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Benefits, implementation and sustainability of innovative paediatric models of care for children with type 1 diabetes: a systematic review

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Abstract

Background and aim The evidence about the acceptability and effectiveness of innovative paediatric models of care for Type 1 diabetes is limited. To address this gap, we synthesised literature on implemented models of care, model components, outcomes, and determinants of implementation and sustainability.

Methods A systematic review was conducted and reported in accordance with Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines. Database searches of Medline, CINAHL, EMBASE and Scopus were conducted. Empirical studies focused on Type 1 diabetes paediatric models of care, published from 2010 to 2022 in English were included.

Results Nineteen extant studies reported on models and their associations with health and psychosocial outcomes, patient engagement with healthcare, and healthcare costs. Thirteen studies described multidisciplinary teamwork, education and capacity building that supported self-care. Four studies involved shared decision making between providers and patients, and two discussed outreach support where technology was an enabler. Fourteen studies reported improvements in health outcomes (e.g. glycaemic control), mostly for models that included multidisciplinary teams, education, and capacity building (11 studies), outreach support or shared care (3 studies). Four studies reported improvements in quality of life, three reported increased satisfaction for patients and carers and, and one reported improved communication. Four of five studies describing shared care and decision-making reported improvements in quality of life, support and motivation. Outreach models reported no negative outcomes, however, accessing some models was limited by technological and cost barriers. Eight studies reported on model sustainability, but only half reported implementation determinants; none reported applying a theoretical framework to guide their research.

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Conclusion Some health and psychosocial benefits were associated with newer models. To address knowledge gaps about implementation determinants and model sustainability, longitudinal studies are needed to inform future adoption of innovative models of care for children with Type 1 diabetes.

Keywords Type 1 diabetes, Children, Families, Model of care, Innovation, Person-centred care, Multidisciplinary teamwork, Health outcomes

Background

Type 1 diabetes (T1D) is typically diagnosed in childhood. In 2021 there were approximately 8.4 million individuals worldwide with T1D and of these, 1.5 million (18%) were younger than 20 years [1]. In Australia, T1D affects around 140,000 young people [2]. Despite advances in care, children and adolescents who develop T1D have a reduced life expectancy by around 12-16 years compared with those without T1D [3, 4]. Increased morbidity and mortality is attributed to an ongoing gap between the recommended glycaemic control levels and the levels achieved in practice, and this poor glycaemic management can lead to cardiovascular disease [5], nephropathy and mental health disorders [6]. Increased attention on the models used to deliver care is needed to better understand how and whether these are associated with improved outcomes, e.g., optimising the maintenance of glycaemic levels among children with T1D.

Paediatric care for T1D is complex as there are a range of developmental stages to consider. For younger children (<14 years) care tends to be family orientated, as they lack the maturity to self-manage their care delivery and planning [5], whereas, adolescents are encouraged to take on more responsibility for self-management as they approach adulthood [7]. Successful outcomes require a psychosocial approach with education and support from clinical teams for the child and their family, to empower and motivate them to effectively manage the treatment 24-h a day, seven days a week, and through life events. Commonly, models of care for paediatric T1D involve routine outpatient clinic visits every 3-4 months with a multidisciplinary team (MDT) [8, 9]. Regular consultations are usually undertaken face-to-face with the child and family, with between visit communication initiated by families/carers as needed [10]. MDT care typically includes specialist doctors and nurses, diabetes educators, dieticians and psychologists, and has been proposed as an effective way of providing care to increase safe levels of glycaemia [11]. A model of care involving an MDT approach is one where professionals bring their skills and expertise from a range of disciplines for a combined team effort that responds to the individual child and family needs. Team members typically collaborate to discuss the patient and family and develop shared care plans. Although several implementation barriers have been identified to adoption of such models, including time constraints, inaccessibility, cost and conflicting recommendations from different providers, this approach has the potential to strengthen clinical care and improve health outcomes, especially when it is consumer focused and involves shared-decision making between the provider and patient [12]. Although the International Society for Pediatric and Adolescent Diabetes (ISPAD) guidelines provide evidence based recommendations for care [13], it is not known whether these are being implemented in practice. Additionally, there have been no systematic reviews to describe current models of care, nor a synthesis of current evidence for the effectiveness of models of care for T1D. It is unclear how the effectiveness of models of care has been conceptualised and, in examining these models, what types of study designs have been used and which outcomes have been measured.

Our group recently conducted a systematic review on models of care for individuals with T1D transitioning from paediatric to adult health care [14]. We identified a gap in the evidence about the implementation determinants and the sustainability of models of care, with few studies applying longitudinal designs to measure longterm outcomes. Given this gap in the transition context, a synthesis of evidence about the implementation determinants and sustainability of non-transitional models of care for T1D is needed.

This review aims, first, to map the scope of implemented models of care for children living with T1D, to describe the model components, how and to what extent these contribute to improved health, psychosocial and health services outcomes. Second, we aim to synthesise evidence about factors associated with implementation and sustainability of effective models of care to inform future adoption of successful models at scale and in different contexts.

Methods

Review protocol

The studies in this review were identified in the results of a concurrent review that reports on models of care for transitioning young adults living with T1D [14]. As such, the methodology sections are similar. The systematic review was guided by the Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for Systematic Reviews (PRISMA) guidelines [15] and follows a registered protocol (CRD42021262727) which is available here: https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=262727.

Search methods

The review team met with a medical librarian to design the search strategy. Four databases were searched: Scopus, Medline, CINAHL, and EMBASE on 6th June 2021 and updated on 11th November 2022. An example of the search strategy is provided in Supplemental File 1. Search limits were applied to include to publications in English, published from January 2010 to November 2022. Snowballing techniques identified other relevant publications in the reference list of included studies and these were screened according to the inclusion and exclusion criteria.

