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Research article

Systematic review of study designs and methods in health transition research for young people with intellectual disabilities

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A B S T R A C T

Background: Transition for young people with intellectual disabilities from paediatric or adolescent services into adult health care services remains a difficult process for all stakeholders. The study assessed the type of interventions, the methodological approaches, study designs and location of existing published evidence in health care transitions.
Methods: A systematic review utilising the PRISMA protocol with an amended quality appraisal tool to explore the nature of published evidence on health care transitions for young people.
Results: Findings demonstrate that health transition research for this population lacks a robust evidence base and researchers favour exploratory studies investigating the experiential dimension of transition. The lack of involvement of young people in the studies indicates a problematic absence of genuinely participatory research.
Conclusion: The study is the first systematic review of empirical studies in health transition of young people with intellectual disabilities exploring the nature of existing evidence. The results will support setting priorities for future research.

What this paper adds

The study is the first systematic review of empirical studies in health transition of young people with intellectual disabilities which looked at the type of research evidence produced. It demonstrates that health transition research for this population lacks a robust evidence base and that researchers engage mainly in exploratory research about the experiences and perceptions of stakeholders, predominantly carers and staff. The absence of young people in the study design and implementation process but also the widespread absence of their voices in the studies themselves as participants is disappointing.

The review also demonstrated that there was only one study investigating the effects of a transition practice. The paper will provide important systematic evidence to inform future empirical research in the field of transition for young people with ID.

1. Introduction

Transition for young people with intellectual disabilities (ID) has benefited from significant policy attention over the last two decades [1, 2, 3, 4]. Yet, the time of transition remains fraught with problems for young people and their carers. Whilst there have been many studies on transition for this population, there is little evidence that transition outcomes for young people have improved [5, 6, 7].

Research has contributed to a better understanding of what young people with ID and their carers want from services during the transition process [8], what young people think is missing and how they think it can improve [9, 10, 11]. However, as some systematic reviews of existing evidence demonstrate [12, 13, 14], the field as a whole appears to be underresearched, as well as lacks strategies to design services that deliver smooth transition into destination services [15, 16, 17].

In this paper we will refer to young people with ID in the sense of the term as used in the United Kingdom. In the UK this term is synonymous with that of 'learning disabilities', whereas in the US the latter term

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denotes what is called a 'learning difficulty' such as dyslexia in the UK. We use the term 'transition' in terms of the move these young people make from children's service provider to health care services provided for adults. We focused on young people with ID to the exclusion of any other group of young people with other conditions because the health care needs of young people with ID are specific to them due to their cognitive and developmental delay [18, 19].

Young people with intellectual disabilities undergo several service transitions at the same time or consecutively, requiring coordinated responses from services working in different professional sectors. Apart from the biological changes leading to adulthood, transition to secondary education occurs first, at the age of 16, followed by transition into employment at 18 onwards, and transition from children to adult health care provision. Moving out of the parental home is often the last transition to take place, sometimes not happening at all, with young people continuing to live with their parents, following a period of time at a residential college [16, 20, 21].

The multiple nature of service transition has led to calls for a unified and coordinated service response with policy guidance urging schools, colleges, social and health care staff to work together to transition the young person to adult services. In the UK, a pillar of transition planning has been the statutory transition planning in schools, starting at age 14 [16,22]. There have been some attempts to combine educational transitions with planning for social and health care transitions, but there is evidence that, where transition plans include planning for other life domains than education, involvement of professionals from social and health care is rare and inter-service transition plans remain poorly coordinated or tokenistic [10, 23].

Given the lack of progress and the insufficient amount of evidence, the problem of transition may be a problem of research. The question arises whether studies conducted on transition produce strong, robust and generalisable evidence that can support changes in services.

The most prominent research frame in the field of transition may be captured by the question of 'what works for whom and when (or under what circumstances)' [24, 25, 26]. The question reflects a long term shift from research paradigms that have traditionally favoured linear cause and effect mechanisms underpinned by positivist ontological and epistemological principles to realist or interpretivist approaches [27, 28, 29]. Realist research seeks to understand the mechanisms that are being activated in certain contexts resulting in outcomes, and why [29, 30, 31, 32, 33, 34].

