



Review Article

Transition From Pediatric to Adult Healthcare for Colorectal Conditions: A Systematic Review



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ABSTRACT

Background: Despite surgical advances for complex congenital colorectal conditions, such as anorectal malformation (ARM) and Hirschsprung disease (HD), many adolescents require transfer from specialist pediatric to adult providers for ongoing care.

Methodology: A systematic review of PubMed, MEDLINE and Embase was conducted to identify what is known about the transitional care of patients with ARM and HD (PROSPERO # CRD42022281558). The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) framework guided our reporting of studies that focused on the transition care of 10–30-year-olds with ARM and HD.

Results: Eight studies were identified that included patient and parent (n = 188), and/or clinician perspectives (n = 334). Patients and clinicians agreed that transitional care should commence early in adolescence to support transfer to adult care when a suitable level of maturation is reached. There was little evidence from patients that transfer happened in a timely or coordinated manner. Patients felt that clinicians did not always understand the significance of transfer to adult services. No models of transition care were identified. Surgeons ranked ARM and HD as the most common conditions to experience delayed transfer to adult care. Beyond pediatric surgeons, patients also highlighted the importance of general practitioners, transitional care coordinators and peer support groups for successful transition.

Conclusions: There is little research focused on transitional care for patients with ARM and HD. Given evidence of delayed transfer and poor experiences, the development of models of transitional care appears essential.

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1. Introduction

Children with complex congenital colorectal conditions, such as anorectal malformation (ARM) and Hirschsprung disease (HD), require surgical management early in life. Despite advances in surgical care, many patients experience sequelae which persist into adolescence and will benefit from transfer of specialist care from pediatric to adult providers. Usual pediatric surgical care offers limited support to engage with adult providers, with concerns of loss of follow-up and poor health outcomes [1–6]. Adolescents and

young adults often only re-engage with health services when their condition deteriorates to a point where there is no option but to seek emergency care from adult health services [5,7–9].

Decades of research into many childhood conditions, such as Type 1 diabetes and cystic fibrosis, have led to increased awareness of the healthcare and wider needs of these cohorts through adolescence [10]. The risks of dropping out of specialist medical care are also well understood, which has fuelled efforts to develop structured and supportive approaches to preparing patients and families to transfer to adult providers [8,11]. In contrast to medical disciplines, the topic of transitional care is a more recent focus for many surgical specialities.

The aim of this systematic review was to identify what is known about transitional care for adolescents with colorectal conditions, including the barriers and enablers to successful transition, with the objective of informing future programs and policies.

Abbreviations: ARM, anorectal malformation; HD, Hirschsprung Disease; MDT, Multidisciplinary team.

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2. Methods

In this review, we differentiate the terms ‘transfer’ and ‘transition’. Using established nomenclature, we have defined ‘transfer of care’ as a point in time when healthcare formally shifts from the responsibility of the paediatric provider to the adult provider [12]. In contrast, we recognise ‘transition of care’ as a process that involves a gradual shift of responsibility from carer to adolescent, that is facilitated by supporting the young person to become empowered to manage their own care [7,12]. Transfer is therefore a component of transition, which is a process that continues beyond the period of engagement with paediatric services [13].

This study was conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines and registered with PROSPERO (CRD420 22281558) [14]. A search of MEDLINE, Embase and PubMed was conducted on 1st February 2022 using the search strategy detailed in Appendix 1.

2.1. Study selection

The search criteria were restricted to peer review articles published in English from January 2011 to February 2022 to ensure that the review captured contemporary practices. Editorials and letters were excluded. Articles were imported into Covidence [15] and two authors (EJM and MYT) independently screened titles, abstracts, and relevant full texts against the inclusion criteria, which were: studies of people with colorectal conditions, with a median age between 10 and 30 years, that focused on transition or transfer of care from pediatric to adult services or providers. Beyond patients, studies of carers and/or clinicians of patients who met these criteria were also eligible. When consensus could not be reached a third author (MT) was consulted.

2.2. Data extraction

Due to the diversity of articles that included patient, parent/carers and clinician experiences, the type of data extracted varied significantly. For papers that largely focused on adolescents, data extracted included: age at transfer, access to transitional care, age that any focus on transitional care commenced, and barriers and

enablers of transfer. For papers that largely focused on clinicians, data extracted included: age of patients, prompts for transfer of care, reasons for not transferring care, responsibility for transitional care, and barriers and enablers of transfer.