Inclusion and exclusion criteria

We included peer-reviewed articles and reviews describing the intervention of a person centred model of care implemented in high income countries [16], and patients under the age of 18 years diagnosed with T1D. Studies that described telehealth, in addition to describing other model components, that was facilitated and delivered by MDT approaches or innovative diabetes education were included. Studies had to report on health related and/or psychosocial outcomes, and/or experiences from the perspective of the health provider, health consumer, and/or caregiver.

Studies that were published prior to 2010, in a language other than English, were conducted in a low- or middle-income country, focused on transitional care of T1D, Type 2 diabetes or maternal health interventions or clinical interventions, e.g., clinical trials involving drugs or specific equipment were excluded. Other reasons for exclusion were commentaries, publications of opinion or perspective, letters to the editor, editorials, and conference abstracts. We also excluded studies that discussed routine consultations through telehealth without a description of a broader model of care.

Study selection

The reference details for articles returned from search results were downloaded to a folder and duplicates were removed and exported into Rayyan, an electronic literature screening program [17]. Ten percent were screened independently by IM and MS, and a separate sample of 10% were screened by YZ and IM. For the updated searches, all title/abstract and full text screening was undertaken independently by two reviewers (AC, GD). Any disagreements among reviewers were resolved by discussion with the whole review team. The full text of selected studies was then examined. Studies were excluded if they failed to meet the inclusion criteria on full-text screening.

Data extraction and synthesis

A custom data extraction workbook was created in Microsoft Excel. The workbook underwent pilot testing on five articles, and adjustments were made as necessary to accommodate the different types of data reported in the articles. Four reviewers (AC, MS, NH, RL) systematically extracted the data, and any discrepancies among the reviewers were resolved through discussion. The relevant information extracted included: specific publication details such as authors, year published, country; design, methods and setting; patient details such as age, gender, race/ethnicity, socio- economic status, mean duration of diabetes, and insurance status; description of the model of care, their components, staffing, resources, and setting; a description of usual care, outcomes related to health, psychosocial aspects, or health service use, the utilisation of an implementation framework, and the determinants, enablers, barriers, and adoption of the model into practice (Table 2). Common themes, features specific to the model, and categories of outcomes were extracted by the research team.

Results

The search for primary studies yielded 1951 results (CINAHL: 727, EMBASE: 572, Medline: 446, Scopus: 205; identified from other sources: 1). Among these, 361 duplications were removed. After screening 1590 title/ abstracts, 1313 papers were excluded as they did not meet the inclusion criteria. Two hundred and seventy studies underwent full text review and a further 258 papers were excluded. Nineteen primary studies were included for data extraction and synthesis, as shown in Fig. 1. Interrater reliability scores (Cohen's kappa) [18] were all above k=0.6, which is considered a "good" interrater reliability score.

Quality assessment

Methodological quality was appraised using the Mixed Methods Appraisal Tool (MMAT) [19]. To ensure consistency, two investigators (AC and RL) appraised 10% of the articles independently. Twelve of the 19 studies reported a quantitative non-randomised design and included a representative sample and complete outcome data [9, 11, 20-29], however, two of these studies did not account for confounders [9, 29]. There were six randomised controlled trials that reported appropriate randomisation and complete outcome data [8, 10, 30-33], however, outcome assessors were not blinded, potentially introducing a bias. One study reported a qualitative design that adopted an appropriate approach, adequate data collection and substantiated interpretation [34]. We did not exclude any studies based on quality assessment. See Supplemental File 2 for further detail.

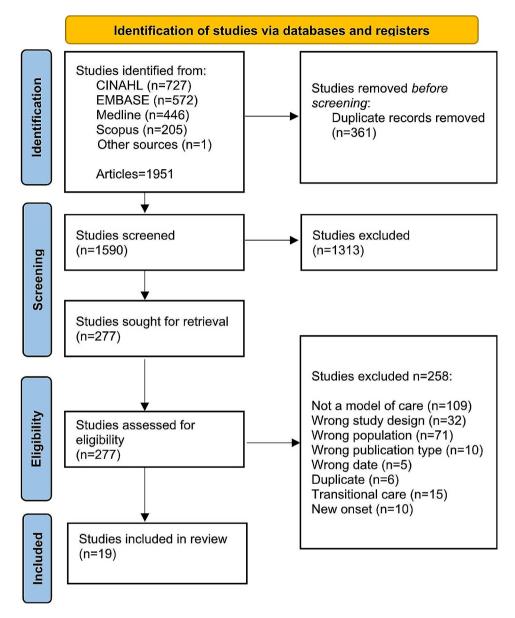


Fig. 1 PRISMA flow chart describing the study selection process

Scope of models

Most studies (13/19, 68%) were from the USA [8–10, 20–22, 24, 26, 27, 30, 31, 33, 34]. The remaining studies were from Australia (4/19, 22%) [11, 23, 25, 28], Canada (1/19, 5%) [29], and Denmark (1/19, 5%) [32], (Table 1). Most studies were conducted using quantitative methodologies (12/19, 63%) [9, 11, 20–29], a third used randomised controlled trial designs (6/19, 32%) [8, 10, 30–33], and there was one qualitative study (1/19, 5%) [34]. The models of care described in the 19 included papers were comprised of several components such as MDT care, shared decision making, capacity building and education for self-care, remote monitoring, and outreach support (Table 1).

Model components

Although there was often overlap among the components (e.g., MDT, shared care and outreach), common components and approaches to care provision were identified as demonstrated in Fig. 2. These included MDT care, shared decision making, capacity building and education to support self-care, outreach support and mentoring and integrated care where technology such as a paging system was used as an enabler.

