster, P.C. Parkin, J. Reisman, I. Maclusky, M. Gold, W. Feldman, Assessing the efficacy of a school-based asthma education program for children: a pilot study, *Can. J. Public Health* 92 (2001) 30–34.
- [54] A. Kennedy, E. Nelson, D. Reeves, G. Richardson, C. Roberts, A. Robinson, A. Rogers, M. Sculpher, D. Thompson, A randomised controlled trial to assess the impact of a package comprising a patient-orientated, evidence-based self-help guidebook and patient-centred consultations on disease management and satisfaction in inflammatory bowel disease, *Health Technol. Assess. (Rockv)* 7 (2003) 1–113, doi:http://dx.doi.org/10.3310/hta7280.
- [55] C. Norton, L.B. Dibley, A. Hart, J. Duncan, A. Emmanuel, C.H. Knowles, N. Stevens, H. Terry, A. Verjee, S. Kerry, N. Hounsou, Faecal incontinence intervention study (FINS): self-management booklet information with or without nurse support to improve continence in people with inflammatory bowel disease: study protocol for a randomized controlled trial, *Trials* 16 (2015) 444, doi:http://dx.doi.org/10.1186/s13063-015-0962-0.
- [56] J. Smith, F. Cheater, H. Bekker, Parents' experiences of living with a child with a long-term condition: a rapid structured review of the literature, *Health Expect.* 18 (2015) 452–474, doi:http://dx.doi.org/10.1111/hex.12040.
- [57] A. Payot, K.J. Barrington, The quality of life of young children and infants with chronic medical problems: review of the literature, *Curr. Probl. Pediatr. Adolesc. Health Care* 41 (2011) 91–101, doi:http://dx.doi.org/10.1016/j.cppeds.2010.10.008.
- [58] S. Saetes, L. Hynes, B.E. McGuire, L. Caes, Family resilience and adaptive coping in children with juvenile idiopathic arthritis: protocol for a systematic review, *Syst. Rev.* 6 (2017) 221, doi:http://dx.doi.org/10.1186/s13643-017-0619-z.