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Transitional Care in Anorectal Malformation and Hirschsprung's Disease: A Systematic Review of Challenges and Solutions

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ABSTRACT

Background: The literature on transitional care in anorectal malformation (ARM) and Hirschsprung's disease (HD) is diverse and heterogeneous. There is a lack of standards and guidelines specific to transitional care in these conditions. We aim to establish and systematically categorize challenges and solutions related to colorectal transition care.

Methods: Systematic review of qualitative studies from MEDLINE, EMBASE, PubMed and Scopus databases (2008–2022) was conducted to identify the challenges and solutions of healthcare transition specific to ARM and HD. Thematic analyses are reported with reference to patient, healthcare provider and healthcare system.

Results: Sixteen studies from 234 unique articles were included. Fourteen themes related to challenges and solutions, each, are identified. Most challenges identified are patient related. The key challenges pertain to: (1) patient's lack of understanding of their disorder, resulting in over-reliance on the pediatric surgical team and reluctance towards transitioning to adult services; (2) a lack of education and awareness among adult colorectal surgeons in caring for pediatric colorectal conditions and inadequate communication between pediatric and adult teams; and (3) a lack of structured transition program and joint-clinic to meet the needs of the transitioning patients. The key solutions are: (1) fostering young adult patient's autonomy and independence; (2) conducting joint pediatric-adult transition clinics; and (3) ensuring a structured and coordinated transition program is available using a standardized guideline.

Conclusion: A comprehensive framework related to barriers and solutions for pediatric colorectal transition is established to help benchmark care quality of transitional care services.

Level of Evidence: IV.

Type of Study: Systematic review without meta-analysis.

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1. Introduction

Transition to the adult healthcare for children and young adults with anorectal malformation (ARM) and Hirschsprung's disease (HD) is a growing concern [1,2]. In part, the complexity of the process for congenital colorectal disorders bears similarity with many other chronic medical conditions of childhood [3–5]. Additionally, ARM and HD have unique needs related to the potential problems of colorectal [6–8], urological [9–11], and gynecological [11] dysfunctions. Significant anomalies such as VACTERL associations may coexist [12,13]. Variation in disease phenotype, compounded by disparate surgical techniques, render the outcomes following surgical repair unpredictable. Living with these functional and anatomical issues is a major cause for psychosocial disorders [14] and problems related to sexuality [15,16] which can impact on their quality of life in adulthood.

Despite the growing attention given to transition of healthcare over the past decade leading to the development of national guidelines [1,17–19] and transition programs [20,21], several reports continue to raise concerns about the quality of transitional care, even from centers that have allocated resources to this area [22,23]. Whilst this may be multifactorial, it has highlighted the importance of setting the standards related to transitional care.

Generic guidelines related to transitional care have offered the fundamental principles required for implementation of healthcare policies. However, condition-specific transition guidelines play a

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Abbreviations: ARM, anorectal malformation; HD, Hirschsprung's disease.

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unique and important role in surgical conditions [24–26]. In inflammatory bowel disease, for instance, a systematic review has identified pertinent issues which set targets for intervention, providing valuable auditable outcome measures [27]. Such guidelines for ARM and HD are lacking despite the abundance of literature reporting the barriers in transitional care. Although much insight could be gleaned from a diverse array of qualitative research, the breadth of methodologies and distinct differences in focus between studies have contributed to the challenge of elucidating key elements for effective transition. Therefore, there is a need to understand the barriers in transitioning children with ARM and HD systematically to effect actionable solutions that conform to optimal standards.

This systematic review aims to identify and categorize the challenges and solutions related to colorectal transitional care in patients with ARM and HD. The findings can provide a framework for standards setting for all transition programs.

2. Methods

2.1. Protocol

This systematic review was performed according to Enhancing transparency in reporting the synthesis of qualitative research (ENTREQ) guidelines [28]. The protocol for this review was registered on International Prospective Register of Systematic Reviews (PROSPERO) (CRD42022348575) [29]. This review aimed to answer two research questions:

1. What are the challenges related to transitional care in ARM and HD?
2. What solutions or models have been used in ARM and HD to overcome these challenges?

2.2. Search strategy and identification of relevant research

This review was performed across four databases (MEDLINE, EMBASE, PubMed and Scopus) to identify articles related to the subject, including publications from 1st January 2008 to 31st August 2022 (15 years). Search was limited to English language using search terms and key words constructed from a modified PICO approach: *Population* (ARM and Hirschsprung patients), the phenomenon of *Interest* (follow-up studies, challenges, solutions), *Context* (transition to adult care, continuity of adult care) and *Outcomes* (challenges and solutions). Further identification of studies was performed through interrogation of the reference lists of selected articles.

