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Review

Transition to adult care in epilepsy: A systematic review

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A B S T R A C T

The transfer from paediatric to adult care can be a complex process in children with epilepsy. Inadequate care during this phase can affect long-term medical and psychosocial outcomes. The aim of this study was to review studies on transitional care from paediatric to adult healthcare for young persons with epilepsy in order to synthesize evidence for best practice. We undertook a systematic review following PRISMA guidelines and employed narrative synthesis. A total of 36 articles were included, of which 11 were interventional studies and 25 observational studies. Study quality was rated as 'good' for only four studies. Interventions included joint or multidisciplinary clinics, education (patient and health professional education) and extended service provision (Saturday clinics, peer-groups). All studies observed a positive effect experienced by the participants, regardless of intervention type. Observational studies showed that transition plans/programmes are asked for but frequently not existing or not adapted to subgroups with intellectual disability or other neurodevelopmental conditions. The results of this systematic review on transitional care in epilepsy suggest that a planned transition process likely enhances medical and psychosocial outcomes for young people with epilepsy, but the body of evidence is limited and there are significant gaps in knowledge of what efficacious transition constitutes. More studies are needed employing qualitative and quantitative methods to further explore the needs of young people with epilepsy and their families but also robust study designs to investigate the impact of interventions on medical and psychosocial outcomes.

1. Introduction

Every year, around 1.1 million children with epilepsy become adults [1,2]. This occurs during adolescence, an important stage for physical and psychological development. While this phase of life is challenging for anyone, young persons with epilepsy (YPE) often have extra difficulties such as social stigma, mental health and neurodevelopmental issues, isolation, and perceived lack of independence [3–5]. YPE have worse psychosocial outcomes in adulthood, even those with pharmacoresponsive epilepsy syndromes associated with intelligence quotients (IQ) within the normal range [6,7]. Modifiable risk factors during adolescence which influence psychosocial outcome in adulthood are: high seizure frequency, unsupportive and unstable family environment, and polypharmacy [7,8]. It is, therefore, important that YPE receive optimal care during adolescence and are adequately prepared for the subsequent transition to adult care [9].

The transfer from paediatric healthcare to adult healthcare is a problematic process in many chronic diseases [10,11]. For YPE, this process is often more difficult because of a high frequency of neurodevelopmental and mental health problems [12,13]. In addition, epilepsy often influences aspects of adolescence such as social life, sexuality, pregnancy, driving, employment and autonomy [14,15]. Furthermore, epilepsy is a heterogeneous disease ranging from severe therapy resistant epilepsy to well controlled epilepsy with normal cognitive functioning. In YPE with intellectual disability there is little expectation for independent living. Thus goals during transition will be different than those with normal cognitive functioning, who are expected to take responsibility for their own healthcare after transition into adulthood [16]. Inadequate transitional care can lead to discontinuation of care and as a result worse somatic and psychosocial outcomes [8,17]. A tailored and well-planned transition from paediatric to adult health care services is therefore paramount for YPE.

The term 'transfer' is the formal handing over from paediatric to adult health care. Often, a referral letter is written to an adult care provider, the patient is seen, and responsibility for care is transferred. The term 'transition' is a process to prepare children, young people,

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families and physicians in the transfer [18]. In the transition process, the maturity of the patients and their knowledge of their disorder has to be taken into account, and appropriate medical information has to be communicated. Along the way, this process can be used as a unique opportunity to get YPE more actively involved in their own care which can also help improve their self-esteem. There has been increasing interest in transitional care for YPE in the last two decades and there is consensus on the importance of an adequate transition [9]. However, little is known about the key features of transitional care and the outcomes which define a successful transition [12,19].

The goal of transitional care in YPE is to improve the individual experience with health care, improve the population health and reduce the per capita costs of care [20]. In this study, we aimed to review and synthesise the available research on the transition process from paediatric to adult healthcare in YPE. An additional aim was to consider the impact of study quality on the findings.

2. Methods

A systematic review method was employed to identify available studies. Due to the heterogeneous study designs, interventions and outcome measures, we employed a 'narrative synthesis' [21,22] to describe and interpret findings. Studies were grouped into observational and interventional studies. We considered also the implications of quality on findings. The protocol for this systematic review was registered prior to commencement on PROSPERO and can be accessed at https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=25000.

2.1. Search strategy and selection criteria

We searched the following databases on 1 May 2021: the Cochrane Central Register of Controlled Trials (CENTRAL), PubMed, CINAHL and PsycInfo. Search terms were related to transition to adult care, adolescence, and epilepsy. Excluded were reviews, guidelines, dissertations, books, letters, editorials and trial registrations without original patient data and articles not in English. See supplemental data 1 and 2 for the inclusion criteria and the searches. The title and abstract of all results were screened independently by two authors (RG and CR) using prior set selection criteria (supplemental data 1). For this, Covidence, a web-based software platform for screening of abstracts and full texts in systematic reviews was used (Covidence systematic review software, Veritas Health Innovation, Melbourne, Australia. Available at www.covidence.org). In case of discrepancy after discussion a third author (IO) was consulted to resolve disagreements and reach consensus.

2.2. Data synthesis and quality assessment

Our data extraction revealed a high variation in data analysis methods from diverse outcome measures in both interventional and observational studies. The results were thus grouped for narrative synthesis in (I) interventional studies, (II) observational studies and (III) observational studies with epilepsy professionals as participants. Narrative synthesis was chosen because of the high degree of variation in data common in observational studies and with respect to a wide variation of study outcome measures [22]. Two reviewers (RG and CR) independently completed quality appraisals for all included studies following the Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies by the National Heart, Lung, and Blood Institute; National Institutes of Health; U.S. Department of Health and Human Services (https://www.nhlbi.nih.gov/health-topics/study-quality-assessment-tools). Risk of bias across selection, data collection, effect measures and reporting was assessed and rated as poor, fair or good according to number of positive responses.

3. Results

3.1. Search results

A total of 5752 records were found in our search across four databases and 5266 unique papers were subsequently screened after removal of duplicates. Eighty-one full text papers were screened and 36 articles met eligibility criteria and were included in this review (see Fig. 1).

