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TITLE

Evaluation of a nurse-led multi-component transition program for adolescents with congenital heart disease

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AUTHOR CONTRIBUTION

Michèle de Hosson (first author): Conceptualization; Data curation; Formal analysis; Investigation; Methodology; Writing - original draft; and Writing - review & editing.

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Eva Goossens & Julie De Backer (last authors): Conceptualization; Supervision; Writing - original draft; and Writing - review & editing.

All authors approved the final manuscript as submitted to be accountable for all aspects of work.

HIGHLIGHTS

- A nurse-led transition program with a transition coordinator as adolescents' guide
- Person-tailored education and transition skills training as program core components
- Transition program participants showed better knowledge scores than controls
- Participating adolescents reported better transition experiences than controls

KEY WORDS

Heart defects, congenital – Transitional Care – Transition Program - Transition to adult care
Chronic Disease

ABBREVIATIONS

AC	= Adult Cardiologist
CHD	= Congenital Heart Disease
CG	= Control Group
CI	= Confidence Interval

CP	= Communication-Paper
IG	= Intervention Group
IPTW	= Inverse Probability Treatment Weighting
ITP	= Individual Transition Plan
HR-QoL	= Health related - Quality of Life
LKQCHD	= Leuven Knowledge Questionnaire for Congenital Heart Disease
MD	= Mean Difference
OYOF	= On Your Own Feet
OYOF-TES (P)	= On Your Own Feet Transfer Experiences Scale (for Parents)
PC	= Pediatric Cardiologist
PedsQL	= Pediatric Quality of Life Inventory
SD	= Standard Deviation
SMD	= Standardized Mean Difference
TP	= Transition Program
TRAQ	= Transition Readiness Assessment Questionnaire

ABSTRACT

Objective

To evaluate the effectiveness of the transition program for adolescents with congenital heart disease (CHD) ‘Transition With a Heart’ (TWAH) on disease-related knowledge, quality of life (QoL), transition experiences, and gaps in follow-up.

Methods

A study with pre-posttest and control group (post-test) using consecutive sampling, including adolescents with moderate to severely complex CHD, without intellectual disability, aged ≥ 12 y, and ~~their~~ parents. After weighting, t-tests were performed. A multivariable regression analysis explored the outcomes’ determinants.

Results

In the intervention group, 28 adolescents and 25 parents were included, and 53 adolescents and 18 parents as controls. Adolescents’ knowledge significantly increased after completing TWAH (from 59.8% to 75.7%; $p < 0.01$). Their knowledge was positively correlated with TWAH ($\beta = +13.3$; $p < 0.01$). Adolescents’ transition experiences were also positively related to TWAH (general experience: $\beta = +5.5$; $p < 0.01$; transfer satisfaction: $\beta = +0.8$; $p < 0.01$). Adolescents’ QoL was mainly determined by CHD complexity and not by TWAH. No one showed gaps in follow-up. TWAH was not associated with parents’ transition experiences.

Conclusion

Implementing TWAH substantially improved adolescents’ disease-related knowledge and transition experiences.

Practice implications

The results regarding transition experiences need to be confirmed by further research. The TWAH design with the person-tailored educational program, skills training, and the transition coordinator can be used in settings with other chronic diseases.

1. INTRODUCTION

Congenital heart disease (CHD) is the most common birth defect, with a global birth prevalence of 8.2 per 1,000 newborns (1). Due to improved therapeutic options, up to 97% of children born with CHD survive into adulthood (2). However, most patients remain at risk for cardiac complications during adult life, such as arrhythmias, heart failure, and infective endocarditis. Across their lifespan, continuous, life-long, age-appropriate cardiac care is recommended to prevent, timely detect, and treat any health- or psychosocial-related problems. The frequency of the cardiac follow-up depends on the complexity of the CHD. For CHDs of severe complexity, such as all forms of cyanotic CHDs, at least yearly cardiac follow-up is recommended, whereas, for moderately complex CHDs, 1-2 years is reasonable (3, 4). Life-long, age-appropriate cardiac care requires adequate transition of care, which is *'the process by which adolescents and young adults with chronic childhood illnesses are prepared to take charge of their lives and their health in adulthood'* (5). Transfer of care is an integral part of this transition and is the event through which adolescents with chronic conditions *'move their care from pediatric to an adult healthcare environment'* (5).