2.3. Inclusion and exclusion criteria

Inclusion criteria for each study include peer-reviewed publications containing original qualitative data that described healthcare transition from pediatric to adult care for ARM (including cloaca) and HD. Studies presenting qualitative data with quantitative descriptors (*e.g.* Likert scale) were included to supplement the interpretation of qualitative data. Systematic or narrative reviews, conference papers, case reports, seminar abstracts or expert opinions were excluded.

The screening of abstracts and titles was performed by two independent reviewers (JP, SS). Full texts were assessed for eligibility criteria by the same two independent reviewers. Any disagreements over the eligibility of a particular study were resolved through discussion between all reviewing authors to make the final decision for data inclusion or exclusion. Data extraction from the eligible articles were completed by all authors independently and concluded through agreement.

2.4. Qualitative data extraction and analysis

The extracted data were organized to record specific characteristics of the included studies, including publication details, country, study design, number of participants and key outcomes. The text from each article was extracted, and common themes across articles were identified and compared for similarities and differences.

Assessment of the quality of studies was examined utilizing the Critical Appraisal Skills Programme (CASP) (scored out of a maximum of ten points) [30]. Each study was evaluated by scoring the number of ‘Yes’ responses in the CASP appraisal, with ranges of 1–3, 4–6, 7–8 and 9–10 categorized as poor, moderate, high, and very high quality, respectively.

Qualitative data extraction and analysis followed Braun and Clark's six phases of reflexive thematic analysis [31]. Data extraction for both research questions (regarding challenges and solutions) involved coding key words and phrases from the Results section of the main text in the included studies. These extracted data were then organized into groups based on shared overarching themes and tabulated in order of importance, considering the number of studies and participants in all the studies.

These themes were categorized into patient, healthcare provider and healthcare system related issues. The term ‘*patient*’ refers to the individual patient undergoing the transition process, as well as their family or caregivers. ‘*Healthcare provider*’ refers to the clinical team led by the pediatric colorectal surgeon handing over the care and the adult colorectal surgical team receiving the care of the patient. ‘*Healthcare system*’ encompasses the local or national governing policies, guidelines, and the environments in which the patient had been cared for, that was beyond the remit of the individual provider.

3. Results

3.1. Literature search and study inclusion

As shown in the PRISMA Flow Diagram (Fig. 1), the initial search strategy returned 302 reports from all four databases. Forty-seven duplicate reports were excluded. Among the unique 234 articles identified, the first stage screening using title and abstract excluded 209 articles consisted of systematic or narrative reviews, expert opinions, and case reports. The remaining 25 articles were subject to full-text review, whereby nine did not fulfil the inclusion criteria and were excluded (Appendix).

There were 16 studies included for the review to address the two research questions based on data derived from 794 participants (Table 1). The included studies were conducted at multi-national ($n = 3$) and national ($n = 13$) levels spanned across seven countries. The majority were from the USA ($n = 5$). Methodologies include survey or questionnaire ($n = 9$), interview ($n = 4$), pilot study ($n = 1$), cross-sectional study ($n = 1$) and mixed methodology ($n = 1$).

3.2. Critical appraisal

Levels of evidence were III ($n = 1$) and IV ($n = 15$) (Table 1). According to the CASP criteria, the quality of the included studies ranged from moderate ($n = 1$) to high ($n = 9$) and very high ($n = 6$) (Table 2).