3.2. Study design and quality assessment

The study designs of the included articles were: interventional (n = 11), observational/cross-sectional studies with patients (n = 18) and cross-sectional studies with medical care professionals (n = 8). One study reported both observational and interventional data [24]. Very few studies had a clearly stated sample size calculation, exposure level or outcome measure and exposure levels and loss to follow-up were mostly 'not applicable'. We therefore summarised the NIH Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies to poor-fair-good based on number of yes responses [25]. In the overall rating, four studies had 'good' quality, 20 studies had 'fair quality' and 12 studies had 'poor' quality. None of the studies reported a sample size calculation or used blinding, less than half reported standardised exposure and outcome measures and five studies adjusted for confounders. For complete results of the quality assessment see Supplementary data 3.

3.3. Results of narrative synthesis

3.3.1. Interventional studies

A total of eleven interventional studies were reported (Table 1). There were three broad categories of interventions, which were directed at: (I) joint clinic (paediatric neurologist (PN) and adult neurologist (AN)) or multidisciplinary clinic with paediatric/adult neurologist/

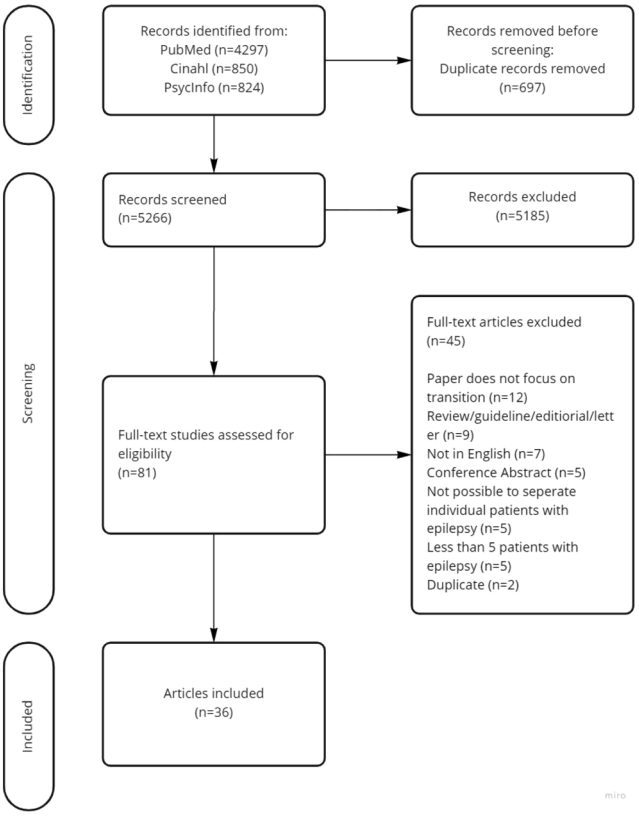

Fig. 1. Flow-chart of the inclusion process following PRISMA guidelines [23].

Table 1
Interventional studies on transitional care in epilepsy patients with main outcomes.

First author (year) *country*	Design	No. patients	Mean age (range)	Epilepsy subtype	Intervention	Outcomes	QA
Appleton [28] (1997) *UK*	IS	120	16 [13–21]	All	Multidisciplinary clinic	Diagnosis revision in 10% to non-epileptic disorder ASM alteration in 22%	Fair
Chiron [29] (2014) *France*	IS	39	22 [16–32]	Severe epilepsies	AN – PN conference	ASM alteration in 67%, of which 62% improvement of seizure frequency.	Poor
Geerlings [30] (2016) *NL*	IS	117	18 [15–25]	All	Multidisciplinary clinic	Diagnosis revision in 12%, ASM alteration in 54%, referred for surgical assessment 6%. Increase of social work assistance 26%	Good
Disabato [76] (2015) *USA*	IS	32	18 [15–25]	All (no ID)	Patient and provider education	Increase of social work referrals 22% No measured change of patient activation in self-care.	Fair
Andreoli [75] (2021) *USA*	IS	38	16 [12–20]	All	Patient and provider education	Increased knowledge of patients and providers after education.	Fair
Geerlings (2016) *NL*	IS	66	21 [16–26]	All	Multidisciplinary clinic 2 year follow-up	Combined transition interventions predict 56% improved medical outcome.	Good
Jurasek (2010) *Canada*	IS	97	[16–18]	All	Multidisciplinary clinic	Transition clinic experienced as beneficial. Decrease of fears associated with moving to adult programme.	Fair
Le Marne (2018) *Australia*	IS	45	16	All	Multidisciplinary clinic 3 month follow-up	Increased knowledge on epilepsy and driving, alcohol/street drugs and birth control/pregnancy. No improvement in self-reported medication adherence, transition readiness or mental well-being.	Good
Lewis [27] (2013) *UK*	NRCCT	16 *	[14–19]	No ID	Multidisciplinary clinic vs. direct transfer	More positive engagement with adult healthcare and retention of epilepsy-related self-care information. Positive factors: continuity of care, on-going and consistent age-appropriate and person centred communication and repeated information exchange.	Fair
Smith [77] (2002) *UK*	IS	207	17 [11–22]	All	AN – PN conference	Transition readiness correlated positively with age, knowledge, and negatively with cognitive problems, expectations, inattention, odd behaviors, and communication problems.	Fair
Crowley [24] (2018) *Ireland*	IS	34	[12–18]	No ID	Saturday transitional care review clinic and transitional and adolescence group	Qualitative outcomes such as "It's good to hear that we all have the same fears and anxiety about our children with epilepsy" and "I feel very comfortable talking about epilepsy here."	Poor

#: study number correlated to supp 2. * number of patients in study group (number of patients in control group). ¤ Mean age in years or interval if mean was not provided.
IS = interventional study without concurrent controls, NRCCT = Non randomized case-controlled trial. ID = intellectual disability, AN = adult neurologist, PN = paediatric neurologist, ASM = anti-seizure medication, QA = quality assessment.

epilepsy nurse/neuropsychologist/social worker/occupational therapist), (II) education (patient and health professional education), and (III) increased service provision (Saturday clinics, peer-groups). All included studies had interventions tailored to their own centre. One study [26] systematically evaluated medical outcomes two years after their multidisciplinary care clinic and one study [27] used a control group. The age range of included patients was between 13-32 years, highlighting that the transition to adult care often takes place after the age of 18 years.