Multidisciplinary team care, education and capacity building for self care

MDT care was a central model component across 13 studies (68%). Teams provided T1D care in different contexts including in hospital settings, outpatient clinics,

Table 1 Key characteristics of the included studies

Study	Country	Setting	Model of care components as implemented	Inter- vention duration	Time at which out- comes measured	Par- tici- pants (n)	Age (years)**
Bergmann et al. 2020 [20]	USA	Metropolitan tertiary care paediatric clinic	Child education and MDT	2 years	Pre-implementation: 4 years, 2 months. Post-implementation: 2 years, 8 months	67	Median (IQR) 15 (1–17)
Ellis et al. 2017 [30]	USA	Community paediatric diabetes clinic	Capacity building for self-care (family education) and MDT	6 months	Baseline and at 7 months.	47	Mean 14
Fiallo-Sharer et al. 2019 [8]	USA	Metropolitan and rural multi- disciplinary diabetes clinics	Capacity building for self-care (family education) and MDT	9 months	Baseline and 3 monthly for 2 years	214	Range 8–16
Floyd et al. 2017 [21]	USA	Metropolitan tertiary care paediatric clinic (University)	Capacity building for self-care (family education) and MDT	9 months	Baseline and at 3, 6, and 9 months	32	Range 12–16
Franklin et al. 2014 [22]	USA	Metropolitan tertiary care children's hospital	Outreach support using a pager service	32 months	Retrospective analysis of visits to ED	979	Mean 11.1
Hannon et al. 2018 [31]	USA	Metropolitan tertiary care academic medical centre	Integrated care where technol- ogy is an enabler	6 months	Baseline, 3 and 6 months	128	Mean 14.7
Hatherly et al. 2011 [23]	Australia	Rural clinics	Shared decision making/inte- grated care	7 months	Baseline	158	Mean 12.9
Herbert et al. 2024 [34]	USA	Metropolitan children's hospitals	Capacity building for self-care (family education) and MDT	Not stated	Baseline	13 child/ par- ent dyads	Mean 15.7
Husted et al. 2014 [32]	Denmark	Metropolitan paediatric outpatient clinics	Shared decision making/inte- grated care	8 to 12 months	Baseline, every 3 months	43	Mean 14.7
Gandrud et al. 2018 [10]	USA	Metropolitan paediatric outpatient clinic	MDT remote monitoring	6 months	6 and 9 months	117	Mean 12.7
Goss et al. 2010 [11]	Australia	Rural outreach clinics	Rural MDT	3 years	Baseline, 12, 24, 36 months	221	Mean 14
Ilkowitz et al. 2016 [24]	USA	Metropolitan tertiary care centre	Capacity building for self-care (family education) and MDT	5 years	Baseline (3 years), post intervention (2 years)	1119	Mean 14
Joshi et al. 2017 [25]	Australia	Metropolitan tertiary care hospital and regional out- reach clinics	Outreach support	12 months	12 months	1017	Mean 12.8
Katz et al. 2012 [26]	USA	Home care	Capacity building for self-care (family education) and MDT	Not stated	Baseline	583	Range 0–17
Nansel et al. 2012 [33]	USA	Metropolitan paediatric endocrine clinics	Capacity building for self-care (family education) and MDT	2 years	Baseline, every 3–4 months, 24 months	390	Mean 12.5
Pascual et al. 2019 [27]	USA	Metropolitan childhood diabetes centre	Education and MDT	2 years	Baseline, 12, 24 months	88	Range 8.4–14.6
Simm et al. 2014 [28]	Australia	Metropolitan tertiary outpa- tient and rural clinics	Outreach support and MDT	6 years	Every 3–4 months	1456	Mean 10.7
Versloot et al. 2022 [29]	Canada	Community paediatric clinics across three sites	Integrated stepped care	34 months	Baseline, 12 months	116	Mean 14.5
Wan et al. 2022 [9]	USA	Metropolitan Centre for Diabetes	Education and MDT	6 months	Baseline, 3 and six months	86	Mean 11.8

USA: United States of America; ED: emergency department; MDT: multidisciplinary team

** Age was reported by different studies as either a mean, median, range or interquartile range (IQR)

outreach services or via telemedicine. MDTs were comprised of medical staff such as paediatric emergency medicine physicians, paediatric endocrinologists, primary healthcare providers as well as nurse practitioners, mental health nurses, dieticians, pharmacists, and patients. MDT model components and the work of MDTs often included education for patients and providers [20, 27]; shared medical appointments that involved the child living with T1D, their caregivers and multiple healthcare providers [9]; capacity building for patient self-care and education [8, 24, 26, 30]; education and remote monitoring in outreach and rural settings [10,

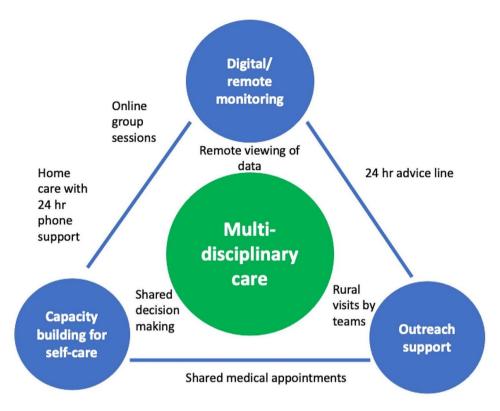


Fig. 2 Components of T1D paediatric models of care

11]; and support between clinic visits from diabetes nurse educators for patients residing in rural settings [28]. The structure and work of the MDT models was variable with no model being described in the same way. Examples include MDTs implementing and embedding evidencebased treatment guidelines into their practice for patients with diabetic ketoacidosis (DKA) [20]; a Spanish speaking MDT involved in shared medical appointments (SMA) for Latino children with T1D [27], and an MDT model where individuals and caregivers participated in SMAs involving education and clinical care [9]. Another MDT was involved in weekly review of outpatient data uploaded from an activity monitor, insulin pump and glucose sensor [10].