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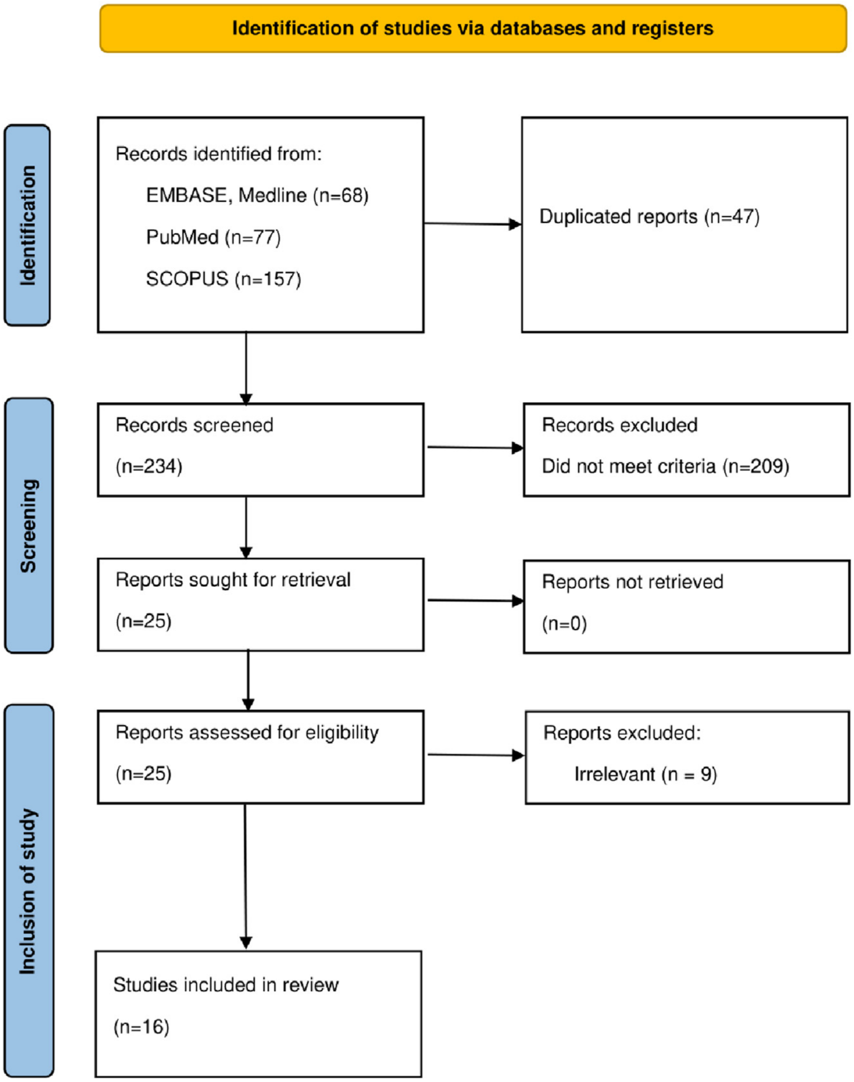

Fig. 1. PRISMA flow diagram of systematic review.

3.3. *Challenges related to colorectal transitional care (Table 3)*

Most challenges identified were found to be related to patient factors. The most common challenge, highlighted in seven studies (n = 231) [32–38], was the lack of patients' awareness of their own condition. This was followed by the reluctance of patients or parents to transition away from their current pediatric surgeon, as shown in six studies (n = 565) [33,36,38–41]. In some countries, the lack of insurance cover also posed a barrier for patient engagement [35,39,42]. The patients' lack of self-advocacy and initiative [33,39], developmental maturity [36] and support from caregivers [36] ultimately resulted in these children being left out once they entered adulthood.

Among the challenges related to healthcare provider, the most common barrier was the lack of education and awareness of adult surgeons in caring for pediatric colorectal conditions, which had been highlighted in 11 studies (n = 678) [32–42]. There was a perceived inadequate communication between pediatric and adult surgeons even when transition had been attempted, as shown in eight studies (n = 305) [32,35–38,41–43]. One study further indicated a lack of interest from adult surgeon leading to ineffective transitional care [44].

In barriers related to the healthcare system, it had been highlighted that a lack of structured transition program [six studies (n = 463) [32,33,36,38–40] led to consequential poorly organized adult clinic for young adult patients with ARM and HD [six studies (n = 343) [33–36,41]]. The studies also highlighted the lack of resource-allocation [43,44] and provision of a multidisciplinary approach [35,38] in developing the effective transition programs.

3.4. *Solutions related to colorectal transitional care (Table 4)*

The solutions which are essential for colorectal transitional care can be broadly divided into the following three categories:

For patients, the most important emphasis was on developing autonomy and independence as highlighted in three studies (n = 163) [33,37,44]. Two studies had also shown education and training specific to patients' condition were effective interventions (n = 122) [33,43]. Young adult patients should also be encouraged to join the relevant colorectal support groups for practical supports [43].

On the other hand, for healthcare providers to achieve optimal transitional care, a joint pediatric-adult colorectal clinic was deemed essential in six studies (n = 221) [33,38,44–47]. As mentioned, promoting autonomy and independence should be

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Table 1
Characteristics of articles included for systematic review.