The study with a 2-year follow-up [26] showed that 56% of the improved medical outcome was attributable to their transition clinic. In a case-controlled study Lewis et al. [27] showed that YPE who attended a multidisciplinary clinic experienced more positive engagement with adult healthcare and better retention of epilepsy-related self-care information than those who did not.

A joint clinic or multidisciplinary clinic led to a change in diagnosis in 10-12% of cases and a change in anti-seizure medication (ASM) in 22-67% [28–30] of cases. The change in diagnosis was often to non-epileptic disorders such as psychogenic non-epileptic seizures (PNES) and vasovagal attacks. Multidisciplinary teams led to an increase of social work referrals (22-26%). One study did not detect a change in self-care or medical adherence after multidisciplinary clinic attendance [31]. The studies which reported qualitative results [24,27,32] indicate that a positive effect was experienced by the participants regardless of intervention type. Specifically, the increase of knowledge, positive experiences, decrease of fears and recognition in having the disease were mentioned.

3.3.2. *Observational studies with patients*

Eighteen observational studies reported on YPE and transition: two case-control studies, four cohort studies and twelve cross-sectional studies (Table 2). Studies including medical record reviews and non-standardised questionnaires showed that a minority (range 0-39% in different subgroups) of YPE experienced a transitional discussion/programme and that many persons with epilepsy aged >18 continued at their paediatric clinic. It was frequently mentioned by YPE and/or their caregivers that the transition process often began too late; between 12 and 16 years would be ideal, according to interviewed caregivers of YPE [24]. Seven studies focused on specific subgroups of YPE, mainly YPE without intellectual disability or specific epilepsy syndromes often with severe intellectual disability, where often the parents were included in the study.

Behavioural, cognitive, and emotional problems affected the transition process more than epilepsy per se according to many YPE, their parents and healthcare professionals [5,33–36]. Specifically anxiety and depression were frequent and caused distress during the transition process [37,38]. Another regularly mentioned problem was that transition programmes were not adjusted to specific subgroups of YPE such as persons with cognitive impairments/intellectual disability or behavioural problems [34,35,39].

Factors which positively influenced the transition process were: a stable medical condition, longer relationship with paediatric neurologist, parental advocacy, and the quality of preparation. Geerlings et al. [5,26] showed that lower intelligence, higher seizure frequency, ongoing seizures and an unsupportive and unstable family environment negatively influenced psychosocial outcome. Additionally, loneliness was associated with a failed transition into independence [5]. The study by Hargreaves et al. [40] reported on a decreased all-cause mortality in adolescents with epilepsy in units where all eligible patients were seen by a tertiary paediatric neurologist versus adolescents in units where patients were only seen by paediatricians and epilepsy nurses This study

Table 2
Observational studies on transitional care in epilepsy patients with main outcomes.

First author (year) *country*	No. patients	Cohort	Methods	Outcomes	QA
Hargreaves [40]	1164	Country-based all epilepsy. PN Contact vs. no PN contact.	CC:	• Absolute reduction of 6% in total mortality risk and 6% post-transition mortality risk between units where all versus no eligible patients were seen by a PN.	Good
(2019) *UK*			Longitudinal data-linkage	• Decrease of access to epilepsy specialist nurse increased epilepsy admissions.	
Reeve [41]	36(31)*	YPE without ID vs. healthy controls	CC:	• No differences on self-esteem, affect and self-efficacy, or measures of psychological adjustment.	Poor
(2002) *UK*			NSQ	• Epilepsy significantly more non-productive coping than control group.	
Baca [42]	308	Community based all epilepsy	CS:	• 31% had a "transition discussion".	Fair
(2018) *USA*			NSQ	• 50% of patients >18 y were being seen by an AN.	
Bar [33]	60	(parents of/) patients with TSC	CS:	• 50% with a normal intellectual development had clear knowledge about their disease.	Poor
(2019) *France*			NSQ	• Unmet needs for psychiatric and behavioral disorders during transition	
Boyce [34]	18	Parents of children with DS	CS:	• 71% of adult patients being seen by PN. Transition programmes began too late.	Poor
(2020) *USA*			Focus-groups	• Existing transition programmes described as "not for our kids." • AN perceived as being uncomfortable with intellectual disabilities and challenging behaviors.	
Crowley [24]	34	Parents of children with all epilepsies	CS:	• 82% wants transition introduction between 12–16 years.	Poor
(2018) *Ireland*			NSQ	• 97% wants peer-support group for their children. Parents want more encouragement for YPE taking their own medication.	
Cui [43]	1238	Tertiary hospital based, all epilepsies	CS: participation coping [44] questionnaire	• Lower participation in healthcare behavior compared to norm. • Participation was positively associated with being female, a longer course of disease, fewer comorbidities, and living in a nuclear family.	Fair
Cui [45] (2022) *China*	1239	Tertiary hospital based, all epilepsies		• Self-care status of adolescents related to coping style and family resilience	
Geerlings(5) (2019) *NL*	59	Tertiary hospital, no ID	CS: Loneliness [46] questionnaire	• 32% failed transition to independence. • Correlation between failed transition to independence and: loneliness, low satisfaction with parent relation and friendships.	Fair
Geerlings [8] (2015) *NL*	138	Tertiary hospital based, all epilepsies	Cohort: Patient record review	• Long-term worse psychosocial outcome associated with lower intelligence, higher seizure frequency, ongoing seizures, and an unsupportive and unstable family environment.	Fair
Gray [37]	60	Tertiary hospital based, all epilepsies	CS: Knowledge on own condition [47]	• YPE knowledge increased throughout adolescence, whilst caregivers' knowledge decreased.	Poor
(2017) *UK*			Anxiety [48]	• 30% YPE high levels of anxiety and depression.	
Kuchenbuch [49]	51	Adults with DS	CS:	• 61% had a "transition discussion"	Fair
(2013) *France*			NSQ	• Positive transition factors: quality of preparation, longer relationship with PN, availability of the PN, age >18 y, and a stable medical condition before transfer.	
Schultz [35]	7	Parents of YPE and ID	CS:	• None had received any transition planning	Fair
(2012) *USA*			Qualitative interview	• Positive transition factors: establishing an interpersonal parent-provider relationship, parental advocacy, and networking. • Not epilepsy but cognitive impairments affected transition most.	
Smith [50]	82	Tertiary hospital based, all epilepsies	CS: Transition readiness [51]	• More transition readiness associated with older age, fewer cognitive problems, better knowledge, better executive functioning/attention, fewer atypical behaviors, and better functional communication skills.	Fair
(2021) *USA*					
Warnell [36]	10	Community based, all epilepsies	CS:	• YPE with behavior/cognitive/emotional problems experienced more distress during transition.	Poor
(1998) *Canada*			NSQ	• Transition readiness is not solely age dependent	
Kossof [52]	10	All YPE on DT	Cohort:	• Patients who received care from Adult Epilepsy Diet Centre more likely to remain on dietary therapy compared to PN/AN care.	Fair
(2013) *USA*			Patient record review		
Kwack [38]	140	Tertiary hospital based, all epilepsies	Cohort:	• Diagnosis revision after transfer in 6%.	
(2022) *Korea*			Patient record review	• High comorbidity among transferred YPE: ID 27%, ADHD 15%, depression 29%. • 35% improvement of seizure frequency in 12 months following transfer.	
Oguni [39]	220	Secondary hospital based, all YPE	Cohort:	• 79% patients with developmental and epileptic encephalopathies had pharmacoresistant seizures which was associated with a late transition age.	Fair
(2021) *Japan*			Patient record review		