Capacity building for self-care included family education programs comprised of education initiatives offered to patients and families in diabetes clinics. A key component was the exploration and identification of self-management barriers that subsequently lead to solutions such as the design and delivery of tailored self-management education resources [8, 30, 33], and coping skills programs [34]. These included tailored delivery of resources to help families improve diabetes management [8], training sessions on cognitive behavioural skills and relapse prevention to improve maintenance of safe glycaemic levels [30], and training in insulin pump usage and the basal bolus regimen [24]. Others described accessible, co-ordinated, family-centred care [26], individualised treatment plans for patients to improve glycaemic maintenance and quality of life, and group support to increase knowledge about T1D, goal setting, and glucose pattern recognition [21]. Additionally, identification of child and family psychosocial issues enabled the development of a stepped care model where the intensity of clinical intervention was matched to each case as needed [29]. Concerns were discussed and addressed among the individuals living with T1D, their caregiver and the care team [29].

Shared decision making

A shared decision making model was described by Hatherly et al. (2011) [23], where paediatric endocrinologists from large metropolitan hospitals provide outreach and travel to rural and regional sites to integrate care with a primary health care provider and patients, to empower patient/family relationships with specialist physicians, nurses and dieticians, providing care that includes self-reflection and the development of life skills [32]. Integrated care with technology as an enabler included real-time mobile health technology that shares patient blood glucose data with their parents, and clinicbased healthcare providers facilitated shared decision making [31].

Outreach support

Outreach support often involved digital and remote monitoring and was described in three studies (16%) [22, 25, 28]. Outreach rural services included a visiting medical specialist who attended routine medical appointments at three-month intervals and follow up by allied health teams or diabetes nurse educators between appointments [28]. In a study in regional and remote Western Australia, all patients are offered 24-h, 7 days-a-week telephone support with an in-hospital or outreach team, and four visits from a metropolitan-based outreach team per year [25]. Triaging in remote settings was undertaken using a paging system that families could access [22].

Model outcomes and effectiveness

Many different outcomes were measured and reported in the 19 included studies. Outcomes could be grouped under the broad categories of health, psychosocial, satisfaction, cost, hospital admissions, regimen adherence and other (Table 2).

Table 3 presents the evidence for effectiveness based on the outcomes, stratified by model components.

Glycaemic control was the most frequently reported health outcome. Five studies (26%), involving MDT, education and capacity building for self-care, reported significant improvements in glycaemic control associated with the model of care [8, 11, 27, 30, 33]. Three studies (16%) reported no change in HbA1c levels indicating that the model of care was comparable to routine care [9, 10, 21]. Two studies (11%) reported a significant reduction in DKA and hospital admissions [20, 24], another two studies (11%) reported comparable insulin regimen adherence compared with usual care [30, 33], one study (5%) showed an increase in insulin pump usage for Latino patients [27], and one study (5%) reported comparable insulin pump usage compared to usual care [9] (Table 3).

For studies describing shared care and decision making, the evidence for health benefits was mixed. Two studies (11%) reported no change in HbA1c levels (perceived benefit) [23, 32], one study (5%) reported short term benefits at three months but this effect was not replicated at six months [31]. One study (5%) reported an increase in HbA1c across 18 months compared to baseline [29]. Compared with usual care, four studies (21%) describing outreach support and mentoring, reported comparable health outcomes including similar levels of HbA1c [25, 35], and episodes of severe hypoglycaemia [25, 28], or DKA [22, 25] (Table 3).

Quality of life and psychosocial outcomes are important measures of the effectiveness of care; however, benefits were not consistently reported. For MDT, education and capacity building for self-care models three studies (16%) reported improved quality of life [11, 21, 30], one (5%) reported better communication [21], and three (16%) reported improved satisfaction for caregivers [30] and patients [27, 34]. Three studies (16%) reported improved quality of life [29], greater support

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Table 2 Outcomes for T1D models of care

Outcome	Outcome measures and citation	Num- ber of stud- ies (%)
Health	Glycaemic control [8–11, 21, 23, 25, 27, 29–33]	13 (68)
	Hypoglycaemia [25, 28]	2 (11)
	Diabetic ketoacidosis [22, 25]	2 (11)
	Continuous glucose monitor usage [9]	1 (5)
	Insulin pump usage [9, 27]	2 (11)
	Insulin regimen adherence [21, 30, 33]	3 (16)
	Hospital admission rate due to diabetic ketoacidosis [20, 22, 24]	3 (16)
Psychosocial	Quality of life [8, 10, 11, 21, 29, 30]	6 (32)
	Psychosocial function [21]	1 (5)
	Diabetes related quality of life [29]	1 (5)
	Diabetes support for children and families [23]	1 (5)
	Perceived competence in diabetes [32]	1 (5)
	Treatment self-regulation [32]	1 (5)
	Degree of autonomy support from health- care provider [32]	1 (5)
	Perceived burden of diabetes related problems [32]	1 (5)
	Amotivation [32]	1 (5)
	Emotional well-being [32]	1 (5)
	Impact of T1D on family [26]	1 (5)
	Communication [21]	1 (5)
	Parent perception [32]	1 (5)
Satisfaction	Patient satisfaction [27, 34]	2 (11)
	Caregiver satisfaction [25, 30]	2 (11)
Cost	Emergency department charges [20]	1 (5)
	Health care use and cost [9]	1 (5)
Other	Ability to function at school [23]	1 (5)
	Attended diabetes educator [23]	1 (5)
	Income [23]	1 (5)
	Visits to clinic [28]	1 (5)
	Number of clinic visits [9]	1 (5)

from a diabetes educator [23], and improved motivation [32]. For outreach support, one study (5%) reported an increased number of rural clinic visits [28].

Two studies (11%) compared healthcare costs for MDT, education and capacity building for self-care models with usual care [9, 20], with one reporting a decrease in ED charges because of decreased presentations of DKA [20]. A further study showed a reduction in the negative impact of T1D on factors such as school attendance and finances for families involved in medical care in their homes compared to usual care [26].