Study	Country	Study type (Level of Evidence)	Population	Outcome
Gribbin et al. (2013) [43]	International	Questionnaire (IV)	Non-profit organisations, parent support groups (n = 26)	Evaluation of the structure and effectiveness of parent support groups in pediatric anorectal anomalies.
Durkin et al. (2015) [39]	USA	Survey (IV)	APSA members (n = 238)	Description of the pediatric surgeon preferences and their current transition practice patterns.
Giuliani et al. (2016) [33]	International	Survey (IV)	Delegates attending UKPCG and IMPCC (n = 96)	Evaluation of long-term FU. Active issues present in teenagers. Transition of care in ARM.
Connor et al. (2017) [46]	UK	Pilot study (III)	Teenagers with ARM (n = 14)	Creation of risk assessment score for stratification of health care needs and interventions in ARM patients transitioning to adult healthcare system.
Witvliet et al. (2017) [47]	Netherlands	Survey (IV)	ARM or HD patients (n = 17)	Assessment of transitional care.
Cairo et al. (2018) [41]	USA	Cross-sectional (IV)	ARM and HD patients and families affiliated with PTN or REACH (n = 118)	Examination of patient and parent's experiences with transitional care.
Nah et al. *(2018)* [37]	Singapore	Interview (IV)	Children with ARM or HD (n = 11)	Exploring physical, social, and emotional impacts of ARM and HD.
Tan et al. *(2018)* [40]	Malaysia	Survey (IV)	Paediatric surgeons in Malaysia (n = 48)	Assessment of participants' demographic characteristics, current practices, and their perspectives regarding TOC.
van der Bent et al. (2018) [35]	USA	Survey (IV)	Patients ≥18 years old with ARM (n = 26)	Exploration of the experiences of patients with ARM.
Tofft et al. (2020) [32]	Norway, Sweden	FGI (IV)	Adolescents and adults born with ARM (n = 16)	Exploration of the needs and expectations of transitional care and of adult healthcare among adolescents.
Hoel et al. (2021) [34]	Norway, Sweden	FGI (IV)	Patients treated for HD in childhood (n = 17)	Acquiring knowledge of HD influence on somatic and psychosocial health. Exploration of transitional care in HD patients.
Loftus et al. (2021) [42]	USA	Survey (IV)	Adults with congenital bowel and bladder conditions (n = 30)	Satisfaction with bladder and bowel management. Interest in reproductive health. Successful transition into adult care. Perceived barriers of transitioning into adult care.
Ahmad et al. (2022) [36]	USA	Survey (IV)	Members of ASCRS, MWPSC, PCPLC (n = 43)	Identification of the most common barriers by pediatric and adult surgeons in the transfer of care of ARM patients.
Kassa et al. (2022) [38]	Sweden	Interview (IV)	Adolescents, young adults and adults with VACTERL association (n = 22)	Exploration of the understanding of transition to adult healthcare services.
Mellerio et al. (2022) [45]	France	Mixed method (qualitative analysis and interview) (IV)	Adolescents with chronic conditions (n = 27)	Assessment of the value of transition preparation consultations as a new service of transitional care.
Violani et al. *(2023)* [44]	Europe, UK	Survey (IV)	Members of ARM-Net Consortium, ERN eUROGEN, ERN ERNICA (n = 45)	Evaluation of transitional care organization.

American Pediatric Surgical Association (APSA), American Society of Colon and Rectal Surgeons (ASCRS), Anorectal malformation (ARM), Hirschsprung's disease (HD), Children's Hospital Colorado (CHCO), Focus Group Interview (FGI), Healthcare provider (HCP), Midwest Pediatric Surgery Research Consortium (MWPSC), Pediatric Colorectal and Pelvic Learning Consortium (PCPLC), Transition of care (TOC), United Kingdom Paediatric Colorectal Group (UKPCG).

strongly encouraged in the transition clinic [33,38,44]. Moreover, adult colorectal surgeons should receive support through training and education to enhance their knowledge in pediatric colorectal conditions and understanding of the long-term needs of these patients [44,47].

In this review, we found that most of the solutions reported the need for optimizing the current healthcare system. First and foremost, there was a strong call for a structured and standardized guideline for healthcare transition (three studies, n = 163 subjects) [33,38,44], complemented by written information about the process (three studies, n = 94) [38,44,45] and the use of risk-assessment score to stratify healthcare needs [33,46]. Adopting a multidisciplinary approach with the support of a transitional care coordinator [38,44] and a counsellor or psychologist [33,44,47], would be the most effective way to manage their needs comprehensively. In some settings, resource-rationalization may determine the need for centralization of the transitional services [38,47]. Outside the hospital system, funding colorectal support groups and ensuring patients could be closely affiliated with them, helped ensure supports are available in the long term within the community [43,44].