* = number of patients (number of healthy controls). CC = case-control study, CS = cross-sectional study, Cohort = cohort study, NSQ = non-standardized questionnaires, TSC = tuberous sclerosis complex, DS = Dravet syndrome, DT = dietary treatment, NSQ = non-standardized questionnaire, SQ = standardized questionnaire, ID = intellectual disability, ADHD = attention deficit hyperactivity disorder, AN = adult neurologist, PN = paediatric neurologist.

also showed a reduction in hospital admissions in units where access to an epilepsy specialist nurse increased.

3.3.3. *Cross-sectional studies with medical care professionals*

A total of eight cross-sectional studies focused on the experiences of medical care professionals on transition for YPE were identified (Table 3). All studies used a non-standardised questionnaire aimed at neurologists and other medical care professionals. In one study, medical doctors in training were interviewed [53].

An epilepsy transition plan/programme existed in 11-83% of the clinics. YPE were frequently followed by paediatric neurologists long after their 18th birthday, there were even patients older than 50 years seen by a paediatric neurologist. Factors associated with paediatric clinic contact after the age of 18 years were personal attachment between the paediatric neurologist and families and perceived inferior care at the adult neurology facilities. Dietary treatment was less accepted as a treatment for adults, also causing a barrier to transition.

Table 3
Observational questionnaire studies on medical care professionals and transitional care in epilepsy patients with main outcomes.

First author (year) *country*	No. subjects	Professional category	Main outcomes
Tirol [53] (2021) *USA*	25	NPMP-residents	All residents generally rate their knowledge and confidence at providing transition care to youth with epilepsy to be poor.
Nabbout [54] (2020) *France*	68	AN/PN	• 70% PN followed patients aged over 18 years. • Factors delaying transfer: personal attachment between PN and families, inadequate adult neurology facilities for adolescents with intellectual disability (59% neurologists, 75% paediatric neurologists)
Borlot [55] (2014) *Canada*	115	AN/PN	• 90% requires multidisciplinary care, most frequently: psychiatrist, psychologist, physiotherapist, speech therapist, and occupational therapist. • AN have lower levels of confidence in diagnosing and treating severe forms of childhood-onset epilepsies and epilepsy associated with cognitive delay.
Carrizosa [56] (2020) *Latin America*	117	AN/PN/EN	• 16% had an epilepsy transition plan/programme. • Negative impact on transition: poor education, inflexible healthcare systems, no financial support, no multidisciplinary teams, scarce communication between AD and PN.
Camfield [57] (2011) *Canada*	133	AN/PN/EN	• 45% treated patients over 18 years of age. • Delaying transition factors: personal attachment between PN and families, concern that AN takes less time. • 11% has a transition clinic, 76% has a contact to transition.
Pieters [58] (2021) *USA*	26	AN/PN	• Transfer is experienced as unnatural and disruptive. • Unique needs for epilepsy patients (rare diagnoses, treatment diversity, discordant developmental age) • Clinicians motivated and committed to providing best possible care.
Iyer [59] (2013) *UK*	15	LC¤	83% specific epilepsy transition clinic, waiting time 1-12 months.
Seaborg [60] (2020) *USA*	74	AN/PN	• Dietary therapy more accepted treatment of intractable epilepsy in children (84%) than adults (17%). • 23% have a documented transition plan. • 87% PN feel the lack of sufficient AN who prescribe dietary therapy as a barrier to transferral.

YPE = young people with epilepsy, NPMP residents = neurology, paediatric and medical paediatric residents, AN = adult neurologist, PN = paediatric neurologist, EN = epilepsy nurse. LC = lead epilepsy clinicians in tertiary paediatric neurology centres.

4. Discussion

The results of this systematic review on transitional care in epilepsy suggest that a planned transition process can enhance medical and psychosocial outcomes for young people with epilepsy. However, there is limited evidence and there are significant gaps in knowledge of what efficacious transition for YPE constitutes. Observational studies showed that transition plans/programmes are wanted but often not available and, if present, not adapted to subgroups of YPE such as individuals with intellectual disability. Also, many YPE experienced anxiety and depression which caused distress during the transition process. Interventional studies on transitional care in epilepsy are heterogeneous in design and inconsistent in evaluation of the impact on patient experience, population health and costs. In general, the following factors appear to positively influence the transition process: multidisciplinary care, continuity of care, a central coordinator, education, and patient-centred communication (Fig. 2).