2.3. Data presentation

Barriers and enablers of successful transfer were categorised using the framework on quality health systems from The Lancet Global Health Commission for High Quality Health Systems (Fig. 1.) [16]. Individual barriers and enablers of transfer were grouped using the foundational level of the framework (population, governance, platforms, workforce, tools). Two authors (EJM and SMS) independently categorised the barriers and enablers into these five domains (Table 1).

3. Results

The initial search identified 758 titles, from which 383 abstracts were screened, and 38 full text papers were reviewed. Supplementary information was obtained from Cairo et al. and Nataraja et al. during the full text analysis stage [9,17]. We were unable to contact Witvliet et al. [18]. This resulted in eight papers which met the search criteria (Fig. 2 and Table 2). All eight studies were conducted in high income settings.

3.1. Patient perspectives

There were 188 patient and carer participants across five studies [9,19–22]. Four studies were of patients, while another did not differentiate findings between patients and carers [9]. There was a wide age range of patients, including 3 largely adult studies (median age ranged from 24 to 29 years). Two studies included patients with ARM, two studies included patients with HD and one included patients with ARM and HD. Twenty-eight males and 42 females participated across those four studies. The 118 participants in the study by Cairo et al. were not differentiated by gender [9].

3.1.1. Patient expectations and experiences

Patients believed that their paediatric surgeon was responsible for transfer of care and felt this should occur in line with

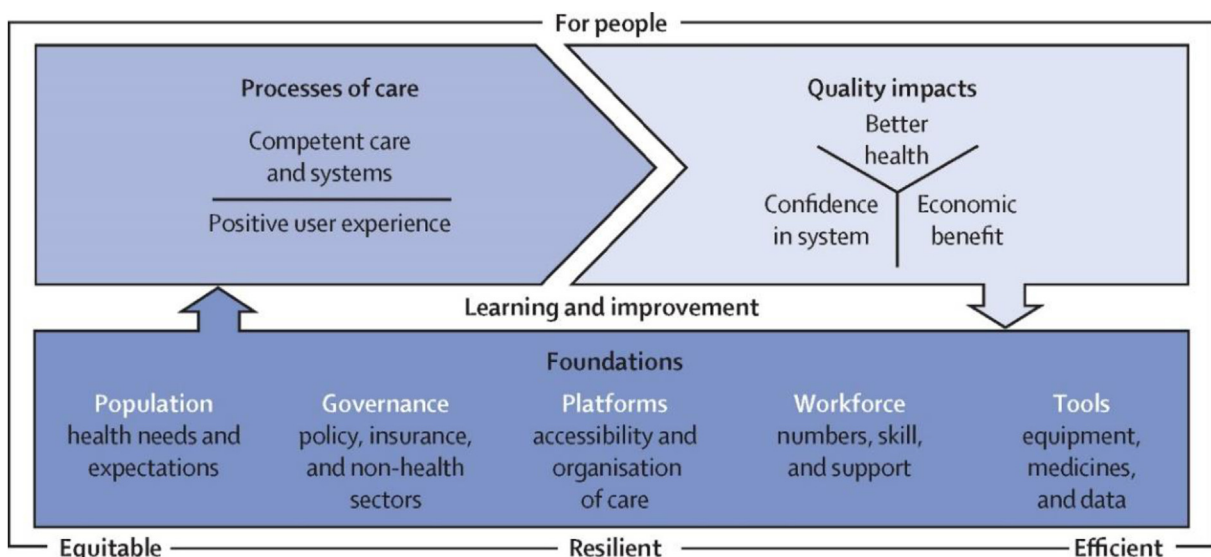


Fig. 1. High Quality Health Systems Framework [16]. Reproduced with permission under STM permissions guidelines.

Table 1
Reported enablers and barriers of transition, classified in accordance with The Lancet Global Health Commission for High Quality Health Systems Framework (16).