Worldwide, 26% of CHD patients are lost to follow-up (6), demonstrating that continuity and transition of care are challenging (7-9). To ensure lifelong medical follow-up and well-being, adolescents and their parents require thorough preparation and care coordination during the transitional phase (10). Therefore, transition programs (TPs) are recommended (4, 6, 10-12), despite scarce scientific evidence of their effectiveness (13). Only a few studies evaluated TPs for CHD (14-21) and partially met the current consensus to implement a multi-component TP, including tailored information provision and self-management skills training (12). Most of these adolescents were transferred but rarely properly transitioned (13). Only the London (22) and the

STEPSTONES TP (23) for CHD adolescents met that current consensus for CHD adolescents and showed promising results on knowledge and empowerment.

To bridge the still-existing evidence gaps, our Pediatric Cardiology and Adult Congenital Heart Disease departments implemented and evaluated a multi-component TP named ‘Transition With a Heart’ (TWAH). This study aimed to assess TWAH’s effectiveness on disease-related knowledge as the primary outcome (from here on, called ‘knowledge’), health-related quality of life (HR-QoL), transition experiences, and gaps in follow-up.

2. METHODS

2.1 Study design

An evaluation study with a pre/post-test design for the intervention group (IG) and a control group (CG) (post-test only) was performed to examine TWAH’s effectiveness on knowledge (primary outcome) and secondary outcomes, and to explore their determinants. The start of TWAH was indicated as the baseline. T1 was the TP’s completion for the IG and the second outpatient visit in adult care for the CG. The first outpatient visit after T1 was indicated as T2 (see Figure 1).

2.2 Intervention and setting

TWAH was based on the Dutch research and innovation program ‘On your own feet’ (OYOF) (24) and was implemented in May 2015. TWAH is dedicated to adolescents with CHDs of moderate and severe complexity (3) (from here on, called ‘adolescents’).

Firstly, TWAH aims to provide adolescent-centered care and communication by initiating independent visits and using a communication-paper. The communication-paper is a practical

instrument based on the OYOF version (25) and for CHD specified by consulting literature (26, 27) and expert opinion. Adolescents can indicate what and with whom they want to discuss the particular topics (See Figure 2). The transition coordinator sends this digital form one week before each outpatient visit.

Secondly, TWAH aims to increase adolescents' knowledge by providing structured and tailored information and improving their transitional skills through an individualized transition plan, detecting transitional skills needing attention. The individual transition plan is also based on the OYOF version (25) and for CHD specified (See Figure 3). Furthermore, several organizational components are integrated (See Figure 4): (a) a transition coordinator, (b) a transition flowchart, and (c) a joint transfer outpatient visit. The joint transfer outpatient visit is the first outpatient visit in adult care, attended by the trusted pediatric cardiologist, the new adult cardiologist, and transition coordinator. This initiative already existed and was integrated into TWAH, encouraging a warm transfer. More details about TWAH are published in the TWAH methods paper (28).

2.3 Sampling

Eligibility criteria to participate in this study were: (a) age \geq 12y, (b) absence of intellectual disability, (c) CHD of moderate to severe complexity conform Baumgartner and colleagues (3), and (d) sufficient proficiency in Dutch.

Adolescents were assigned to the IG if their pediatric cardiologist participated in TWAH (n=3 pediatric cardiologists). Consecutive sampling was applied for composing the IG. From May 2015 to July 2019, adolescents were selected and invited by their pediatric cardiologist to participate in TWAH and were subsequently included in the IG. Adolescents cared for by a non-participating pediatric cardiologist (practical reason: reduced employment and approaching

retirement) and adolescents who had already reached the transfer age (+/-16y) when TWAH started were assigned to the CG. Therefore, convenience sampling was applied by the transition coordinator from May 2015 to February 2020. She selected and invited the adolescents and parents to participate in the study as controls. CG adolescents and parents received standard care. Standard care during the study period included only a transfer to adult care via the joint transfer outpatient visit. So, additional transitional guidance from the transition coordinator, including the provision of disease-related information and transitional skills training, was not offered to the adolescents and parents of the CG. The parent (both IG and CG) attending the T1 visit and most visits in the transitional phase was invited to participate (post-test only – from December 2017) (See Figure 5).