Health transitions represent a specific challenge for researchers. They involve complex interventions, containing several, potentially mutually confounding, active components influencing transition outcomes. Health transition interventions, such as joint clinics, multi-disciplinary transition teams, or key working, may also exhibit properties characteristic of complex phenomena, such as non-linearity, feedback loops and emergence [35, 36].

There is a need to obtain an overview of the types of study design and study methods that health transition research currently produces, and assess its relevance in relation to the challenges encountered by young people with ID and all stakeholders. The present study thus aimed to generate systematic knowledge that will help researchers to assess the current relationship between research methods, study design and service practice. It produces a picture on 'who' researchers focus on, which interventions they investigate, in which service sector their research takes place and which research designs they favour.

The study investigated the question: what is the current type of evidence produced by empirical studies in health care transitions for young people with intellectual disabilities? In the context of health transition, appraising the methodological type, thematic focus and participatory approach of published empirical studies is a key step in defining future research priorities. The aim of the systematic review was hence not to produce a narrative of findings underpinned by thematic analysis but to present an overview of the existing empirical evidence in health transition, the methods and study designs used, their thematic focus, service sector as well as participatory approach.

The study aimed to produce systematically obtained information about health transition where 'health' refers to the main primary and secondary health providers, such as family doctors (paediatrician or general practitioners) and acute and hospital services. In most health systems, core transition health planning either occurs at the primary or secondary health care sector, yet rarely through coordination in tertiary or allied health care systems such as physiotherapy or speech and language therapy.

2. Methods

The study started with the premise that the question 'what works for whom and when' represents the most prominent approach when examining the nature and scope of the evidence produced in health transition research. Different parts of the question were operationalized by establishing an analytical framework with 10 investigative domains: description of the transition intervention examined; type of participants; sampling practice; size of sample; study design; descriptive versus analytical design; information about the location of the study (adult or children's services); instruments used; service sector in which the research took place; and whether or not participatory research approaches were used to involve young people with ID.

Preferred Reporting Items for Systematic Reviews guidelines (PRISMA) were followed [37]. Electronic searches of the following databases were conducted by a trained information specialist. Medline, Embase, Cinahl, PsycInfo, Health Business Elite, HMIC, Social Care Online. Searches took place in November 2016 and were updated in November 2017. Search parameters were English language and having been published between 1990 and 2017. A search terms example is given below.

Example of Search Terms (Medline).

1. exp LEARNING DISORDERS/(19596)
2. exp INTELLECTUAL DISABILITY/(83643)
3. exp DEVELOPMENTAL DISABILITIES/(15887)
4. ("learning disorder*" OR "learning disability*" OR "intellectual disability*" OR "developmental disability*" OR "developmental delay*" OR "mental retardation").ti.ab/(48434)
5. 1 OR 2 OR 3 OR 4/(135272)
6. TRANSITION TO ADULT CARE/(452)
7. CONTINUITY OF PATIENT CARE/(15419)
8. (continu* ADV3 care).ti.ab/(17004)
9. (transition* ADJ3 (health* OR adult* OR care)).ti.ab/(8063)
10. 6 OR 7 OR 8 OR 9/(35734)
11. 5 AND 10/(351)
12. 11 [Limit to: Publication Year 1990–2016 and (Language English)]/(301)

2.1. Inclusion criteria

Papers included in the review were studies with an empirical content, where data were collected and analysed (this could include secondary data analyses), studies on health transition, and studies including participants who have an intellectual disability. Transition was defined as moving from children or adolescent to adult health care providers, this could occur in either the primary or secondary and tertiary care sector. Intellectual disabilities was taken to be synonymous with the term of learning disabilities where studies originated in the UK, and with the term developmental disabilities. Studies conducted in the US which used the term learning disabilities were not included since this term conventionally denotes what are called learning difficulties such as dyslexia in the UK.