Implementation determinants and model sustainability

None of the 19 studies mentioned implementation frameworks or theories to guide their adoption of multidisciplinary models of care. Twelve studies (63%) discussed

Table 3 Evidence for effectiveness of model components stratified by outcome type

Model compenents	Outcome	Variable measured	Study	Evidece	Benefit?
MDT, education, and capac- ity building for self-care	Health	Glycaemic control	Ellis et al. 2017 [30]	Clinically significant (0.7%) improvements in HbA1c for those who received an individualised treatment plan and training sessions ($p = 0.05$).	Benefit
		Glycaemic control	Fiallo- Scharer et al. 2019 [8]	No difference in mean HbA1c levels between those who received tailored self-management resources to identify barriers and subsequent delivery of resources, compared with usual care. However, there was a significant improvement in HbA1c levels for teens (ages 13–16 years) at one site compared with usual care (9.68% vs. 10.76%, p < 0.05).	Mixed
		Glycaemic control	Floyd et al. 2017 [<mark>21</mark>]	No change in HbA1c levels for those engaged in SMAs ($p = 0.98$).	No benefit
		Glycaemic control	Gandrud et al. 2018 [<mark>10</mark>]	Compared with conventional care, intensive remote monitoring combined with outreach resulted in a statistically non-significant decrease in HbA1c levels of 0.34 (3.7 mmol/mol, $p = 0.071$) at 6 months.	No benefit
		Glycaamic control	Goss et al. 2010 [11]	Compared with usual care, the intervention group who received education and support via a rural MDT model of care had significantly lower levels of HbA1c (9.6% vs. 8.1%, $p < 0.001$) from 2006 to 2009.	Benefit
		Glycaemic control	Nansel et al. 2012 [33]	There was a significant decrease in HbA1c levels for a clinic integrated behavioural intervention compared with usual care observed at 24 months (0.44 vs. 0.76, p =0.03). (Ages 12 to 14 years, p =0.009; ages 9 to 11 years no effect).	Mixed
		Glycaemic control	Pascual et al. 2019 [27]	Routine care with SMAs for Latino patients resulted in a significant improvement in HbA1c levels from baseline to year 1 (p =0.0146) and from year 1 to 2 (0.0082), compared to routine care alone.	Benefit
		Glycaemic control	Wan et al. 2022 [9]	There was no significant difference in HbA1c levels for patients undertaking SMAs compared to usual care ($p = 0.49$).	No change (benefit)
		Continuous gly- caemic monitor (CGM) usage	Wan et al. 2022 [9]	There was no significant difference in CGM usage for patients undertaking SMAs compared to usual care (1 vs. 4, $p = 0.18$).	No change (benefit)
		Insulin pump usage	Pascual et al. 2019 [27]	Routine care with SMAs for Latino patients resulted in a signifi- cant increase in insulin pump usage from baseline to year 2 for participants aged < 12 years (p = 0.0455) and > 12 years (p = 0.0027) compared to routine care alone.	Benefit
		Insulin pump usage	Wan et al. 2022 [<mark>9</mark>]	There was no significant difference in insulin pump usage for patients undertaking SMAs compared to scare (18 vs. 13, $p = 0.16$).	No change
		Insulin regimen adherence	Ellis et al. 2017 [<mark>30</mark>]	Non-significant improvements based on self-report for those receiving individualised treatment plans with cognitive-behavioural skills training ($p > 0.05$).	No change (benefit)
			Nansel et al. 2012 [<mark>33</mark>]	No effect for a clinic integrated behavioural intervention compared with usual care based on child and parent reports ($p > 0.05$).	No change (benefit)
		Hospital admis- sion due to DKA	Bergmann et al. 2020 [<mark>20</mark>]	A quality improvement initiative resulted in a decreased rate of hospitalisations compared to usual care (74% vs. 55%, p = 0.011).	Benefit
			Ilkowitz et al. 2016 [24]	A three-pronged intervention (intensive insulin management, education program, increased access to appointments and support events and groups resulted in a decrease in DKA admissions (16.7% vs. 9.3%, $p = 0.006$), and a decrease in unique patient 30-day readmissions (20% vs. 5%, $p = 0.001$), and a reduction in length of stay ($p < 0.0001$).	Benefit
		Clinic visits	Wan et al. 2022 [9]	There were significantly more clinic visits for the SMA group compared with usual care (2.38 vs. $1.7, p < 0.01$)	Benefit
	Psychosocial	Quality of life	Ellis et al. 2017 [30]	Those receiving individualised treatment plans with cognitive-be- havioural skills training reported improved diabetes related quality of life compared with control group ($p=0.001$).	Benefit

Table 3 (continued)