	Hoel 2021	Tofft 2020	Cairo 2018	Nah 2018	van der Bent 2018	Giuliani 2016	Durkin 2015	Connor 2017
<ul style="list-style-type: none"> This may enable successful transfer of care X When this does not occur it is potential barrier to transfer of care								
Adolescent is able to care for their condition independently			• X	X	•	• X		
Adolescent understands disease progression and long term needs					• X			
Adolescent understands the benefits of transfer to an adult service								
Adolescent understands their condition	• X	• X				X		
Adolescent/carer have confidence in adult providers			X		X			
Adolescent/carer's positive relationship with paediatric provider					X		X	
Age appropriate relationship between parent/carer and adolescent				•		• X		
Effective strategies for managing sensitivity of diagnosis			X		X			
Family/social supports are available				•	•		•	
Adolescent has access to private health insurance					• X			
A written transition plan is used to guide transitional care		•	•			X		
Adequate communication between paediatric and adult teams		• X	•	•	X	• X	•	
Adolescent is given a list of referral options prior to transfer	•		•		X	X		
Adolescent is able to contact relevant clinicians in the adult setting	X	X			X			
Adolescent is being seen alone for at least part of their appointment	•			•		•		
Adolescent is provided a copy of their history prior to transfer			•					
Appropriate support groups exist	•	•						
Availability of specialised adult care centres	• X	•			X			
Care coordination is available in the adult setting		•			X			
Care coordination is provided during transition period		•	X	•				
Early commencement of transition planning	•	•	•		X	• X		
Education in navigating the health care system	•	•	•			•		
Mental health support is available during adolescence and transition	•		• X		• X			
Multidisciplinary team is involved during transition period	•		•					•
Patient is familiar with adult environment				X				
Reside close to adult care			• X				X	
Transitional care is tailored to the adolescent		•				•		
Adolescent has a role in any medical decisions that pertain to their care		•	•	•	• X	•		
Adult clinician is familiar with ARM/HD	• X	X	X		• X	•	X	
Adult clinician is willing to assume care of adolescent/adult patient		X					•	
Adult clinician is willing to seek guidance from other clinicians when unsure					X			
Adult clinician's understands the challenges of chronic illness in adolescence	•		X		X			
Adult provider is willing to treat adolescent based on disease complexity			X		X			
Dedicated transitional care clinician		• X						
General/ Family Practitioner is involved in the transition process	X				•			
Sufficient availability of qualified adult colorectal surgeon	X	X	X				• X	
Accurate information is readily available in a variety of media		• X						
Standardised assessment tool is used to determine transition risk						X		•
Standardised assessment tool is used to measure ARM/HD complications						X		
Standardised care of ARM/HD in adolescence						X		
Population								Adolescent participants
Governance								Clinician participants
Platforms								Tool creation study

All elements of care in this table are presented in the affirmative which align to the enablers of successful transfer, as indicated by a dot in the table. A cross in the table indicates that there is evidence within the article that when this element of care is not present during transition, it may act as a barrier to successful transfer. The one exception to this, is positive relationship with paediatric provider, as a positive relationship with paediatric provider may act as a barrier to successful transfer.