The findings from observational studies on transition for YPE can be synthesised into three messages: (I) only a minority of YPE experienced a transitional discussion/programmes, (II) behavioural, and cognitive problems affected the transition process often more than epilepsy and (III) anxiety and depression were frequent in YPE and had a negative effect on the transition process. It is therefore paramount to consider cognitive, behavioural and emotional functioning during the transition process and initiate screening/assessment in the context of unmet need.

Interventional studies on transitional care in YPE are scarce, heterogeneous in terms of design and outcome measures and of limited quality. A majority of participants in the various studies experienced a positive effect, regardless of the intervention and outcome measure. The intervention was often a 'multidisciplinary clinic', but the number of disciplines and the participants varied widely. The outcome measures were rarely specified, and ranged from strictly medical (i.e. change in anti-seizure medication), to qualitative experiences, and to improved knowledge without further specification. Control-groups were largely absent and the follow-up duration was short. Overall, a positive effect of transitional care on the individual patient experience is suggested, but the quality of the studies precludes clear conclusions.

Our review identified a care gap in transitional programmes for epilepsy. Adequate support during adolescence and the transfer to adult medical care is very important according to the participating YPE, their caregivers and medical professionals. This has been stated in previous literature [12,19], and the findings of this review highlight that this is a clear wish from the young people and their families as well. Moreover, transitional care appears to improve individual care experience in YPE. This is consistent with studies on other chronic diseases, examples include: decreased hospital admission rates and surgeries in patients with inflammatory bowel disease [61], increased survival in cystic fibrosis [62], and protective effect on continuity of care in congenital heart diseases [63]. However, transitional programmes for YPE were often absent, even in high income countries. In low and middle income countries, transition programmes rarely exist [64]. Furthermore, specific subgroups of YPE such as epileptic syndromes with developmental and epileptic encephalopathy may be excluded from regular transition programmes [34]. It can therefore be assumed that the majority of YPE globally do not have access to transitional care. To get a better picture on the current state of transitional care, the results of the ILAE questionnaire will be highly welcomed: (https://www.ilae.org/news-and-media/news-about-ilae/transition-in-epilepsy-a-survey-for-patients-and-healthcare-practitioners).

Another key finding is that subgroups of YPE have different needs during transition. The goal of transitional care for some YPE will be full independence whilst in others it will be a smooth transition to continued dependence. Those with epilepsy and a low developmental age at transition will continue to need comprehensive multidisciplinary care and support. The transition is often more experienced as a necessity driven by the medical and legal system than a positive process.

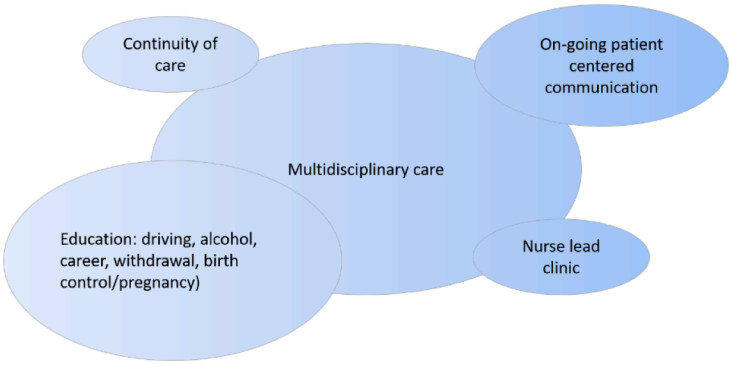

Fig. 2. Successful factors for transition in interventional studies.

Moreover, these YPE experience multiple transitions simultaneously; new contacts for their neurological care, psychological/psychiatric and rehabilitation care, change of school/educational setting, and social support services after childhood, and legal guardianship needs to be considered. Thus, for YPE and intellectual disability, coordination of these different transitions and the clarification of who is responsible for what, appeared generally inadequate and not adjusted to the specific individual. In the literature, alternative effective approaches such as lifespan clinics without transition are found [65]. On the other hand, for YPE with a normal intellectual function without other comorbidities, the transition goal is independence: they are expected to take responsibility when they become adults. Here, the focus is more on increasing disease knowledge and health self-efficacy and decreasing parental dependence. More information, available knowledgeable staff and peer-support possibilities are desired by these YPE and their caregivers, and there is a clear need to start the transition process in a timely manner so that these YPE are ready for adult care when the transferal takes place.

4.1. Limitations

This review of the literature is mainly limited by the quality of data and the heterogeneity of interventions and outcome measures in the included studies. The interventional studies lacked control-groups and clearly defined interventions/outcomes, while observational studies used a myriad of scales and scores as outcomes. Additionally, there is a large geographic bias with a lack of studies from low and middle income countries, which limits the global generalizability of the findings. With regard to quality assessment, none of the studies satisfied all the quality assessment criteria and only four studies were graded as good.

4.2. Future research directions

In spite of the clear recognition of the importance of transitional care, the level of evidence to determine best practice during transition in YPE is currently low. The heterogeneity of interventions and outcome measures highlights the need for well-designed observational and interventional studies with validated outcome measures. Future studies should focus on the triple outcome of improvement of individual patient experience, population health and cost-effectiveness. Information on the cost-effectiveness is essential for the financial outlay and sustainability of transitional programmes [66,67]. Here, inspiration and knowledge comes from other chronic conditions, with well-constructed randomised clinical trials in for example diabetes and spina bifida [68,69] and validated outcome scales in for example congenital heart disease and rheumatic and musculoskeletal diseases [70–72].

4.3. Clinical implications

Despite the limitations, this review can help in guiding transitional care in epilepsy by highlighting key areas of importance. Epilepsy is a heterogeneous disease and transition care builds on local health-care systems. Factors in epilepsy complicating transitional care are: (I) high prevalence of cognitive, behavioural and emotional problems, [13,73], (II) rare conditions underlying epilepsy, and (III) specific treatments such as dietary treatments [60]. Thus, there will not be one universal transition care programme. However, the results of this review suggest that the following features may generally improve transition to adult care in young people with epilepsy:

1 Early start of introduction of transition planning; between ages 12-16 years.
2 Identification and coordination of different transitions in medical and social care
3 Meeting the adult care team before transition.
4 One contact person (often a nurse with epilepsy specialisation) throughout the transition process who is easily accessible.
5 Consideration of possible behavioural and emotional problems during transition.
6 Promoting knowledge of disease and health self-efficacy.
7 Consideration of parental involvement/dependence.
8 Supplementing verbal information with written and visual information (hand-out or digital).
9 Peer-groups for adolescents and parents can be supportive
10 Adapt for YPE with cognitive impairment (hospital visits, waiting room, and invitations) and dietary treatment.