Model compenents	Outcome	Variable measured	Study	Evidece	Benefit?
			Fiallo- Scharer et al. 2019 [8]	No difference in quality of life for the child with diabetes or their parent between those who received tailored self-management resources, compared with usual care ($p > 0.05$).	No change
			Floyd et al. 2017 [<mark>21</mark>]	Significant improvement in quality of life for those engaged in SMAs with a MDT (p =0.005).	Benefit
			Gandrud et al. 2018 [10]	Compared with conventional care, intensive remote monitoring combined with outreach resulted in no difference in quality of life $(p > 0.05)$.	No change (benefit)
			Goss et al. 2010 [11]	Compared with usual care, the intervention group aged greater than 10 years of age who received education and support via a rural MDT model of care had significant improvements in quality of life.	Benefit
		Communication	Floyd et al. 2017 [<mark>21</mark>]	Significant improvement in communication for those engaged in SMAs with a MDT (p =0.02).	Benefit
		Impact of T1D on the family	Katz et al. [26]	Compared to usual care, a medical home resulted in a 15% reduction in family impact (p =0.01), care coordination resulted in a 18% reduction in family impact (p =0.002), family-centred care resulted in a 10% reduction in family impact (p =0.08).	Benefit
	Satisfaction	Patient	Herbert et al. 2014 [34]	Half reported the TeamWork Project as "pretty helpful". Specifically, the model reinforced current skills. Compared with the education group, the coping group felt that the model helped with communication, problem solving, new perspectives and skills (qualitative data).	Perceived benefit
			Pascual et al. 2019 [27]		Perceived benefit
		Caregiver	Ellis et al. 2017 [30]	All of those receiving individualised treatment plans with cogni- tive-behavioral skills training reported high levels of satisfaction with treatment location and the program.	Perceived benefit
	Cost	Emergency Department	Bergmann et al. 2020 [20]	Total adjusted median ED charges decreased from pre to post implementation period.	Benefit
		Health care use and cost	Wan et al. 2022 [9]	Compared to usual care, there were no differences in 6-month total costs between SMAs and usual care ($p=0.84$).	No change (positive)
	Other	School function	Floyd et al. 2017 [<mark>21</mark>]	Significant improvement in school function for those engaged in SMAs with a MDT (p =0.006).	Benefit
		Treatment adherence	Floyd et al. 2017 [<mark>21</mark>]	Significant improvement in treatment adherence for those en- gaged in SMAs with a MDT ($p=0.01$).	Benefit
		Barriers to treat- ment adherence	Floyd et al. 2017 [<mark>21</mark>]	Significant reduction in barriers for those engaged in SMAs with a MDT ($p = 0.02$).	Benefit
Shared care and decision mak- ing, and integra- tion of care	Health	Glycamic control	Hatherly et al. 2011 [23]	Glycamic control was not compromised in a shared care model compared with specialist care ($p > 0.05$).	No change (positive)
		Glycamic control Glycamic control	Husted et al. 2014 [32] Hannon et al. 2018 [31]	No change in HbA1c levels for those receiving a life skills program compared with usual care ($p = 0.85$). Patients using a combined approach of real-time mobile health technology and family centred goal setting, had a significant decrease in HbA1c levels from baseline to three months, but not at	No change (positive) Mixed
		Glycamic control	Versloot et	six months. For the stepped care model, HbA1c levels worsened between 6–18 months compared to baceling $(n = 0.001)$	No benefit
	Psychosocial	Quality of life	al. 2022 [29] Versloot et al. 2022 [29]	months compared to baseline ($p = 0.001$). Significant improvement in quality of life for those participating in a stepped care model at 6–18 months compared to baseline ($p = 0.048$).	Benefit
		Diabetes-related quality of life	Versloot et al. 2022 [29]	Significantly less diabetes related concerns for those participating in a stepped care model at 6–18 months compared to baseline (p=0.001)	Benefit

Table 3 (continued)

Model	Outcome	Variable	Study	Evidece	Benefit?
compenents		measured Support	Hatherly et al. 2011 [23]	Those receiving shared care were more likely to visit a diabetes educator in the prior 12 months compared with specialist care $(p = 0.02)$.	Benefit
		Amotivation	Husted et al. 2014 [<mark>32</mark>]	Compared with usual care, those receiving a life skills program had significantly reduced amotivation ($p < 0.0001$).	Benefit
		Wellbeing and perceived bur- den of diabetes	Husted et al. 2014 [<mark>32</mark>]	Compared with usual care, those receiving a life skills program had no change in wellbeing or perceived burden of diabetes ($p > 0.05$).	No change
		Parent perception	Husted et al. 2014 [<mark>32</mark>]	Compared with usual care, those receiving a life skills program had no change in parent perception ($p > 0.05$).	No change
	Other	Hospital admissions	Hatherly et al. 2011 [<mark>23</mark>]	Those receiving shared care were more likely to be admitted to hospital in the prior 12 months compared with specialist care ($p = 0.002$).	No benefit
Outreach support	Health	Glycaemic control	Joshi et al. 2017 [<mark>25</mark>]	No change in mean HbA1c for those who attended an outreach clinic compared to a metropolitan clinic ($p = 0.39$).	No change (positive outcome)
		Severe hypoglycaemia	Joshi et al. 2017 [<mark>25</mark>]	No change in severe hypoglycaemic events for those who attended an outreach clinic compared to a metropolitan clinic ($p = 0.74$).	No change (positive outcome)
			Simm et al. 2014 [<mark>28</mark>]	No difference in mean HbA1c levels between an urban diabetes clinic and outreach, rural clinics (8.3% for both groups, $p = 0.87$).	No change (positive outcome)
		Diabetic ketoacidosis	Franklin et al. 2014 [<mark>22</mark>]	Those using a pager service to access care were 2.75 times more likely to visit the ED for DKA, compared to nonusers ($p < 0.00001$).	Benefit
		Diabetic ketoacidosis	Joshi et al. 2017 [<mark>25</mark>]	No change in the DKA rate for those who attended an outreach clinic compared to a metropolitan clinic ($p = 0.68$).	No change (positive outcome)
		ED and inpa- tient admissions due to DKA	Franklin et al. 2014 [<mark>22</mark>]	Those using a pager service to access care were less likely to be admitted to the ED for DKA, compared to nonusers (odds ratio 0.58 , $p < 0.02$).	Benefit
	Other	Number of clinic visits	Simm et al. 2014 [<mark>28</mark>]	Increased number of clinic visits for those who attended a rural, outreach clinic and an urban diabetes clinic ($p < 0.001$)	Benefit

SMA: shared medical appointment; MDT: multidisciplinary team; ED: emergency department; DKA: diabetic ketoacidosis

implementation enablers, and 11 (58%) reported barriers to implementation (Table 4). For models incorporating MDT, education, or capacity building for self-care, the reported enablers included: group-based sessions, [8] care managed by a single facilitator [8, 11], a small team [11], adequate provider to patient ratio [24], integration into existing clinics [33], close collaboration between primary care doctors and the diabetes team in hospital [26], and adopting a multicultural approach [27]. Barriers included length of program being too long for patients to engage with consistently [30], low frequency of sessions [8], lack of skilled staff to cover periods of leave [11], low health literacy of patients and caregivers [26], and a lower drive among older children (>12 years) to undertake diabetes related education [27].