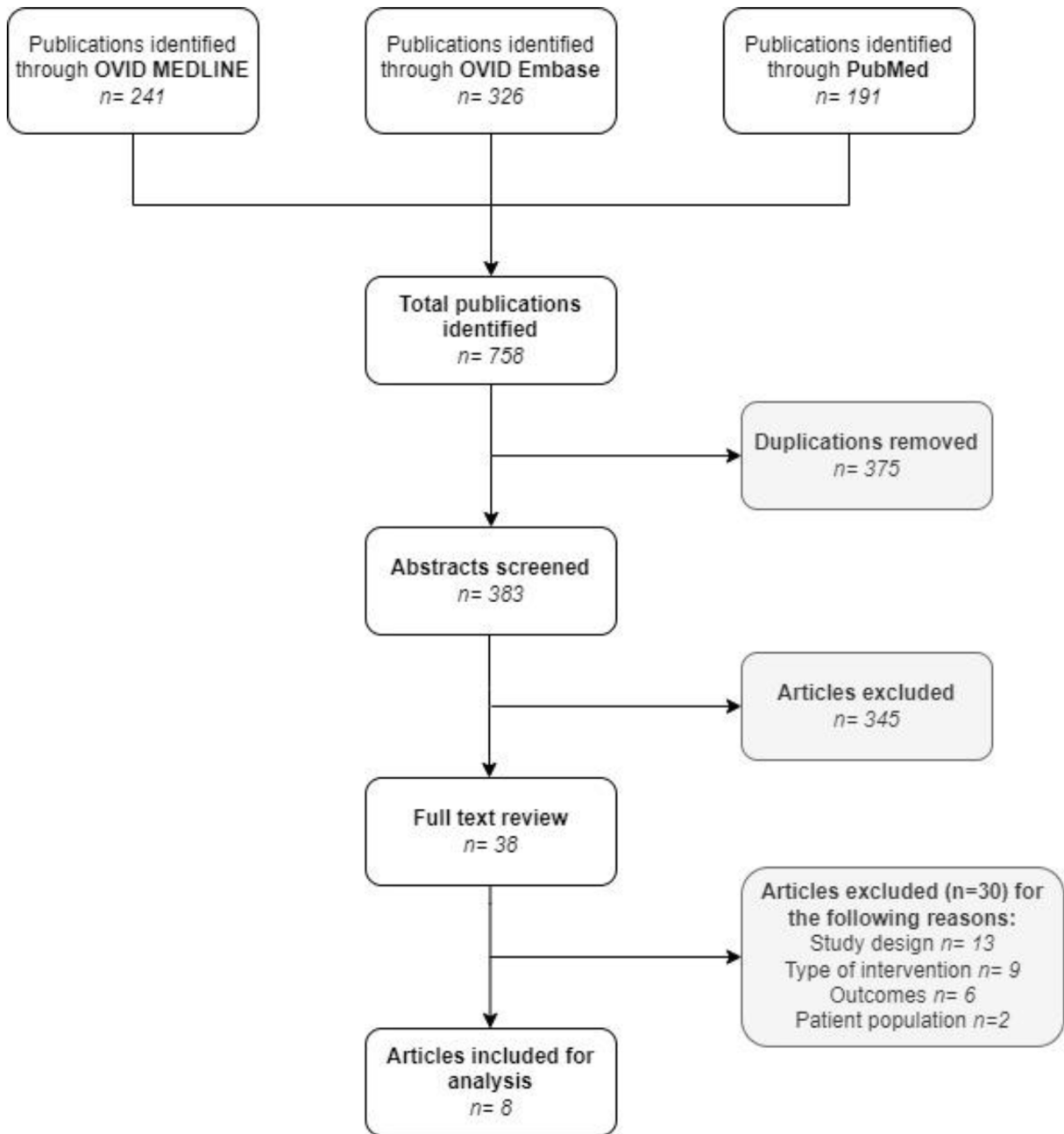


Fig. 2. Study selection following the PRISMA guidelines.

adolescent maturation [20,21]. Additionally, patients believed that transitional care should commence in early adolescence [19]. Patients reported the onset of new medical and psychosocial complications during adolescence, including bullying, and sexual and reproductive health concerns [19,20]. Some patients described their teens as “the worst period to be living with a dysfunctional bowel disorder” [20].

3.1.2. Current model of care

A varied proportion of patients (3–33%) received care from a multidisciplinary team (MDT) [9,21]. Only one person (7%) reported that they were cared for by an MDT in the adult setting [21]. Discussions about transfer to adult care providers appeared infrequent; only 13% of patients surveyed by Cairo et al. reported having had any preparatory discussions, and 67% of respondents in the

study by van der Bent et al. reported no discussion regarding transition or transfer [9,21].

There was no consistency in the timing of transfer to adult health services, even within the same service. For example, one study that included patients who had transferred to adult care reported that five patients had transferred between 16 and 18 years of age and six between 19 and 21 years of age. A further 10 patients at that site remained under the care of the paediatric team, despite the study participants' median age of 28.5 years [21].

3.1.3. Transfer readiness

Symptoms and function were found to be better predictors of cognitive and emotional adjustment than the diagnosis of ARM or HD [22]. Researchers reported “lightbulb” moments when adolescents understood the “chronicity and continuing impact” of their

Table 2
Characteristics of included studies.