2.2. Exclusion criteria

Studies were excluded if it was not clear whether participants had an intellectual disability, or where they had autism, epilepsy or physical disabilities without specifically noting that they also had an intellectual disability. The following types of documents were excluded: policy and guidance publications, conceptual papers, commentaries, discussion pieces or reflection papers, as well as systematic reviews, Cochrane reviews and meta-analyses of previously published studies. Studies about transition in tertiary sector providers were also excluded. We did so because aspect of transition from primary or secondary health care providers are different from tertiary services such as mental health or occupational therapy, physiotherapy and speech and language therapy. The main access point for health care services in the adult sector is a family doctor or general physician, whereas in the children's health care sector it would be a pediatrician. Whilst occupational therapists and physiotherapist play an important role in providing holistic care to people with ID, they are unlikely to lead on care transitions from children's to adult services. Where tertiary services were combined in a wider study with investigations of primary and secondary health care services, the papers qualified for inclusion. Studies related to all other kinds of transition were also excluded, such as transition in education, into employment or housing.

2.3. Data extraction

The aim of the review was to assess the type of evidence as judged against a specific set of indicators, papers were appraised by extracting information across 10 predefined items:

1. Transition practice or intervention
2. Participants
3. Sampling practice
4. Sample size
5. Study design - experimental/quasi-experimental vs/observational
6. Analytic versus descriptive design
7. Location of study (adult or children's health service)
8. Data collection instrument used
9. Sector - primary or secondary health services
10. Involvement of young people with ID in the study.

Studies were categorised as either: (1) a service needs analysis, (2) study of service perceptions and experiences of stakeholders, (3) an assessment of service change outcomes, or (4) audits. The investigative categories reflect different research paradigms, for example studies assessing the outcomes of service changes may preferably utilise empiricist research designs, whereas studies investigating the experiences of carers, young people or staff may favour interpretivist approaches. In turn, service audits may be popular to establish service needs to inform future service commissioning. However, the literature notes, no category is exclusively aligned with only one research paradigm [30].

Information about participants included the category of respondents such as carer, young people and/or staff. Sampling referred to information regarding the sampling method (purposive, convenience, random etc.). Studies were then analysed as to their study design. The main differentiation related to the difference between experimental or observational designs. The information was then appraised as to whether studies used an analytic (comparison) or a descriptive design.

The site of study was also noted. We were interested in whether the study was conducted across several service organisations or service sites (multiple site study vs. single site). Multiple site studies may enhance the generalizability of study findings. Sites could have variable patient population footprints, ranging from the largest (secondary analysis of national data sets) to the smallest (patients in a single clinic). Data collection instruments were also appraised. Instruments could be online, face to face, or phone surveys, focus groups and semi-structured interviews. Finally, we recorded the sector in which the study took place, i.e. primary or secondary care, or both.

2.4. Data analysis

Extracting information for the ten investigative domains inevitably involved some measure of judgement. Whilst our review included mostly objectifiable information, the question as to the level of involvement of young people in research required a definition of what constitutes meaningful involvement of young people with ID. We were guided by the participatory research paradigm formulated since the mid-1990s [38, 39, 40, 41, 42].

We took a pragmatic approach [43], using independent rating by two researchers, followed by open discussion where colleagues could challenge each other's interpretation and reasons for rating. At least two researchers worked on each full text of every paper to extract the information independently and then compared the results. Where there was disagreement, researchers discussed it and reached consensus. A third researcher cross checked the consolidated table of results and reasons given and queried issues where further justifications were needed.

3. Results

The search produced 1613 papers. After duplicates had been removed, 1339 papers remained. Title screening by three independent researchers (AK, JR, and JK) excluded an additional 712 papers, which left 627 eligible for inclusion in abstract screening. Abstract screening was also conducted by at least two independent researchers and 89 papers were thought to qualify for full text analysis. An updated search identified another 6 papers which were included in the full text analysis. Full text analysis, again by two independent researchers, reduced the field to 17 papers. Reasons were: policy paper (23); commentary (7); systematic review or meta-analysis (2); no young people with ID in study (34); results not reported separately for young people with ID (6). No authors were contacted for clarifications or additional information Fig. 1).

3.1. Descriptive results

Seventeen (n = 17) papers were included in the final analysis. They represented a range of study methods, data collection instruments and drew on various stakeholder groups for participation. Categories were not mutually exclusive so studies investigating various transition aspects could gain multiple entries. Studies are referred to in brackets relating to their number in the overview table (Table 1).

3.1.1. Research topic and investigative focus

The analysis showed that six studies investigated the service needs of young people with intellectual disabilities (1, 7, 9, 10, 13, 16). Particularly prominent were scoping studies assessing population health care needs through secondary analysis of transition surveys in the US.