4. Discussion

This systematic review has analyzed the existing literature on pediatric colorectal surgery by assimilating the challenges that impact on the feasibility and quality of transitional care. To further address the variability of the literature, it has grouped these challenges into three categories based on the stakeholders involved i.e., patients, healthcare providers and healthcare system. As a result, modifiable factors were elucidated as targets for implementing evidence-based and relevant solutions. This enabled us to generate a framework which can be actioned upon by clinical teams and healthcare commissioners, using the summaries outlined in

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Table 2
Evaluation of quality of studies using CASP appraisal.

Study	Critical Appraisal Skills Programme (CASP) criteria										Methodological quality
	1	2	3	4	5	6	7	8	9	10	
Mellerio *et al.* [45]	Y	Y	Y	Y	Y	U	Y	Y	Y	Y	Very high
Loftus *et al.* [42]	Y	Y	Y	Y	Y	N	N	Y	Y	Y	High
Tofft *et al.* [32]	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	Very high
Giuliani *et al.* [33]	Y	Y	Y	Y	Y	N	N	Y	Y	Y	High
Gribbin *et al.* [43]	Y	Y	Y	Y	Y	N	N	Y	Y	Y	High
Durkin *et al.* [39]	Y	Y	Y	Y	Y	N	N	Y	Y	Y	High
Hoel *et al.* [34]	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	Very high
van der Bent *et al.* [35]	Y	Y	Y	Y	Y	N	N	Y	Y	Y	High
Ahmad *et al.* [36]	Y	Y	Y	Y	Y	N	N	Y	Y	Y	High
Tan *et al.* [40]	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	Very high
Connor *et al.* [46]	Y	Y	Y	Y	Y	N	N	Y	Y	Y	High
Witvliet *et al.* [47]	Y	Y	N	Y	N	N	Y	Y	N	U	Moderate
Cairo *et al.* [41]	Y	Y	Y	Y	Y	N	N	Y	Y	Y	High
Nah *et al.* [37]	Y	Y	Y	Y	Y	N	N	Y	Y	Y	High
Kassa *et al.* [38]	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	Very high
Violani *et al.* [44]	Y	Y	Y	Y	Y	N	Y	Y	Y	Y	Very high

https://casp-uk.net/casp-tools-checklists/.
Y = Yes, N = No, U = Unclear.
1-3 = poor, 4–6 = moderate, 7–8 = high, 9–10 = very high quality.

Tables 3 and 4. They consist of 14 themes related to challenges and solutions respectively, forming the core elements in standards setting for future care quality improvement and audit in pediatric colorectal transition.

The importance of a structured transitional program is evidenced by the positive impact it has on patients, families and the healthcare system [48]. Within the realm of pediatric colorectal patients, consisting mostly of ARM and HD, where bowel, urological and sexual dysfunctions are known to extend beyond childhood, effective transitional care is especially important [15,16]. Currently, the majority of healthcare settings globally necessitate children with long term medical conditions to move from pediatric to adult healthcare system when they reach adulthood [40]. The latter system is predominated by a different conglomerate of pathologies with disparate care models. This is a challenge because the divide between pediatric and adult healthcare systems defaults transitional care to be disjointed unless concerted effort is given to consolidate the process.

Several guidelines do exist to incorporate recommendations for transitional care [1,17–21] The American Academy of Pediatrics and National Institute of Clinical Excellence (NICE) in the UK have recommended early transition commencing at the age of 12–13 years of age for allowing more time for preparation and navigating through the adult healthcare system, since as early as 2011 [17,18]. These guidelines offer common principles which deserve careful consideration but are not specifically tailored towards colorectal care. Furthermore, there is a lack of evidence of definitive benefits from these guidelines both in the USA [22] and other parts of the world [23], when measured up against the Six Core Elements of Health Care Transition 3.0 (2018) [49]. Successes attained by current transition services are therefore difficult to measure given the lack of benchmark to compare with, a void that this study has intended to fill with regard to colorectal transition.