2.4 Data collection and outcome measures

Demographic and clinical data were collected from the medical records. The other data were collected through paper-based surveys (See Figure 1). The questionnaires were voluntary and independently completed by adolescents and parents, under the supervision of the transition coordinator, avoiding interaction and consulting external sources.

2.4.1 Disease-related knowledge

The primary outcome was the adolescents' level of knowledge, since establishing a healthy lifestyle requires a good understanding of the CHD, treatment, and lifestyle. Better disease-related knowledge empowers adolescents and is associated with lower risk behaviors and better transitional skills (29, 30) and is assessed using the Leuven Knowledge Questionnaire for Congenital Heart Disease (LKQCHD), a cross-cultural validated questionnaire (31-33), focusing on a spectrum of CHD-related domains with 25 and 27 items for boys and girls, respectively (34).

The total score was the percentage of correct answers to the total number of questions. The power analysis estimated that at least 34 adolescents (17 per group) were required to achieve 80% power for detecting an average difference of 12% for the knowledge score (35) at a significance level of $\alpha=0.01$. This percentage was the demonstrated effect of an educational intervention in CHD adolescents (35) and was assumed effective. The knowledge was adequate if more than 80% of the answers were correct and poor if less than 50% were correct (36).

2.4.2 Health-related quality of life

HR-QoL was assessed using the Pediatric Quality of Life Inventory (PedsQL) Dutch version 4.0 for adolescents, a valid measure for HR-QoL in pediatric chronic conditions (37). The PedsQL's generic and cardiac scales involved ten domains and 50 items (38). Adolescents indicated the degree of experienced problems with the items over the past month using a five-point Likert scale (0=never/4=almost always). The answers were computed to domain and total scores on a 0-100 scale. Higher scores reflected better HR-QoL.

2.4.3 Transition experiences

The validated questionnaires 'On Your Own Feet Transfer Experiences Scale' (OYOF-TES) (39) and OYOF-TES for Parents were used to assess transition experiences. Both questionnaires consisted of two subscales with several items that could be rated on a five-point Likert scale: collaboration between pediatric and adult care (range 11-55) and transfer preparation (range 7-35), resulting in the total score (range 18-90). Transfer satisfaction and confidence in the pediatric and adult cardiologist were indicated on visual analog scales (range 0-10). Higher scores reflected better experiences (39).

2.4.4 Gaps in follow-up

If the outpatient visit after T1 (=T2) exceeded the guideline-recommended time interval between two visits, depending on the CHD complexity(3), a gap in follow-up was recorded (40).

2.5 Statistical analysis

Descriptive statistics were used to describe adolescents' characteristics and gaps in follow-up. Continuous data were presented as means with standard deviations, and categorical data as absolute numbers with proportions. A paired t-test was performed as pre-posttest within the IG.

Given that there was no randomization and the IG and CG were not completely similar in baseline characteristics, inverse probability treatment weighting (IPTW) was used, creating a balance between relevant covariates that could confound the results. Weights were calculated by inverting propensity scores, estimated in a regression analysis with whether or not participating in TWAH as the dependent variable (41). More details of IPTW are illustrated as supplementary material.

After weighting, comparisons were made between the IG and CG with independent t-tests. A multivariable regression analysis was conducted to explore if TWAH independently determined the set of outcomes. Based on previous research (35) and expert opinion, the following covariates were included: group allocation (intervention or control), age, CHD complexity (3), sex, educational level, and number of cardiac surgeries. No violations were found when testing the assumptions of normality, linearity of residuals, and multicollinearity applicable to the performance of multivariable regression. A double robust estimation could be applied by including the propensity scores (41, 42).

All data were analyzed using SPSS 27.0 (SPSS Inc., Chicago, IL) with two-sided tests. The significance level was set at $\alpha < 0.01$, compensating for multiple testing given the large number of outcomes.