Ten studies examined the experiences and perceptions of stakeholders in the transition process (1, 2, 4, 5, 6, 8, 9, 10, 11, 14). It is noteworthy that whilst parents/carers as well as staff perceptions were studied, young people' perceptions of the health transition process were rarely obtained (1, 4, 7, 12).

Service changes were investigated in one study (3), whilst a clinical intervention was studied in one paper (16). Transition outcomes were explored in one further study, where authors looked at the transition destination of young people with intellectual disabilities as recorded by a large national transition survey (17). One study reported the results of tool development where a transition preparation tool was used to facilitate smooth health care transitions (12).
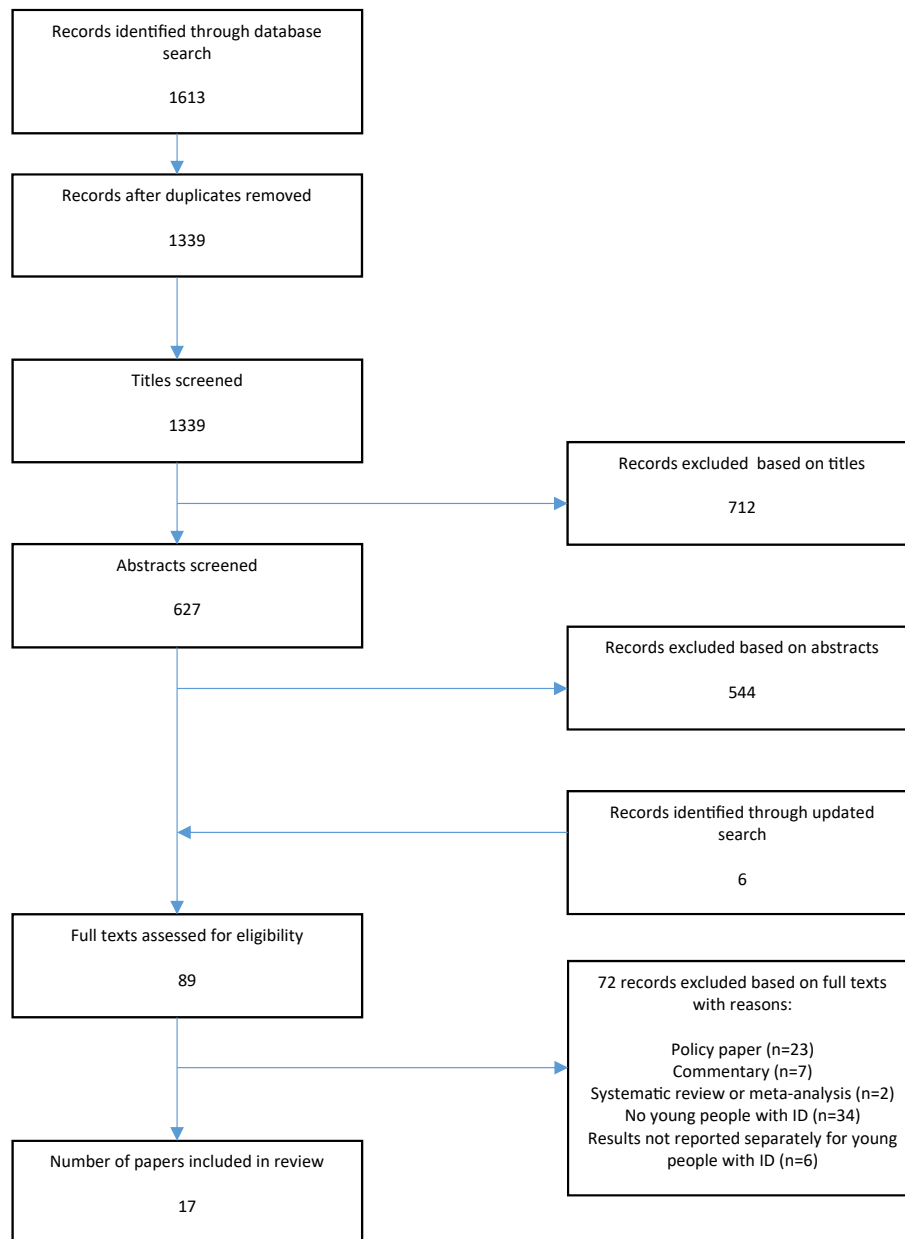
Fig. 1. PRISMA flow diagram of study selection