Among the many challenges faced, one prominent issue is the patients' lack of understanding of their conditions [32–38] leading to reliance on and emotional attachment to their pediatric providers and parents [33,36,38–41]. During a long and complex childhood follow-up, the lapse of time could render significant details related to their care difficult to be recalled and to be explained to the patients affected by either ARM or HD in their teenage years, especially without the full benefit of clear standardized record. We believe having a comprehensive understanding of their clinical condition is crucial to patients making appropriate dietary adjustment, adhering to medication regimen, and engaging in bowel management program, where appropriate. For patients with urological and renal anomalies, particularly in ARM, the ability to attend follow-up appointments for investigation and treatment can significantly impact long-term renal function and continence outcomes [10]. This insight emphasizes the need for a novel and more sustainable approach to empower patients (and carers) in obtaining, maintaining, and managing their health information in the long term independently.

For healthcare providers, another major issue is inadequate preparation of adult colorectal surgical teams, who are expected to

Table 3
Challenges associated with transitional care.

Challenges and barriers to successful transition of patients to adult services		
Patient	Healthcare provider	Healthcare system
Lack of patient education and awareness of healthcare condition [32–35,37,38]	Adult surgeons have inadequate education and awareness [32–38,40–42]	Lack of a structured transition program [32,33,36,38–40]
Reluctance of patient or parent to transition away from current pediatric surgeon [33,36,38–41]	Inadequate communication between pediatric and adult surgeon [32,35–38,41–43]	Poor adult clinic organization and set-up [33–36,41,42]
Inadequacy of insurance cover [35,39]	Absence of discussion between surgeon and patient about transition process [35]	Inadequate funding allocated to transition services [43,44]
Lack of patient self-advocacy and initiative [33,39]	Lack of interest from adult surgeons [44]	Inadequate multi-disciplinary provision to bolster transition process [35,38]
Developmental immaturity of patient [36]		
Absent or unsupportive caregiver [36]		

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Table 4
Solutions associated with transitional care.

Solutions identified for overcoming the challenges in transition patients		
Patient	Healthcare provider	Healthcare system
Develop patient autonomy and independence [33,38,44]	Conduct a joint pediatric and adult surgeon clinic [33,38,44–47]	Create a structured & standardized guideline of the transition process [33,38,44]
Re-visit education and training specific to patient's condition requiring healthcare [33,43]	Promote patient autonomy and independence [33,38,44]	Provide written information about transition process [38,44,45]
Join support groups, face-to-face or online [43]	Training and education for adult surgeons [44,47]	Provide access to a counsellor or psychologist as part of transition program [33,44,47]
		Centralize transition services [38,47]
		Employ a transitional care coordinator [38,44]
		Utilize an evidence-based transition risk-assessment score to objectively stratify healthcare needs [33,46]
		Organize or signpost support groups, face to face or online [43,44]
		Provide professional affiliation and funding for support groups [43]

take over the care of these complex patients, during the transition. These teams often lack the necessary clinical information, knowledge, and experience to effectively meet the needs of these patients [32–42]. This is compounded by a healthcare system that lacks structure and policy or condition-specific guidelines due to constraint in resources [32–42]. As a result, these patients and their families remain 'stuck' in the pediatric system or 'lost' entirely. Whilst challenges exist at various levels, the literature suggests an overarching issue of inadequate communication between adult and pediatric surgical teams [32,35–38,41–43]. Therefore, hospital teams within reasonable geographical proximity should form strong transition network for better collaboration. Both pediatric and adult colorectal teams share the responsibilities of developing joint transitional services and supporting these children through combined clinics [33,38,44] and facilitated by a transition coordinator [38,44,46]. Collaborative learning and education between pediatric and adult colorectal teams can be fostered through co-working and potentially co-operating in the operating room.

Tools for needs-evaluation are available to assess patients individually, such as the transition risk assessment score (TRAS), which allows for risk-stratification to determine and target the frequency of follow-up, assessment of function in the long term and interventions required [46]. During the routine follow-up period, standardized functional scoring system can also be used as long-term monitoring tool for bowel, urological, sexual, and psychological functions, as well as quality-of-life assessment, in a consistent and standardized manner [33,46].

Encouragingly, our findings indicate that efforts to address these issues, albeit partial, have yielded positive results. It is now evident that a multimodal approach, starting with the establishment of a clear transition program for HD and ARM, can facilitate multidisciplinary implementation. Transitional clinics provide a supportive environment for children and young adults to ask questions, be treated as individuals to promote maturity, and to have their needs assessed [33,38,44]. A study has demonstrated that attendance at a multidisciplinary pediatric colorectal clinic focusing on education significantly increased the health literacy among caregivers after 3–4 visits [50]. This suggests that young adults undergoing the transition process themselves can benefit from starting early and placing equal emphasis on education. Through accurate information-sharing and parental coaching, young adults' literacy can be enhanced, and autonomy attained.