First author (Year)	Study Aims	Data Collection	Population description	Number of participants (n) & response rate (%)	Age	Condition
Cairo, S. (2018)	To examine both patient and parent experiences with transitional care for those who have undergone a childhood operation for ARM or HD	Web-based survey, with some open-ended response options	Patients/carers affiliated with the Pull Through network or REACH. No age limitations and no differentiation between patient and carer respondents. No details on patient location; however, the majority of PTN and REACH members are located in USA and Canada.	118 (estimated response rate 26.2%)	Mean (SD) 12.3 (11.6) years	57 (61.3%) ARM, 9 (9.7%) Cloaca, 27 (29%) HD
Durkin, E. T. (2015)	To determine whether individual surgeon's demographics (e.g. gender, years of experience, practice setting) influence their perspectives on transition.	Survey of members of the American Pediatric Surgical Association	Pediatric surgeons <5 yrs post fellowship (n = 38), 6–10 yrs (n = 34), 11–15 yrs (n = 42), 16–20 yrs (n = 35), >20 yrs (n = 34)	238 (22%)		ARM and HD.
Giuliani, S. (2016)	To evaluate current international practice and identify future research priorities around long-term follow-up, management of active problems during adolescence, and transition of care to adult services for children with ARM.	Survey of delegates attending the UK- Pediatric Colorectal Group Meeting and the 22nd International Meeting of the Pediatric Colorectal Club.	76 colorectal surgeons, 12 trainees, 11 others working in colorectal care (e.g., allied health professionals).	96 (41%)		ARM
Hoel, A. T. (2021)	To acquire in-depth knowledge about how HD influences somatic and psychosocial health from childhood to adulthood.	Focus group discussions (patients grouped by gender)	Patients with HD attending adult Colorectal clinics in Norway and Sweden.	17 (33%)	Median (range) 29 (19–43) years	17 HD (9 male, 8 female)
Tofft, L. (2020)	To explore the needs and expectations of adolescents and adults born with ARM around transitional and adult healthcare.	Focus group discussions (patients grouped by gender)	Patients with ARM who had been treated for ARM at one of two pediatric tertiary centres (Norway and Sweden).	16 out of 82 eligible patients (20%), 8/46 in Sweden (17%), 8/36 in Norway (22%).	Median (range) 24 (19–47) years	16 ARM (6 male, 10 female)
van der Bent, A. (2018)	To explore the transition experiences of a cohort of adult patients with ARM and identify factors that influence the ability to achieve satisfactory transition.	Survey.	Patients with ARM who had received treatment at Boston Children's Hospital between 1993 and 2017, and were now older than 18 years. Participation also invited through support groups.	26 (19 from Boston Children's Hospital and 7 from support groups).	Median (range) 28.5 (23–29) years	26 ARM (7 male, 19 female)
Nah, S. A. (2018)	To explore adolescents' and young adult's perceptions of their illness, healthcare, bowel symptoms, and the lifestyle adjustments that they make.	One-on-one interviews using a question guide developed from a literature review and informed by clinician expertise.	Patients who had received primary corrective surgery at KK Women's and Children's Hospital (Singapore) and continued to be regularly followed.	11 (9% of eligible patients).	Mean (range) 16.9 (14–21) years	7 ARM (3 male, 4 female) 4 HD (3 male, 1 female)
Connor, M. J. (2017)	To create an evidence-based transition risk assessment score that can identify adolescents most at risk during transition period.	Pilot study of new tool administered in clinic.	Patients of anorectal clinic at St. George's University Hospital, UK.	7 completed the tool on one occasion, 4 completed on two occasions, 3 completed on three occasions	Median age at each wave of 13, 15 and 16 years, respectively.	14 ARM

HD - Hirschsprung Disease, ARM - Anorectal malformation.

condition, which typically did not occur until the “late preteen or early teen years” [22]. In the work of van der Bent et al., 42% of patients over 18 years of age remained under the care of their paediatric surgeon; 22% of surveyed patients were not ready for transfer, and 13% explicitly stated that they would never be ready to transfer to adult care providers [21]. The main reasons patients reported not wishing to transfer to an adult surgeon were the established relationship (70%) and comfort (70%) with their paediatric surgeon [21].

3.2. Clinician perspectives

From the two studies that recruited clinicians, Durkin et al. surveyed paediatric surgeons who were members of the American Pediatric Surgical Association and Giuliani et al. surveyed colorectal clinicians attending two conferences in the UK (the UK-Paediatric Colorectal Group, and the 22nd International Meeting of the Pediatric Colorectal Club) [23,24].

3.2.1. Clinician expectations and experiences

Many surgeons were noted to have “never considered their role in the process of transition, seeing it as a “strictly medical” issue” [23]. In contrast, Giuliani et al. found that 86% of surveyed clinicians felt it was the role of the paediatric surgeon to coordinate transfer of care [24]. In that study, 89% of clinicians reported a significant proportion of patients with ARM developed new problems in adolescence that required extending the MDT prior to transfer of care. The majority (90%) of clinicians did not use any objective scoring system to risk stratify adolescent patients [24]. However, when asked, 81% reported that they felt a practical tool may be useful in determining which adolescents would require the most transitional support. Addressing this gap, the Transition Risk Assessment Score (TRAS) was created, which combines three existing scoring systems; the Krickenbeck (colorectal), Pena (nephrourological) and Bai (Quality of Life) assessment tools into a single measure to classify patients as low, medium or high risk of requiring ongoing medical care during adolescence [25]. A pilot study found that the majority of patients were rated in the medium risk category, with suggested benefit from continuing engagement in health care, management of active problems, and attention to various psychosocial morbidities specific to this age group [25].