3.1.2. Study participants

Fifteen studies recruited participants on the basis of health care records of young people. Eight of those investigated the content of young people's health care data in terms of service utilisation or service needs and outcomes. Yet, only one of the 17 studies actually interviewed young people (4). Eight studies interviewed, conducted focus groups or surveyed carers (1, 2, 4, 6, 8, 9, 10, 11) and six studies interviewed or surveyed professionals (3, 4, 5, 6, 8, 14).

3.1.3. Sampling

Most studies in the review utilised convenience sampling (n = 4) or used a self-selection process (n = 4) where carers or staff were approached and asked to participate. Five studies conducted secondary analyses of existing health care data or national survey data (9, 10, 13, 15, 17). One study used a purposive sample. In general, information about the sampling strategy were difficult to obtain as it was not always clear how participants were selected, approached or recruited.

3.1.4. Sample size

Four studies used a national register of young people with health care needs or clinical register of patients to identify and approach participants (9, 10, 15, 17). Where respondents were recruited through services, the sample sizes varied from 140 (maximum) to 2 (minimum). Information about sample size is only useful in conjunction with research approach so we cross-checked sample size with qualitative research design and found that four studies using qualitative research instruments such as interviews and focus groups had a sample size ranging from 24 (1) to 2 (6). If the study with a slightly unconventional data collection method specified as a 'discussion' was excluded (11), the sample sizes for the remaining studies appeared small, yet reasonable, with 24, 16 and 17 participants respectively.

3.1.5. Experimental versus observational design

All studies used an observational design which may demonstrate the difficulties of conducting experimental or quasi-experimental studies in the field of intellectual disabilities, a challenge for all research in people

Table 1
Data extracted from included papers.

		Practice	Participants	Sampling	Sample size	Experimental vs. observational design	Analytic vs. descriptive	Data collection instrument	Site	Sector
1	Bhaumik et al., 2011 [67]	scoping service need and perception	YP, Carers	self-selecting carers	140 YP from scoping, 24 carers	observational	descriptive	postal questionnaire, interviews	local services, city and county UK	community paediatric, CAHMS, social services
2	Bindels-de Heus, van Staa, van Vliet, Ewals, & Hilberink, 2013 [68]	experiences	carers	convenience	131 carers	observational	descriptive	online questionnaire	region	hospital and community
3	Caan, Lutchmiah, Thomson, & Toocaram, 2005 [70]	health facilitator role	staff	service contacts/ convenience?	150	observational	descriptive	focus group schedule, interviews, reflective diaries,	2 council areas (London)	community?
4	Pickler, Kellar-Guenther, & Goldson, 2010 [80]	barriers	all stakeholders	convenience	16 YP, 15 professionals, 17 carers =48	observational	descriptive	focus group	state (Colorado)	community (paeds)
5	Durkin, Zurakowski, Rangel, Lillehei, & Fishman, 2015 [73]	staff experiences	staff	self-selecting	238	observational	descriptive	survey online	US	paediatric surgeons
6	Olsen & Swigonski, 2004 [79]	parent staff interaction	carers and staff	convenience	3	observational	descriptive	discussions	pilot site US (not specified)	paediatrician
7	Betz, Redcay, & Tan, 2003 [66]	needs analysis YP	non and disabled youth	audit	25, 7 disabled	observational	analytic, cross-sectional	chart (scale?)	local transition clinic	community based, nurse led
8	Geenen, Powers, & Sells, 2003 [74]	perceptions/ attitudes parents of level of involvement of health care staff in transition	parents and staff	public school register, paediatric surgeon register Oregon	753 parents, 141 staff	observational	analytic, cross-sectional (parents vs. staff attitudes)	mailed survey	state (Oregon)	paeds, GPs and other providers
9	Lotstein, McPherson, Strickland, & Newacheck, 2005 [76]	perceptions of future needs planning	parents/ guardians	national register	5533	observational	descriptive	telephone survey	national US	not specified
10	Scal & Ireland, 2005 [81]	health care needs perceived by parents	parents	national register	4332	observational	descriptive	telephone survey	national	not specified
11	Davies & Beamish, 2009 [72]	experiences	parents	purposive	17	observational	descriptive	interviews	local US	hospital, paediatric clinic
12	M. McManus et al., 2015 [78]	tool development	young people	self-selecting	35	observational	descriptive	tool and survey	local US	3 paediatric primary care clinics
13	Borlot et al., 2014 [69]	pathway transition and different needs analysis depending on transition origin (community vs. clinic)	young people	patient data	302 (170/ 132)	observational	analytic, cross-sectional	clinical data audit	local US	paeds clinics and tertiary centres
14	Camfield, Gibson, & Douglass, 2011 [71]	perceptions barriers to transition	clinical staff	convenience (conference attendees)	133	observational	descriptive	survey	US conference	paeds, nurses
15	M. A. McManus	transition preparation received?	YP	national register	17144	observational	descriptive	survey (phone)	national	childrens