During the peri-transition period, young adults should be encouraged to join support groups such as Pull Through Network (PTN) [51] and the Research, Education, and Awareness for Children with Hirschsprung's Disease (REACH) [52], for ARM and HD, respectively. They form a life-long source of information as well as a channel for discovering practical strategies to tackle daily challenges of living with their condition.

One importance of this study is its call for a more holistic approach that prioritizes information-sharing and close follow—up processes from birth. The demand for better care is increasing. Therefore, the next generation of colorectal transitional care, like other congenital childhood conditions, must be equipped to provide up-to-date and comprehensive health information, potentially powered by electronic health records and digital platforms accessible to patients to address the issues highlighted here.

This study has been designed based on the '*pragmatism*' approach described by the late philosopher, John Dewey [53], through clear definition of the problems, followed by exploring solutions and their consequences derived from experiences of others who strived to provide transitional care in the past two decades. The strengths of this systematic review include adoption of a robust protocol, with qualitative data from selected papers which are mostly of high or very high quality (94 %) even though, by nature, are of low evidence levels. The analyses were carried out systematically with a protocol set *a priori* following PROSPERO registration. Therefore, our results are presented in a format that promotes pragmatic clinical utilization. Recognizing the qualitative nature of our data, the themes related to challenges and solutions could have overlapped between groups within our categorization system. In addition, the order of importance of these themes within each category had only considered the number of studies and participants involved in the studies, which is inherently arbitrary by nature and may not constitute a rigorous quantitative assessment. Nonetheless, as expected of qualitative research, the repetitiveness of several themes described would reach saturation so much so that the pertinent points made would become apparent [54].

5. Conclusion

Based on the recent 15 years of literature, this systematic review identifies 14 barriers hindering progress in transitional care in ARM and HD, stemming from inadequate communication, inconsistent engagement, and the lack of planning and funding of the current healthcare systems. While generic guidelines for transitional care are available, there has been a lack of condition-specific guidelines for these conditions. This review offers a unique perspective for systematically examining challenges and solutions specific to

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pediatric colorectal practice. A framework of standards has been established through a summary of themed issues, which may translate into actionable and measurable outcomes for guideline development, to improve the quality of future transition services.

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Appendix 1

Search strategies

OVID (EMBASE, MEDLINE)

((“Anorectal malformation*" OR “Hirschsprung* disease” OR “Anorectal anomaly*" OR "Hirschsprung's Disease” OR “HD” OR “Colorectal anomaly” OR “Colorectal anomalies” OR “Colorectal defects” OR “Colorectal disorders” OR “Colorectal conditions” OR “cloaca” OR “cloacal”) AND.

(“Follow-up studies” OR “Lost to follow-up*" OR “Challenge$" OR “Barrier$" OR “Assessment$" OR “Score$") AND.

(“Care, transitional” OR “Transition to Adult Care” OR “Continuity of Patient Care” OR “Transition to adult services” OR “Transitional Care”)).

SCOPUS

((“Anorectal malformation*" OR “Hirschsprung* disease” OR “Anorectal anomaly*" OR "Hirschsprung's Disease” OR “HD” OR “Colorectal anomaly” OR “Colorectal anomalies” OR “Colorectal defects” OR “Colorectal disorders” OR “Colorectal conditions” OR “cloaca” OR “cloacal”) AND (“Follow-up studies” OR “Lost to follow-up*" OR “Challenge$" OR “Barrier$" OR “Assessment$" OR “Score$") AND (“Care, transitional” OR “Transition to Adult Care” OR “Continuity of Patient Care” OR “Transition to adult services” OR “Transitional Care")).

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((“Anorectal malformation*" OR “Hirschsprung* disease” OR “Anorectal anomaly*" OR "Hirschsprung's Disease” OR “HD” OR “Colorectal anomaly” OR “Colorectal anomalies” OR “Colorectal defects” OR “Colorectal disorders” OR “Colorectal conditions” OR “cloaca” OR “cloacal”) AND (“Follow-up studies” OR “Lost to follow-up*" OR “Challenge$" OR “Barrier$" OR “Assessment$" OR “Score$" OR “Scoring system” OR “Long-term care” OR “Long term care”) AND (“Care, transitional” OR “Transition to Adult Care” OR “Continuity of Patient Care” OR “Transition to adult services” OR “Transitional Care")).