3.2.2. Current standards of care

In the UK, while 73% of clinicians reported providing MDT care from birth, the majority (82%) did not hold MDT meetings with adult practitioners during the transition process [24]. In the USA, Durkin et al. found that ARM and HD were overrepresented in patients with delayed transfer to adult care [23]. Hirschsprung disease was the most prevalent condition that continued to be treated by paediatric surgeons (25% of surgeons treated patients over 25 years), with ARM equal second (22% of surgeons). Conversely, Giuliani et al. reported that 33% of clinicians routinely discharged patients with ARM before the age of 10 years [24].

3.2.3. Transfer readiness

Between 28% and 52% of surgeons reported institutional policies governing the commencement of transitional care [23,24]. While many surgeons reported that this process should occur early, starting between the ages of 9 and 12 years, Giuliani et al. also found that 27% suggested that transitional care should only commence between 17 and 18 years [24]. Transfer and transition were not defined by Giuliani et al., so it is unclear if clinician responses referred to the timing of preparatory transitional care or the timing of transfer to adult services [24].

Adult medical comorbidities, pregnancy, substance abuse and poor adherence with treatment commonly prompted transfer to adult care [23]. Clinicians also reported that they would transfer a patient if initiated by another doctor of a mutually managed patient. In contrast, patients were not transferred to adult care for a variety of reasons, including developmental delay, enteral access, difficult venous access, ongoing surgical needs and patient attachment.

3.3. Barriers and enablers of high-quality transitional healthcare

3.3.1. Population

The population subgroup comprises factors that relate to the health needs and expectations of the patient population [16] (Fig. 1.). Two areas emerged as enablers of successful transfer: patient understanding and skills, and the role of family. Multiple studies reported that an adolescent's ability to care for themselves, and their understanding of the condition supported successful transfer to adult care [9,19–21,24].

The role of family was shown to influence the likelihood of transfer. Appropriate adjustment of the carer/adolescent relationship, as well as more generalised family and social supports, were considered to support transfer [21–24]. Conversely, carers who retained too much control within the adolescent/carer relationship were considered a barrier to successful transfer [24].

A number of factors were only discussed in the context of barriers to transfer. This included an adolescent or carer who lacked confidence in adult providers, had insufficient strategies for managing and coping with the sensitivity of a colorectal disorder, or had a positive relationship with paediatric providers [9,21,23]. None of the studies explored the importance of adolescents understanding the potential benefits of transferring to adult providers.

3.3.2. Governance

Governance seeks to understand the role of policy, insurance and non-health sectors in the provision of high-quality healthcare [16]. In the case of colorectal care, the only evidence was in relation to health insurance. In Boston, USA, van der Bent et al. found that access to health insurance enabled successful transfer of care; not having health insurance served as a barrier [21].

3.3.3. Platforms

Platforms focus on the accessibility and organisation of care [16]. Early commencement of transitional care was found to be key to increasing the likelihood of successful transfer, as was ensuring transition was individualised and that adolescents were regularly seen alone for at least a portion of their appointments [9,19,20,22,24]. Conversely, failing to initiate transitional care early and not ensuring that adolescents are familiar with the hospital to which they will be transferred was thought to jeopardise successful transfer [21,22,24].

Care coordination during the transition period was thought to promote a smoother transfer [9,20–22]. Similarly, mental health support was thought to enable transfer, while its absence led to increased difficulty [9,19,21]. Multiple studies also found that MDT involvement during transition was thought to improve transfer outcomes [9,19,25].

Providing written resources, such as transition plans and lists of referral options, was considered to increase the chance of successful transfer [9,20,24]. Education in navigating the healthcare system, access to their complete medical history, and availability of support groups were all considered likely to support transfer to adult care [9,19,20,24].

While the structure of service provision varied significantly depending on location, both access to specialised adult colorectal centres and the patient residing near these centres were found to

increase the likelihood of successful transfer [9,19,20]. Conversely, a patient not being able to access a specialised adult colorectal centre due to geographical distance or lack of availability of a service, could hamper transfer [9,19–21,23].