There are similarities between these features and those described in other chronic conditions. For example, appropriate parent involvement, promotion of health self-efficacy, and meeting the adult team before transfer were the three key features associated with better outcomes after transition in type 1 diabetes, cerebral palsy and autism spectrum disorder [74].

4.4. Conclusion

Overall, this review highlighted the need for, and, significant gaps in knowledge on, optimal transitional care in epilepsy. Transitional plans/programmes for the transfer from paediatric to adult care are desired but often not available and, if present, not adapted to subgroups of YPE such as individuals with intellectual disability. A planned transition process may enhance medical and psychosocial outcomes for young people with epilepsy, but the evidence of what efficacious transition for YPE constitutes is limited. Multidisciplinary care, continuity of care, a central coordinator, and patient-centred communication appear to positively influence the transition process. Standardised evaluation of patient experience, population health and cost-effectiveness on transition programmes or interventions are needed.

CRediT authorship contribution statement

Rianne J.M. Goselink: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Visualization, Writing – original draft. **Ingrid Olsson:** Conceptualization, Methodology, Funding acquisition, Investigation, Writing – review & editing. **Kristina Malmgren:** Conceptualization, Methodology, Funding acquisition, Writing – review & editing. **Colin Reilly:** Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Supervision, Writing – review & editing.

Declaration of Competing Interest

None of the authors has any conflict of interest to disclose.

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Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.seizure.2022.07.006.