Appendix 2

Reasons for exclusion

Authorship	Reason for exclusion
Michihata et al.	Study does not offer any meaningful, primary data for the purpose of this systematic review
Chulani et al.	Study does not offer any meaningful, primary data for the purpose of this systematic review
Hartman et al.	Study published in 2006. This is outside the scope of this systematic review
Lemacks et al.	Study does not offer any meaningful, primary data for the purpose of this systematic review
Ebert et al.	Study does not offer any meaningful, primary data for the purpose of this systematic review
Danielson et al.	Study does not offer any meaningful, primary data for the purpose of this systematic review
Schmidt et al.	Study does not offer any meaningful, primary data for the purpose of this systematic review
Blyth et al.	Focus of study is on transition of patients with gynaecological pathology, not ARM or HD
Acket et al.	Study does not offer any meaningful, primary data for the purpose of this systematic review

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Appendix 3

ENTREQ statement

Item	Guide and description	Reported on page
Aim	State the research question the synthesis addresses.	4
Synthesis methodology	Identify the synthesis methodology or theoretical framework which underpins the synthesis, and describe the rationale for choice of methodology *(e.g. meta-ethnography, thematic synthesis, critical interpretive synthesis, grounded theory synthesis, realist synthesis, meta-aggregation, meta-study, framework synthesis).*	5
Approach to searching	Indicate whether the search was pre-planned (*comprehensive search strategies to seek all available studies*) or iterative (*to seek all available concepts until they theoretical saturation is achieved*).	5
Inclusion criteria	Specify the inclusion/exclusion criteria *(e.g. in terms of population, language, year limits, type of publication, study type).*	5, 6
Data sources	Describe the information sources used (*e.g. electronic databases (MEDLINE, EMBASE, CINAHL, psycINFO, Econlit), grey literature databases (digital thesis, policy reports), relevant organisational websites, experts, information specialists, generic web searches (Google Scholar) hand searching, reference lists*) and when the searches conducted; provide the rationale for using the data sources.	5
Electronic Search strategy	Describe the literature search *(e.g. provide electronic search strategies with population terms, clinical or health topic terms, experiential or social phenomena related terms, filters for qualitative research, and search limits).*	5
Study screening methods	Describe the process of study screening and sifting *(e.g. title, abstract and full text review, number of independent reviewers who screened studies).*	6,18
Study characteristics	Present the characteristics of the included studies *(e.g. year of publication, country, population, number of participants, data collection, methodology, analysis, research questions).*	20,21
Study selection results	Identify the number of studies screened and provide reasons for study exclusion *(e,g, for comprehensive searching, provide numbers of studies screened and reasons for exclusion indicated in a figure/flowchart; for iterative searching describe reasons for study exclusion and inclusion based on modifications t the research question and/or contribution to theory development).*	18,19
Rationale for appraisal	Describe the rationale and approach used to appraise the included studies or selected findings *(e.g. assessment of conduct (validity and robustness), assessment of reporting (transparency), assessment of content and utility of the findings).*	6,7
Appraisal items	State the tools, frameworks and criteria used to appraise the studies or selected findings *(e.g. Existing tools: CASP, QARI, COREQ, Mays and Pope* [25]*; reviewer developed tools; describe the domains assessed: research team, study design, data analysis and interpretations, reporting).*	6,7
Appraisal process	Indicate whether the appraisal was conducted independently by more than one reviewer and if consensus was required.	6,7
Appraisal results	Present results of the quality assessment and indicate which articles, if any, were weighted/excluded based on the assessment and give the rationale.	22
Data extraction	Indicate which sections of the primary studies were analysed and how were the data extracted from the primary studies? *(e.g. all text under the headings "results/conclusions" were extracted electronically and entered into a computer software).*	6, 7
Software	State the computer software used, if any.	n/a
Number of reviewers	Identify who was involved in coding and analysis.	n/a
Coding	Describe the process for coding of data *(e.g. line by line coding to search for concepts).*	n/a
Study comparison	Describe how were comparisons made within and across studies *(e.g. subsequent studies were coded into pre-existing concepts, and new concepts were created when deemed necessary).*	7
Derivation of themes	Explain whether the process of deriving the themes or constructs was inductive or deductive.	7
Quotations	Provide quotations from the primary studies to illustrate themes/constructs, and identify whether the quotations were participant quotations of the author's interpretation.	n/a
Synthesis output	Present rich, compelling and useful results that go beyond a summary of the primary studies (*e.g. new interpretation, models of evidence, conceptual models, analytical framework, development of a new theory or construct).*	23, 24

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