For models that included outreach support, the enablers included having a dedicated and well-trained team [22], the adoption of a MDT approach [25] that is consistent and cohesive, and coupled with effective communication [28]. In one study, support between routine consultations was facilitated by 24-hour phone access to

the diabetes team [25]. Access to clinics for those living in remote regions was a reported barrier by one study [28].

For models using shared care and decision making and integration of care, a reported enabler was having all the model components working well together [21]. A barrier was a lack of access to 24-hour ambulatory care and inequitable access for families of lower socio-economic status due to higher costs [23].

In the assessment of the sustainability of the models or care, three of 19 studies (16%) measured outcomes at two years [20, 27, 33], and five (26%) measured outcomes two years or longer after model implementation [11, 22, 24, 28, 29] (Table 1). Four of these studies (21%) did not mention sustainability of the model of care after implementation, although outcomes were measured [20, 27, 29, 33]. The remaining studies presented evidence for model sustainability by measuring outcomes once across a period of time [11, 22, 24, 28]. Several barriers and enablers associated with the sustainability of the models of care were described (Table 4).

Model	Study	Model description	Enablers	Barriers
MDT, educa- tion, and capacity ouilding for self-care	Bergmann et al. 2020 [20]	Evidence based guidelines: EMR order set up- dates, ED physicians and stakeholders' regular updates and nursing education.	Nil stated.	The providers were concerned about the early discharge of patients increasing the risk of readmission for low-risk DKA.
	Ellis et al. 2017 [<mark>30</mark>]	Individualised treatment plans, mandatory and optional skills training and relapse pre- vention delivered at home.	Nil stated.	Over one third did not com- plete the program as it was felt to be too long and home visits too frequent.
	Fiallo-Sharer et al. 2019 [8]	"Project ACE (Achieving control, Connecting resources, Empowering families.)" Tailored self- management resources.	Model acceptance was boosted by group-based sessions where a single facilitator managed several families together.	Program intensity was reduced by the low frequency of the sessions: every three to four months rather than weekly or every two weeks.
	Floyd et al. 2017 [21]	Shared medical appointments, groups ses- sions and peer support.	The model was well accepted by prac- titioners, patients and their families as all the components were perceived to work together in a group setting in a single appointment.	Nil reported.
	Goss et al. 2010 [11]	"Rural Australian Diabetes – RADICAL)" - MDT team meetings with education sessions.	Positive attitudes promoting hope and empowerment improved outcomes. The model was facilitated by a counsel- lor who helped develop a personalised and realistic approach to care. A small team allowed for provider consistency for consultations.	Reduced support in less populated regions for similarly skilled members of the MDT when team members on leave.
	Ilkowitz et al. 2016 [24]	Education, and support events and groups and increased access to clinics.	Improved patient to provider ratio, decreased wait times for appointments and increased number of sessions.	Nil stated.
	Katz et al. 2012 [<mark>26</mark>]	"Medical Home" - family centred, coordinated, comprehensive, compassionate, and cultur- ally effective.	A close collaboration between primary care doctors and the diabetes team.	Reduced health literacy for some families.
	Nansel et al. 2012 [33]	A program to facilitate problem-solving, com- munication skills and responsibility sharing via telephone calls and in-person contact. Program delivered by a specialised health advisor.	Integration into existing clinics.	Nil stated.
	Pascual et al. 2019 [<mark>27</mark>]	Shared medical appointments within a Latino program.	Multicultural team involvement in a culturally sensitive educational program.	Study attrition was reported for teenager group thought to be due to a lower drive to be educated compared to parents and their dependent children.
	Wan et al. 2022 [9]	Shared medical appointments among providers and peers	Team environment increased provider satisfaction	In person visits only (rather than online option) prevented some individuals from attend- ing clinics.
Shared care and decision making, and ntegration of care	Hatherly et al. 2011 [23]	Shared care among paediatric endocrinolo- gists from large metropolitan hospitals who travel to rural and regional sites to integrate care with a primary health care giver.	Nil reported.	A lack of access to 24-hour ambulatory care increased hospital admissions. There was a suggestion of inequity for those of lower socioeconomic status.
Outreach support	Franklin et al. 2014 [<mark>22</mark>] Joshi et al.	Allied health personnel support with 24/7 paediatric diabetes self-management. A 24 h/7 days telephone support with an in-	The team were reported to be dedi- cated and well-trained. The model involves a centralised and	Nil reported. Nil reported.
	2017 [22]	hospital or outreach team, and four visits from a metropolitan-based outreach team per year.	MDT approach.	nireported.

 Table 4
 Enablers and barriers of implementation of T1D models of care

Table 4 (continued)

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Model	Study	Model description	Enablers	Barriers
	Simm et al. 2014 [<mark>28</mark>]	Outreach rural services included a visiting medical specialist who attended routine med- ical appointments at three-month intervals and follow up by allied health teams (diabetes nurse educators).	protocols allowed for the successful	The number of clinic visits was higher for rural patients and the access to services for those living in more remote regions was difficult.
	Versloot et al. 2022 [29]	Individualised stepped care.	Having access to external funding to embed mental health support in the clinic.	There was a suggestion of ethnic diversity preventing access.

 $\mathsf{MDT} = \mathsf{Multidisciplinary\ team,\ \mathsf{EMR}} = \mathsf{electronic\ medical\ records,\ \mathsf{ED}} = \mathsf{emergency\ doctors}$

Discussion

Our evidence synthesis identified a variety of implemented innovative models of care for T1D, with an MDT approach most frequently reported. The key components of the models included MDTs, shared medical appointments, shared decision making, capacity building for self-care, outreach support and mentoring and integrated care enabled by technology such as telehealth consultations and interactive educational programs delivered online or by teleconference.

The literature shows enormous variety of outcome measures reported across studies, which makes it difficult to synthesise evidence of benefits across different models. For example, some reported benefits in terms of improved maintenance of glycaemic levels [10, 21, 27, 30], and increased insulin pump usage, while others reported no improvements in health outcomes [29, 31, 32].