2.6 Ethical aspects

Written informed consent was obtained from all participants. Giving informed consent was not a prerequisite for TWAH participation but provided the option for study participation. The local ethical committee approved the study protocol (B670201525795), complying with the principles outlined by the Declaration of Helsinki (43).

3. RESULTS

3.1 Sample characteristics

In total, 33 adolescents were eligible to participate in the IG. Five of them did not start with TWAH for several reasons: request for faster transfer ($n=3$), heart transplantation ($n=1$), and discontinuity of care ($n=1$). The 28 remaining adolescents started with TWAH and were recruited as IG. In total, 53 adolescents were recruited in the CG. Of the 28 IG adolescents, 26 parents (maximum one per adolescent) were eligible. The other two did not attend the T1 visit and could not evaluate the entire TP. From December 2017, 21 parents could be invited as CG. Finally, one IG parent and three in the CG were unwilling to participate (lack of time), resulting in 25 IG and 18 CG parents.

The adolescents' characteristics are shown in Table 1.

3.2 Pre-post transition comparisons

The knowledge significantly increased from baseline to post-transition (from 59.8% to 75.7%; $p<0.01$) in IG adolescents (unweighted sample). Moreover, the proportion of adolescents with adequate knowledge significantly increased from 3.6% to 57.1% ($p<0.01$). Regarding HR-QoL, the subscale representing school functioning increased significantly from 76.3/100 to 83.0/100 ($p<0.01$) (see Table 2).

3.3 Comparisons between the intervention and control group after transition

After weighting, IG adolescents had significantly better knowledge post-transition than controls (75.9% versus 62.1%; $p<0.01$). Significant more IG adolescents showed adequate knowledge (=score>80%) than controls (54.3% versus 10.1%; $p<0.01$), and significantly fewer IG adolescents showed poor knowledge (=≤50%) than controls (5.0% versus 19.0%; $p<0.01$) (see Table 3).

IG adolescents reported significantly better HR-QoL than controls among the subscales treatment anxiety (respectively on average 93.9/100 versus 85.3/100; $p<0.01$), school functioning (82.8/100 versus 76.1/100; $p<0.01$), and psychosocial health (85.0/100 versus 78.2/100; $p<0.01$).

Regarding transition experiences, IG adolescents indicated significantly better general transition experience (78.5/90 versus 73.8/90; $p<0.01$) and better experienced collaboration between pediatric and adult care than controls (48.2/55 versus 45.0/55; $p<0.01$). IG adolescents were significantly more satisfied with their transfer than controls (9.0/10 versus 8.2/10; $p<0.01$). Adolescents and parents in both groups expressed great confidence in pediatric and adult cardiologists. Confidence in the adult cardiologist was significantly greater in IG adolescents than in controls (9.1/10 versus 8.7/10; $p<0.01$).

All participating adolescents attended the visit after T1 (=T2) within the recommended time interval. Hence, there was no short-term gap in follow-up.

3.4 Determinants of knowledge

The multivariable linear regression analysis indicated that knowledge was primarily influenced by TWAH participation. After weighting and correction for all selected covariates, TWAH participation was significantly associated with better knowledge ($\beta=+13.3\%$; 99CI [7.9; 18.8]; $p<0.01$). Furthermore, knowledge was associated with CHD complexity when adjusting for the other covariates. Adolescents with severely complex CHD had significantly better knowledge than those with moderately complex CHD ($\beta=+9.1\%$; 99CI [2.0; 16.2]; $p<0.01$).

3.5 Determinants of the secondary outcomes

Table 4 presents the regression analysis of the secondary outcomes and indicates that HR-QoL was, to a lesser extent, associated with TWAH participation. After weighting and correction for other covariates, IG adolescents demonstrated significantly better HR-QoL scores than controls for three of the 12 subscales: anxiety for future treatment, emotional functioning, and psychosocial health. HR-QoL was mainly associated with CHD complexity. It was found while holding other covariates constant, that adolescents with a severely complex CHD had lower HR-QoL scores than adolescents with moderately complex CHD regarding eight subscales (i.e., heart problems and treatment, medical treatment, cognitive problems, communication, physical functioning, social functioning, psychosocial health, and general HR-QoL).