Table 1 (*continued*)

		Practice	Participants	Sampling	Sample size	Experimental vs. observational design	Analytic vs. descriptive	Data collection instrument	Site	Sector
	et al., 2013 [77]									
16	Berens & Peacock, 2015 [65]	clinic intervention evaluation/ needs	young people	audit data clinic	332	observational	descriptive	clinical data audit	Houston, Texas	childrens hospital
17	Lin, Lee, & Adirim, 2015 [75]	transition outcomes compared dis vs non dis group	YP	national register	1438 no dis/ 413 disab) = 1851	observational	analytic, cross-sectional	survey	national	health general, no info

services where ethical review often rules out randomization of services to participants.

3.1.6. Analytical versus descriptive design

The studies in the review were predominantly of descriptive character. Four studies utilised a comparative design (7, 8, 13, 17). One study investigated the effect of a local transition clinic, comparing the differential impact of the clinic between disabled and non-disabled young people (7). Another study examined different complexities of health care needs between cohorts from different origins (13). A third study compared the experiences of transition by parents of young people with the perceptions of staff involved in the same process (8).

3.1.7. Instruments

Studies included in our review used a range of instruments. Studies bridged the analogue with the digital era, so the medium of survey data collection differed. Three surveys were administered over the phone and two where postal questionnaires, whilst five surveys were conducted online. Three studies used interviews (1, 3, 11) and two studies conducted focus groups (3, 4). One study also asked participants to keep a reflective diary (3) and one study provided insufficient information as to which instrument was used for data collection (6).

3.1.8. Site characteristics

The study distinguished between single and multiple site as well as geographical size of the site. For the latter, we used the categories 'local', 'state or region', 'national', and 'other'. The analysis did not yield sufficient information about whether or not any studies recruited multiple sites to participate. Whilst we noted that some papers made reference to various providers included in the study, information was insufficient to draw any conclusions about the nature of the multiple sites. Since drawing on data from multiple sites provides stronger evidence and improves generalizability of study finding, this lack of information was frustrating. However, since a second reason to include a second site in a study is often to use a comparative design, and few of the studies used such a design, the lack of information about multiple design features of studies may be reflective of the wider opportunistic methodological approach in much of intellectual disabilities research.

3.1.9. Health care sector

A lack of clear information was apparent in most studies about the sector under investigation. One often criticized aspect of transition research is that most studies focus on children's and adolescent services and underemphasize the importance of the adult sector in facilitating good transition outcomes. This leads to the impression that transitions, even where well organized and coordinated amongst children's services may lead into 'nowhere' [5, 18, 44].

No study in our review included adult sector providers. All studies were based in, or recruited participants from adolescent or paediatric services and focused on transition preparations or services facilitating

transition from the children's site. One study (17) did investigate transition outcomes but did not inquire further into the role of adult services in shaping and informing transition destinations.

In terms of location of provision, there has been some debate in the literature about the nature, quality and costs of community versus hospital based health care provision following de-institutionalisation [45, 46, 47] and we scrutinized the information in the selected papers about this. Four papers did not provide sufficient information as to whether community or hospital based services were focus of the study (3, 9, 10, 17). Six papers investigated transition services in community services (1, 2, 4, 7, 8, 12) and three papers focused on hospital based services (2, 11, 16). We recognize that community services vary significantly across regions and states, but still thought this was important information that highlighted a trend in health care research with young people with intellectual disabilities. The predominance of community based service studies may reflect easier recruitment strategies for participants or the dominant service model with community providers leading on transition arrangements.