References

[1] Fiest KM, Sauro KM, Wiebe S, Patten SB, Kwon CS, Dykeman J, et al. Prevalence and incidence of epilepsy: a systematic review and meta-analysis of international studies. Neurology 2017;88(3):296–303.
[2] WHO. 2022 [Available from: https://population.un.org/wpp/DataQuery/.
[3] Healy SA, Fantaneanu TA, Whiting S. The importance of mental health in improving quality of life in transition-aged patients with epilepsy. Epilepsy Behav 2020;112:107324.
[4] MacLeod JS, Austin JK. Stigma in the lives of adolescents with epilepsy: a review of the literature. Epilepsy Behav 2003;4(2):112–7.
[5] Geerlings RPJ, Gottmer-Welschen LMC, Machielse JEM, de Louw AJA, Aldenkamp AP. Failed transition to independence in young adults with epilepsy: The role of loneliness. Seizure 2019;69:207–12.
[6] Nickels K. Seizure and Psychosocial Outcomes of Childhood and Juvenile Onset Generalized Epilepsies: Wolf in Sheep's Clothing, or Well-Dressed Wolf? Epilepsy Curr 2015;15(3):114–7.
[7] Valova V, Kochan A, Werry B, John R, Prager C, Schneider J, et al. Early onset, long illness duration, epilepsy type, and polypharmacy have an adverse effect on psychosocial outcome in children with epilepsy. Neuropaediatrics 2020;51(2):164–9.
[8] Geerlings RP, Aldenkamp AP, Gottmer-Welschen LM, de With PH, Zinger S, van Staa AL, et al. Developing from child to adult: risk factors for poor psychosocial outcome in adolescents and young adults with epilepsy. Epilepsy Behav 2015;51:182–90.
[9] Devinsky O. Transition to adult care for children with epilepsy–a call for action. Epilepsia 2014;55(Suppl 3):54–5.
[10] Betz CL. Approaches to transition in other chronic illnesses and conditions. Pediatr Clin North Am 2010;57(4):983–96.
[11] Stam H, Hartman EE, Deurloo JA, Groothoff J, Grootenhuis MA. Young adult patients with a history of disease: impact on course of life and transition into adulthood. J Adolesc Health 2006;39(1):4–13.
[12] Geerlings RP, Aldenkamp AP, de With PH, Zinger S, Gottmer-Welschen LM, de Louw AJ. Transition to adult medical care for adolescents with epilepsy. Epilepsy Behav 2015;44:127–35.
[13] Watson R, Parr JR, Joyce C, May C, Le Couteur AS. Models of transitional care for young people with complex health needs: a scoping review. Child Care Health Dev 2011;37(6):780–91.
[14] McEwan MJ, Espie CA, Metcalfe J, Brodie MJ, Wilson MT. Quality of life and psychosocial development in adolescents with epilepsy: a qualitative investigation using focus group methods. Seizure 2004;13(1):15–31.
[15] Stefanidou M, Greenlaw C, Douglass LM. Mental health issues in transition-age adolescents and young adults with epilepsy. Semin Pediatr Neurol 2020;36:100856.
[16] Nabbout R, Camfield P. Bringing the transition of epilepsy care from children to adults out of the shadows. Epilepsia 2014;55(Suppl 3):52–3.
[17] Jonsson P, Jonsson B, Eeg-Olofsson O. Psychological and social outcome of epilepsy in well-functioning children and adolescents. A 10-year follow-up study. Eur J Paediatr Neurol 2014;18(3):381–90.
[18] Blum RW, Garell D, Hodgman CH, Jorissen TW, Okinow NA, Orr DP, et al. Transition from child-centered to adult health-care systems for adolescents with chronic conditions. A position paper of the Society for Adolescent Medicine. J Adolesc Health 1993;14(7):570–6.
[19] Burke L, Kirkham J, Arnott J, Gray V, Peak M, Beresford MW. The transition of adolescents with juvenile idiopathic arthritis or epilepsy from paediatric healthcare services to adult health-care services: a scoping review of the literature and a synthesis of the evidence. J Child Health Care 2018;22(3):332–58.
[20] Prior M, McManus M, White P, Davidson L. Measuring the "triple aim" in transition care: a systematic review. Pediatrics 2014;134(6):e1648–61.
[21] Ryan CCaCRG. Cochrane Consumers and Communication Review Group: data synthesis and analysis'. 2013.http://cccrg.cochrane.org (accessed DATE)
[22] Egger M, Schneider M,D, Smith G. Spurious precision? Meta-analysis of observational studies. BMJ 1998;316(7125):140–4.
[23] Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Murow CD, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. BMJ 2021;372:n71.
[24] Crowley SL, Byrne S, McNulty S, Keating K, Nestor T, Owen Y, et al. The Temple Star Transitional Model of Care for epilepsy; the outcome of a quality improvement project. Epilepsy Behav 2018;79:4–8.
[25] 2021. Available from: https://www.nhlbi.nih.gov/health-topics/study-quality-assessment-tools.
[26] Geerlings RP, Aldenkamp AP, Gottmer-Welschen LM, van Staa AL, de Louw AJ. Long-term effects of a multidisciplinary transition intervention from paediatric to adult care in patients with epilepsy. Seizure 2016;38:46–53.
[27] Lewis SA, Noyes J. Effective process or dangerous precipice: qualitative comparative embedded case study with young people with epilepsy and their parents during transition from children's to adult services. BMC Pediatr 2013;13:169.
[28] Appleton RE, Chadwick D, Sweeney A. Managing the teenager with epilepsy: paediatric to adult care. Seizure 1997;6(1):27–30.
[29] Chiron C, An I. Pharmacology aspects during transition and at transfer in patients with epilepsy. Epilepsia 2014;55(Suppl 3):8–11.
[30] Geerlings RP, Aldenkamp AP, Gottmer-Welschen LM, de With PH, Zinger S, van Staa AL, et al. Evaluation of a multidisciplinary epilepsy transition clinic for adolescents. Eur J Paediatr Neurol 2016;20(3):385–92.
[31] Le Marne FA, Towns SJ, Gaskin C, Ho J, Baker R, Beavis E, et al. Implementing a new adolescent epilepsy service: improving patient experience and readiness for transition. J Paediatr Child Health 2019;55(7):819–25.
[32] Jurasek L, Ray L, Quigley D. Development and implementation of an adolescent epilepsy transition clinic. J Neurosci Nurs 2010;42(4):181–9.
[33] Bar C, Ghobeira R, Azzi R, Ville D, Riquet A, Touraine R, et al. Experience of follow-up, quality of life, and transition from pediatric to adult healthcare of patients with tuberous sclerosis complex. Epilepsy Behav 2019;96:23–7.
[34] Boyce DM, Devinsky O, Meskis MA. Barriers to transition from pediatric to adult care for patients with Dravet syndrome: a focus group study of caregivers. Epilepsy Behav 2020;109:107096.
[35] Schultz RJ. Parental experiences transitioning their adolescent with epilepsy and cognitive impairments to adult health care. J Pediatr Health Care 2013;27(5):359–66.
[36] Warnell P. The transition experience of epilepsy patients/families: results of a telephone survey. Axone 1998;20(2):31–3.
[37] Gray V, Palmer L, Whelby K, Vinten J, Gait L. Exploring the role of knowledge of condition and psycho-social profiles of young people with epilepsy during transition. Epilepsy Behav 2017;73:156–60.
[38] Kwack DW, Lee H, Lee R, Kim DW. Treatment outcome following the transition to adult epilepsy care in childhood-onset epilepsy. Seizure 2022;96:43–5.
[39] Oguni H, Ito S, Nishikawa A, Otani Y, Nagata S. Transition from pediatric to adult care in a Japanese cohort of childhood-onset epilepsy: prevalence of epileptic syndromes and complexity in the transition. Seizure 2021;88:1–6.
[40] Hargreaves DS, Arora S, Viveiro C, Hale DR, Ward JL, Sherlaw-Johnson C, et al. Association of quality of paediatric epilepsy care with mortality and unplanned hospital admissions among children and young people with epilepsy in England: a national longitudinal data linkage study. Lancet Child Adolesc Health 2019;3(9):627–35.
[41] Reeve DK, Lincoln NB. Coping with the challenge of transition in older adolescents with epilepsy. Seizure 2002;11(1):33–9.
[42] Baca CM, Barry F, Berg AT. The epilepsy transition care gap in young adults with childhood-onset epilepsy. Epilepsy Behav 2018;87:146–51.
[43] Cui C, Li SZ, Zheng XL, Cheng WJ, Ting W. Participation in healthcare behavior by adolescents with epilepsy and factors that influence it during the transition period: a cross-sectional study in China. Epilepsy Behav 2020;113:107576.
[44] Petersen C, Schmidt S, Bullinger M, Group D. Brief report: development and pilot testing of a coping questionnaire for children and adolescents with chronic health conditions. J Pediatr Psychol 2004;29(8):635–40.
[45] Cui C, Shuang-Zi L, Cheng WJ, Wang T. Mediating effects of coping styles on the relationship between family resilience and self-care status of adolescents with epilepsy transitioning to adult healthcare: a cross-sectional study in China. J Pediatr Nurs 2022;63:143–50.
[46] Hutten E, Jongen EMM, Vos A, van den Hout A, van Lankveld J. Loneliness and mental health: the mediating effect of perceived social support. Int J Environ Res Public Health 2021;18(22).
[47] Jarvie S, Espie CA, Brodie MJ. The development of a questionnaire to assess knowledge of epilepsy: 2–Knowledge of own condition. Seizure 1993;2(3):187–93.
[48] Spence SH. A measure of anxiety symptoms among children. Behav Res Ther 1998;36(5):545–66.