Finally, the most significant determinant of success of transfer was linked to the quality of communication between paediatric and adult clinicians. Six of the eight studies showed evidence that when communication occurred well, transfer was more likely to be successful than when there was insufficient communication between care providers [9,20–24]. Failure of paediatric and adult clinicians to communicate and work collaboratively prior, during and following transfer was considered to reduce the chance of successful transition [20,21,24]. Similarly, the inability of an adolescent or adult patient to make direct contact with the adult clinician was also found to be a likely barrier to successful transfer [19–21].

3.3.4. Workforce

Workforce relates to the number of employees, their skill level and the supports available to them [16]. The most commonly identified workforce enabler was clinicians who allowed the adolescent to have a role in all medical decisions [9,20–22,24]. Other enablers of transfer included access to adult clinicians who were familiar with ARM or HD, who understood the challenges of living with a chronic condition during adolescence and who were willing to assume care for adolescents whom, in many cases, had complex surgical histories [19,21,23,24]. van der Bent et al. found that adult clinicians who were unwilling to seek guidance from other clinicians when unsure of the best approach to treatment harmed the transition process [21]. Ensuring linkages with family and/or general practitioners may also support a successful transfer to adult care [19–21].

3.3.5. Tools

Tools relates to the equipment, medicines and data used within the healthcare system [16]. The absence of standardised ARM or HD care, and the absence of standardised tools to determine those at greater risk of difficult transition, may be barriers to successful transfer [24]. The provision of accurate information through a variety of media was found to potentially enable transfer [20].

4. Discussion

Many themes arise from this small but complex body of research about transitional care for patients with ARM and HD. Our review suggests that there are many commonalities between the transitional care needs of the colorectal cohort and other adolescent patients whose needs are embodied in various general transition guidelines [26–28]. In contrast to the expectation within these guidelines that transitional care is routine, our findings provide little confidence about this for adolescents with complex colorectal conditions. The scarcity of transitional care research in high income countries with significant healthcare resources suggests that those living in low and middle income countries will have an even lower chance of supported progression from paediatric to adult care.

These data indicate an unusual combination of both early discharge to primary care (for example, in late childhood) and delayed transfer to adult specialist services (for example, the mid-twenties). This variability in the timing of transfer may suggest that there is a group of patients with ARM/HD who are discharged in childhood without expectation that future specialist care will be required. Little is known about the natural history of this cohort and whether they indeed go on to enjoy good health in adult life. In the absence of such data, discharge to primary care in early adolescence rather than to adult specialist surgical services may be appropriate. This contrasts with a significant group of patients with

ARM/HD who remain in paediatric care beyond the age that would be considered timely to transfer [29]. While the marked variability in timing of discharge or transfer may reflect different complications and comorbidities for adolescents with ARM/HD, it is also likely to reflect the absence of institutional policies.

In the absence of clearly defined policies, paediatric surgeons must make individual judgements about the best timing of patient transfer. Our data show that in some cases, this resulted in them retaining young adults within the paediatric system, while in other cases, especially when 'adolescent issues' arose, such as substance abuse and pregnancy, clinicians promptly transferred care to adult providers. Beyond the need for policy around transition, including the time of transfer, this suggests that enhanced adolescent health training for both paediatric and adult providers, and the MDTs they work with, could maximise the likelihood of successfully managing comorbidities in either system.

While this review has highlighted that transitional care for colorectal patients is infrequently provided, it suggests that many of the enablers and barriers of transition for this cohort align with those of well documented guidelines that largely reflect medical cohorts [26–28,31]. Transitional research emerging from large surgical cohorts, such as congenital cardiac and transplant patient groups, infers substantial alignment with the transitional care needs of these medical cohorts [30,32–34]. There is an absence of transitional guidelines for patients with less prevalent surgical conditions, such as the colorectal conditions of interest within this review [30,32–34]. The European-based ARM-net consortium released a consensus statement about transitional care for patients with ARM that was informed by a thorough literature review of this largely medical literature [4]. While the ARM-net expert panel felt there was good agreement between the recommendations and the needs of the colorectal surgical cohort, this was based on expert opinion rather than empirical research.

Many of the key enablers identified in both medical and surgical cohorts were evident in the studies within our review. As found by Suris et al., communication between paediatric and adult care providers and between providers and patients were both identified as key to successful transition, including the provision of a complete health summary at the time of transfer [27]. Aspects such as the opportunity for the adolescent to be seen alone for at least part of the appointment, the presence of peer support, and the use of technology such as telehealth and electronic medical records (EMR) are also consistent with recommendations within contemporary transitional care guidelines [27,30,33].