3.1.10. Involvement of young people in research

None of the papers noted any involvement of young people in either the design, implementation or analysis of data in the research. This is not surprising given that meaningful involvement may require participatory research approaches that are related to different research paradigms [48, 49, 50]. However, it was disappointing to see that there has been so little progress in effectively involving young people, moving research from research on to research with the population [41, 51, 52, 53].

The table above shows that, in our sample of selected studies, the most likely study to be undertaken was an investigation of health transition involving either young people, carers or staff as participants, most likely investigating their experiences or perceptions of transition, where the research was bound to be conducted in the children's sector, favouring descriptive over comparative designs, using mainly surveys and lacking any meaningful involvement of young people with ID in the research process.

4. Discussion

The study set out to examine the type of evidence produced by empirical studies on health care transition for young people with intellectual disabilities since 1990. Seventeen studies that qualified for inclusion were analysed in line with an amended quality appraisal instrument. The findings show that there is relatively little empirical research in the field on this topic, even though there are now some systematic reviews emerging [12, 13, 54, 55]. Our findings demonstrate that there is limited reliable evidence available to service planners or commissioners of health care on what works for whom and under which circumstances.

Overall, the papers in the review revealed that there is a healthy methodological pluralism in the field, without qualitative or quantitative

approaches being unduly privileged. When looking at the topics and domains investigated, few studies aim to scope the needs of the population, particularly by talking to young people themselves. Experiences and perceptions of parents and carers appear to be a predominant concern of health researchers. It should be cause for concern that the views of young people was the focus of only one study. Given that there is some evidence that the way in which young people perceive transition differs from the opinions of their parents, and that professionals in turn view the process differently to parents [56], this lack of investigative focus on young people's perceptions of transition is worrying. Knowing what parents or staff want can only provide a partial picture of what a smooth health care transition should look like.

Only one study examined the effects of service changes, and whilst it is encouraging to see such a study, the relative absence of intervention research speaks to the fact that health care transition research in intellectual disabilities is still in the exploratory phase. There is however no reason why specific transition practices, such as joint clinics cannot be explored through studies utilising qualitative research approaches. Doing this type of research is essential if we want to gather knowledge about what works for whom and in which service context. Whilst we acknowledge that it is difficult to design and implement (quasi-)experimental studies, there may be some potential in constructing controlled, multi-site studies which would provide good reliable evidence about transition practices and their impact on young people.

The lack of tool development and their evaluation is also disappointing. Transition in education and employment has been marked by a plethora of transition tools [12, 57], some more useful than others, and it is not clear why there is such an absence of tools to facilitate health care transitions. One reason may be that the statutory process of transition in education provides a more conducive environment to tool development as educational policy mandates the use of some tools in the school's planning process. With paediatric services usually fragmented between acute settings and community provision tools may appear too dependent on singular provider organisations and hence of limited use. There is some anecdotal evidence that some hospitals have developed transition support tools but it is not clear how widespread this is [58, 59].

A significant advantage of the transition survey in the US is that it provides a data point for all school leavers ready for secondary analysis [60, 61]. This vital resource has engendered several transition studies which were included in our review. Whilst there are some data in the public domain in other countries, such as the UK, there are no equivalent studies, even though Emerson et al. have helpfully set out how to generate similar datasets for health services from local commissioning groups in England [62]. Researchers in countries other than the US should explore the possibility to exploit similar national databases to facilitate transition outcome analyses for this population.

Our analysis of the sampling strategy revealed that too few studies provide clear information about the way in which their study participants have been selected, approached and recruited. We are therefore precluded to make any conclusive remarks about the sampling strategies employed but note that, where researchers work hand in glove with health care services, good sample sizes can be achieved. Working with local providers may be a promising route to recruit young people and their carers for studies on health care transition. Whilst we recognise the challenges of collaborating with GPs, hospital providers and community services may offer an alternative way to recruit participants. When publishing empirical papers on transition, researchers should be clear about the sampling strategy employed in their studies allow reviewers the estimation of sampling biases.