[49] Kuchenbuch M, Chemaly N, Chiron C, Dulac O, Nabbout R. Transition and transfer from pediatric to adult health care in epilepsy: a families' survey on Dravet syndrome. Epilepsy Behav 2013;29(1):161–5.
[50] Smith AW, Gutierrez-Colina AM, Roemisch E, Hater B, Combs A, Shoulberg AM, et al. Modifiable factors related to transition readiness in adolescents and young adults with epilepsy. Epilepsy Behav 2021;115:107718.
[51] Sawicki GS, Garvey KC, Toomey SL, Williams KA, Chen Y, Hargraves JL, et al. Development and validation of the adolescent assessment of preparation for transition: a novel patient experience measure. J Adolesc Health 2015;57(3):282–7.
[52] Kossoff EH, Henry BJ, Cervenka MC. Transitioning pediatric patients receiving ketogenic diets for epilepsy into adulthood. Seizure 2013;22(6):487–9.
[53] Tirol FG, Kumar A. Resident training in transitioning youth with epilepsy into adult care. J Child Neurol 2021;36(1):60–4.
[54] Nabbout R, Teng T, Chemaly N, Breuillard D, Kuchenbuch M. Transition of patients with childhood onset epilepsy: perspectives from pediatric and adult neurologists. Epilepsy Behav 2020;104(Pt A):106889.
[55] Borlot F, Tellez-Zenteno JF, Allen A, Ali A, Snead OC, Andrade DM. Epilepsy transition: Challenges of caring for adults with childhood-onset seizures. Epilepsia 2014;55(10):1659–66.
[56] Carrizosa-Moog J, Isaza-Jaramillo S. Perceptions of adult and child neurologists of transition programs in epilepsy in Latin America: a cross-sectional study. Epilepsy Behav 2020;110:7.
[57] Camfield PR, Gibson PA, Douglass LM. Strategies for transitioning to adult care for youth with Lennox-Gastaut syndrome and related disorders. Epilepsia 2011;52(Suppl 5):21–7.
[58] Pieters H, Watson M, Baca CM. Pediatric and adult epileptologist perspectives and experiences of pediatric to adult epilepsy care transition: "Saying goodbye and opening a door. Epilepsy Behav 2021;120:107981.
[59] Iyer A, Appleton R. Transitional services for adolescents with epilepsy in the U.K.: a survey. Seizure 2013;22(6):433–7.
[60] Seaborg K, Wang X, Olson C, Felton EA. Pediatric to adult transitions of ketogenic dietary therapy for epilepsy. J Child Neurol 2020;35(13):896–900.
[61] Garcia-Rodriguez F, Raygoza-Cortez K, Moreno-Hernandez L, Garcia-Perez R, Garza Lopez LE, Arana-Guajardo AC, et al. Outcomes of transitional care programs on adolescent chronic inflammatory systemic diseases: systematic review and meta-analyses. Pediatr Rheumatol Online J 2022;20(1):15.
[62] Okumura MJ, Kleinhenz ME. Cystic fibrosis transitions of care: lessons learned and future directions for cystic fibrosis. Clin Chest Med 2016;37(1):119–26.
[63] Moons P, Skogby S, Bratt EL, Zühlke L, Marelli A, Goossens E. Discontinuity of cardiac follow-up in young people with congenital heart disease transitioning to adulthood: a systematic review and meta-analysis. J Am Heart Assoc 2021;10(6):e019552.
[64] Camfield PR, Andrade D, Camfield CS, Carrizosa-Moog J, Appleton R, Baulac M, et al. How can transition to adult care be best orchestrated for adolescents with epilepsy? Epilepsy Behav 2019;93:138–47.
[65] Balestrini S, Guerrini R, Sisodiya SM. Rare and complex epilepsies from childhood to adulthood: requirements for separate management or scope for a lifespan holistic approach? Curr Neurol Neurosci Rep 2021;21(12):65.
[66] Fantaneanu TA, Jacobs CS, De Gusmao C, Bergin AM, Milligan TA, Pearl PL, et al. Pediatric and adult neurologist perspectives on the challenges of sustaining a transfer clinic. Neurol Clin Pract 2020;10(4):356–61.
[67] Hauser WA, Camfield CS, Camfield PR. Can an epilepsy transfer clinic be sustained in the United States? Neurol Clin Pract 2020;10(4):274–6.
[68] Campbell F, Biggs K, Aldiss SK, O'Neill PM, Clowes M, McDonagh J, et al. Transition of care for adolescents from paediatric services to adult health services. Cochrane Database Syst Rev 2016;4:CD009794.
[69] Christie D, Thompson R, Sawtell M, Allen E, Cairns J, Smith F, et al. Structured, intensive education maximising engagement, motivation and long-term change for children and young people with diabetes: a cluster randomised controlled trial with integral process and economic evaluation - the CASCADE study. Health Technol Assess 2014;18(20):1–202.
[70] Ridosh MM, Stiles-Shields C, Stern A, Winning AM, Anderson L, Sawin KJ, et al. Erratum to: The Adolescent and Young Adult Self-Management and Independence Scale (AMIS-II): Expanding evidence for validity and reliability. J Pediatr Rehabil Med 2022.
[71] Saarijarvi M, Wallin L, Moons P, Gyllensten H, Bratt EL. Transition program for adolescents with congenital heart disease in transition to adulthood: protocol for a mixed-method process evaluation study (the STEPSTONES project). BMJ Open 2019;9(8):e028229.
[72] Walter M, Kamphuis S, van Pelt P, de Vroed A, Hazes JMW. Successful implementation of a clinical transition pathway for adolescents with juvenile-onset rheumatic and musculoskeletal diseases. Pediatr Rheumatol Online J 2018;16(1):50.
[73] Singh SP, Paul M, Ford T, Kramer T, Weaver T, McLaren S, et al. Process, outcome and experience of transition from child to adult mental healthcare: multiperspective study. Br J Psychiatry 2010;197(4):305–12.
[74] Colver A, McConachie H, Le Couteur A, Dovey-Pearce G, Mann KD, McDonagh JE, et al. A longitudinal, observational study of the features of transitional healthcare associated with better outcomes for young people with long-term conditions. BMC Med 2018;16(1):111.
[75] Andreoli A, Klingbeil C. Implementing pediatric transition education initiative during inpatient admissions in the epilepsy monitoring unit. J Pediatr Nurs 2021;57:50–5.
[76] Disabato JA, Cook PF, Hutton L, Dinkel T, Levisohn PM. Transition from pediatric to adult specialty care for adolescents and young adults with refractory epilepsy: a quality improvement approach. J Pediatr Nurs 2015;30(5):e37–45.
[77] Smith PE, Myson V, Gibbon F. A teenager epilepsy clinic: observational study. Eur J Neurol 2002;9(4):373–6.