Current guidelines encourage empowering adolescent patients through supporting their self-management skills and engagement in decision making [26,27]. While clinicians, patients and families all agreed that these were enablers of successful transfer, there was limited evidence to suggest that this was occurring in practice. Similarly, few patients reported discussions about transfer or the opportunity to meet adult clinicians before transfer, another standard recommendation. Much research on transitional care highlights the role of MDT care in meeting the holistic health needs of the adolescent, such as the range of psychosocial complications that were described during adolescence [5,9,19,25]. Disappointingly, there was no evidence of routine psychosocial assessment of health risks, including mental health, despite evidence that adolescents with chronic health conditions are at least as likely, if not more likely, to engage in risk taking behavior than their peers [10,35]. Enhanced MDT support throughout the transition period would provide a valuable opportunity to support and educate families and patients with the goal of enhancing wellbeing and supporting timely transfer.

While not unique to the adolescent colorectal cohort, some of the challenges identified by both patients and health professionals

are commonly seen in at least some surgical cohorts [36]. The relative rarity of congenital colorectal conditions means that few surgeons in adult settings are able to develop experience or expertise in them. This can make finding an adult provider challenging, which often requires significant travel (and cost). Some adult clinicians are unwilling to assume care for young adult patients with complex surgical histories whose earlier procedures are largely unique to the paediatric setting [9,21]. This perpetuates the fear in patients and carers, documented in at least some studies, that adult colorectal surgeons are not equipped to care for congenital colorectal conditions [9,21]. One arguably unique feature of this cohort is the sensitivity of the diagnosis itself or of anorectal surgery; a number of adolescents and young adults talked of not wanting to share their diagnosis even with health professionals [20], an area deserving further research.

It is apparent that transitional care for colorectal patients is in its infancy and that work is required at a systems level to meet the recommendations of professional bodies, such as the Society for Adolescent Health and Medicine [28]. Improved training of both paediatric and adult clinicians would help ensure that all are better equipped to manage the raft of unique challenges presented by adolescents with chronic health conditions. Hospitals must also ensure that staffing and budgets are allocated to promote appropriate continuity of care. Improved knowledge of the progression of ARM and HD over time, and the prevalence of comorbidities would promote greater standardisation of care across adolescence. Despite strong evidence that transition is most beneficial when coordinated by a nominated member of the MDT, much of this responsibility currently falls to surgeons, with no evidence of contingency should surgeons not have capacity to support such complex periods of change [27].

Paediatric surgeons face a difficult balancing act in their relationship with adolescent patients. They need to maintain a positive connection with patients, while preparing them for adult care. This includes instilling confidence in adult healthcare providers despite them being unlikely to have the same expertise. Interestingly, none of the studies included in this review analysed any potential benefits of transfer. While adult care is fundamentally different to paediatric care, it is likely that there are only benefits for young people to transfer their care to a facility where the breadth of health needs may be more appropriately managed. This does not preclude the value of ongoing input from paediatric surgeons in adult settings if this were deemed appropriate. Assisting adolescents (and their families) to understand these benefits, which will be fostered by paediatric providers having positive relationships with adult providers, may lessen the anxiety and reluctance that some adolescents and their families experience surrounding transfer.

Most of these studies used convenience samples of adolescents and young adults who were recruited through hospital clinics. We do not know how representative these findings are of birth cohorts of patients with ARM/HD. Several studies were also limited by small sample size and low response rates. It is likely that adolescents who remain engaged with services in adolescence and adulthood have more complex colorectal presentations, which means that these findings may not be reflective of the population of patients with ARM/HD. Finally, while we had anticipated a critical appraisal of the literature, the nature of the outcomes did not lend themselves to a meta-analysis so we chose not to proceed with this.

5. Conclusion

Continuity of healthcare is fundamental to the optimal care of adolescents and young adults with ARM and HD which will be supported by clinician education, policy development and system change. Further research with larger and more representative birth

cohorts is also required, as creating a fully informed model of care requires better understanding of the natural history of ARM and HD and their comorbidities across the life course.

Previous communication

Nil.

Data availability statement

Data sharing not applicable to this article as no datasets were generated or analyzed during the current study.

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Conflict of interest

None of the authors have any conflicts of interest to disclose.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.jpedsurg.2024.02.012>.

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