The analysis of the studies in our review clearly showed a preference for observational, descriptive non-comparative designs. This is easily explained by the difficulty to gain ethical approval for studies with randomised allocation to different services [63]. Restricting access or granting privileged access to services requires justification and is rarely countenanced by ethical review panels [64]. Another challenge may be the nature of evaluative work where researchers are often asked to assess the impact of a service change long after changes have been implemented. A lack of baseline data makes it difficult to ascribe potential effects or impact to a singular intervention or practice change within a complex health care provision. This is especially the case for young people with intellectual disabilities who are often recipients of several health care services due to co-morbidities. As mentioned above, the use of multi-site, controlled studies, or difference in difference designs may be a useful mitigating strategy in this context.

In line with the preferred exploratory design, most of our studies favoured the use of instruments requisite to the qualitative research approach. The majority of studies used focus groups and interviews to gather data from participants. This is indicative of the largely exploratory nature of the studies in our review and, as mentioned above, the absence of any assessment of actual transition practices is of concern. Making datasets and data collection instruments publicly available would aid the standardisation of data collection instruments and may contribute to the strengthening of the evidence base.

The papers also rarely specified clearly in which health care sector the study took place. Whilst all studies focused on children's and adolescent services, it was not clear whether they conceptualised sufficiently the significant fragmentation in children's health care between community, paediatric and hospital services. Since GPs are the providers of choice in the adult sector, the transitions in community and acute settings are likely to be radically different for young people with intellectual disabilities who often transfer to various different consultants in, sometimes, different hospitals after transition. We believe that health transition research should reflect this fundamental division between community and hospital transition. A first step would be to ensure that clear reporting standards are being used.

Looking at the summarizing table (Table 2) there appears to be a poor match between the main research methods, the research objectives, research foci (adult versus children's and adolescent health services), and data collection instruments with the primary service target population, young people with ID. There is ample evidence that people with low adaptive functioning, have difficulty using some instruments such as online surveys and using surveys with the population with ID yields suboptimal results. Involving young people in the design, analysis and reporting of studies is therefore likely to improve the quality and usefulness of the research. It stands to reason that participatory, comparative studies using mainly qualitative research methodologies would enhance the ability to form the 'interpretive community of professional knowledge and lived experience' (Daviter 2019, p.74) that is necessary to produce useful and reliable evidence in the transition field.

Table 2

'Who', 'What', 'Where' and 'How' research was undertaken.

	Investigative Focus		Number of studies
Who	Young people with ID		8
	Carers		8
	Staff		6
What	Experiences/perceptions		10
	Needs		6
	Outcomes		3
	Other[1]		1
Where	Sector	Adult	0
		Children	15
		Unclear	2
How	Comparative design?	Descriptive	13
		Anadlytical	4
	Data collection instrument	Survey	10
		Interview	3
		Focus group	3
		Audit	2
		Other[2]	2
	Participatory design?	Yes	0
		No	17

[1] Tool development.

[2] Informal discussion and unspecified instrument.

The absence of transition research relating to the destination services, the adult sector is, also a considerable disappointment. It appears that researchers who often lament the 'transition into nothing' provide little insight themselves into what this destination looks like, and how to improve it for young people with ID.

5. Limitations

The study has some limitations. We did not utilise a quality appraisal tool, but instead used a set of indicators for the extraction of information from the selected studies. Since the study aimed to explore the nature, scope and study design of papers, a focus on the ten extraction indicators instead appears justified. Quality appraisal tools assess the quality and strength of evidence which this study did not examine.

6. Conclusion

The study is the first systematic review of empirical studies in health transition of young people with intellectual disabilities which looked at the type of research produced. It demonstrates that health transition research for this population lacks a robust evidence base and that researchers engage mainly in exploratory research about the experiences and perceptions of stakeholders, predominantly carers and staff. The absence of young people in the study design and implementation process but also the widespread absence of their voices in the studies themselves as participants is disappointing.

In addition, it is surprising that there was only one study investigating the effects of a transition practice. Intellectual disability research needs to do better in this area if it wants to provide reliable answers to the question of what works for whom in health transition.

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