Improved psychosocial outcomes, including quality of life were consistently reported for models that included shared care, decision making, and integrated care [23, 29, 32]. These benefits are consistent with previous studies on integrated care models for other conditions such as chronic kidney disease [36], cancer survival [37], and mental illness [38]. It remains uncertain, however, whether improvements, or lack of improvements, can be directly attributed to the innovative model of care or its components, because of the differing models in use, and the large variety of outcome tools and measures being used across studies. Versloot et al. (2022) [29] for example, applied both a general measure of quality of life and a diabetes-specific measure of quality of life, showing that effects were stronger when the diabetes-specific measure was used [29]. This suggests that a greater consistency and standardisation of diabetes specific outcome measures may be needed to improve the reliability of evidence synthesis across studies and settings in the future.

Outreach support had previously been reported to contribute to positive health behaviors for adults living with diabetes [39], and for the provision of social support and capacity for skill building among people living with chronic conditions in rural areas [40]. Consistent with our findings, these positive outcomes are enabled by technology – usually a combination of telephone and online support, in addition to face-to-face consultations as previously described in a model of care for children living with medical complexity [41].

Over half of the studies reported on the implementation determinants of the model of care, however none used implementation science frameworks or theories to guide their work. Commonly reported enablers of successful model of care implementation included communication and team cohesiveness [9, 11, 21, 22, 25–28, 33]. These findings are consistent with those reported in other studies of paediatric integrated care models [41], and by adult consumers and providers commenting on integrated care models [42]. A review conducted by Bradford et al. (2016) [43] reported that establishing clear, efficient processes to support teams to manage care is important for the successful implementation and sustainability of models of care [43].

The key barriers identified for patient engagement with T1D models of care included program intensity requiring frequent engagement or long educational sessions [30], difficulties in accessibility due to connectivity problems for online components [39], or additional travel and cost for families living in rural regions [28]. This suggests that groups developing new models of care may benefit with closer engagement and co-design with end-users before launching new models of care or programs for T1D. Limited availability of skilled staff when team members were on leave also limited access [11], suggesting the need for adequate human resource planning and staff training when implementing new models of care.

Short follow-up periods after implementation limited assessment of the evidence for models' sustainability. Nine (47%) of studies were evaluated at 12 months post implementations, and the follow-up period was not reported in two studies (11%). Only eight studies (42%) measured outcomes two years or longer after implementation, the longest follow-up period being six years [28]. For example, a rural MDT model of care that offered education and support showed significant benefits for glycaemic management and quality of life measures every 12 months for 3 years [11]. These findings are consistent with the known gaps in evidence for the sustainability of health programs [44], including for models of care for chronic health conditions [45]. Understanding the determinants of successful model of care implementation and sustainability should be a research priority to inform future T1D models of care development and co-design, as well as informing strategies for implementation and scaling up. For example, the Consolidated Framework for Implementation Science Research (CFIR) [46] and the Integrated Sustainability Framework (ISF) [45] provide comprehensive, flexible frameworks underpinned by robust theoretical grounding that lends itself to the study of implementation and sustainability of models of care while taking account of specific internal and external contexts [46].

Strengths and limitations of the current review

Applying the PRISMA guidelines, a rigorous multi-step team-based study selection process, a comprehensive search of four international academic databases and adhering to a registered protocol are all methodological strengths of this study. Most studies were from the USA and were conducted in large metropolitan settings which limits the generalisability of findings to other settings. Other limitations include selecting only articles written in English and from high income countries, and omitting grey literature may have impacted the comprehensiveness of results. Grey literature may be particularly important in this area as models of care may be reported in "in-house" service reports or evaluations that are not submitted for peer review and publication in journals. Additionally, our search criteria focused on implemented T1D models of care, and did not focus on other ways of driving improved health outcomes for T1D based on population registries or networks, for example the Swedish paediatric diabetes quality registry, SWEDIABKIDS [47] or the SWEET global network [48]. Future reviews should consider including grey literature and the impacts of registry data to drive improvements in health outcomes for T1D. Due to the wide heterogeneity of study methodologies, analysis methods, and outcome measures we were not able to pool data, and synthesis was challenging, limiting the generalisability of our findings. Other limitations included small sample sizes [21], limited scope [34], and single site models [10, 20–22, 24, 30, 31].

Conclusions

The evidence for the effectiveness, acceptability and sustainability of paediatric T1D models of care is currently limited and inconsistent, although a pattern is emerging. Across three broad categories of model components, that include an MDT approach, some studies reported benefits for children living with T1D, including improved glycaemic management, although other studies reported no benefit. Several psychosocial benefits such as improved quality of life and motivation were reported, especially for shared care models, but again some studies reported no benefit. Barriers to accessing these models by patients/families included program intensity, time commitment required, cost and location. Enhanced communication and team cohesiveness were reported as important enablers for effective care delivery. There is a need for greater consistency of outcome measures to enable more robust evidence synthesis across studies and contexts. We also identified a need to adopt longitudinal study designs with larger sample sizes and longer follow-up periods to better understand model of care sustainability. Future studies should be guided by robust theoretical frameworks to study implementation determinants and sustainability of T1D models of care.

Supplementary Information

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Supplementary Material 1: Example of the search strategy for Medline Ovid

Supplementary Material 2: Quality Appraisal of included studies using the Mixed Methods Appraisal Tool (MMAT)

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A review describing similar methodologies has been published in BMC Health Services Research [14].

Author contributions

YZ, ED, TJ and JB designed the research. YZ, AC, MS, IM, GD, NH, and RL conducted the research. AC and YZ analysed the data and drafted the manuscript. All authors read and approved the final manuscript.

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Data availability

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

Not required as a review article.

Consent for publication

Not applicable.

Competing interests

The authors declare no competing interests.

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