Adolescents' transition experiences were mainly related to TWAH participation. After adjusting for other covariates, TWAH participation was positively associated with four out of six aspects: general transition experience, alignment and collaboration, preparation, and transfer satisfaction. Higher disease complexity significantly correlated with worse general transition experience, worse experienced preparation, and less confidence in the pediatric cardiologist.

Amongst parents, TWAH participation was not significantly associated with their transition experiences but mainly related to their child's CHD complexity. Parents of children with severely complex CHD had significantly worse transition experiences. The confidence in pediatric and adult cardiologists was unrelated to TWAH participation or other covariates in adolescents and parents.

4. DISCUSSION AND CONCLUSION

This study evaluated the multi-component TP 'TWAH', including tailored information provision, transitional skills training, guidance by a transition coordinator, joint transfer outpatient visits, and continuation of the TP in adult care, specially designed for adolescents with CHDs of moderate to severe complexity and their parents. Significant improvements in adolescents' knowledge and better transition experiences were observed after completing TWAH. We noted a slightly positive influence on HR-QoL. When interpreting these study results, some elements need to be considered.

4.1 Discussion

Disease-related knowledge

TWAH aimed to improve adolescents' knowledge by providing person-tailored information. After the first thorough information session, topics on adolescents' request were discussed. Moreover, autonomous conversation with healthcare providers was stimulated. This person-tailored method

aligns with previous recommendations (12, 44, 45). The TWAH program appeared to be effective in improving adolescents' knowledge. In previous studies, young people with CHD also demonstrated higher levels of knowledge after completing educational interventions (16, 21-23, 35, 46). However, TWAH resulted in markedly better knowledge scores (76%) than an one-time education intervention in young adults with CHD (57%) (35). In addition, the proportion of adolescents in our study with sufficient knowledge was significantly higher in the IG than the CG (54.3% versus 10.1%). Furthermore, significantly fewer IG adolescents showed poor knowledge ($\leq 50\%$) compared to controls (5.0% versus 19.0%; $p < 0.01$). These positive results were perhaps caused by the tailored TWAH approach with multiple information moments and discussing topics on the adolescent's demand.

Health-related Quality of Life

This study showed that HR-QoL was mainly determined by CHD complexity, with significantly lower scores in adolescents with severely complex CHDs. The limited contribution of transitional interventions on HR-QoL has been reported previously (47). Young CHD patients have good HR-QoL and sometimes even better than healthy peers. These results suggest they cope well with the disease burden and probably explain why a TP has little to add (48). Although the ultimate goal of transitional care is improving HR-QoL, the use of HR-QoL as an outcome measure in transitional research might be debatable, given the limited room for improvement.

Adolescents' and parents' transition experiences

In the current study, adolescents and parents reported good transition experiences. Our IG adolescents scored slightly higher than adolescents suffering from rheumatic diseases (49) or cystic

fibrosis (50) after completing comparable TPs. An average score of 78.5/90 was observed in our study versus 74.5/90 (49) and 71.7/90 (50) in the other samples. Moreover, our study showed that adolescents' general transition experiences were positively related to TWAH participation ($\beta=+5.5$; $p<0.01$). This positive association may result from the multi-component TWAH approach. TWAH differentiates with this approach from previous studies in CHD adolescents merely evaluating a single-component or one-time transition intervention (19-21, 51).

The experiences of IG parents were relatively good but could not be associated with TWAH participation since parents' experiences in the CG were comparable good. These good parental experiences are possibly related to the joint outpatient visit, an organizational intervention previously experienced very positively for building a new treatment relationship (52). Parents of children with severely complex CHD in our study expressed significantly worse transition experiences. The more intensive cardiac follow-up, inherent to more complex CHDs (3), may cause more parental distress and worse experiences.

Regarding transition experiences, no comparison with other studies on CHD adolescents and their parents could be made, revealing room for further transition research. We are currently performing qualitative research to get an in-depth understanding of participants' experiences.

Self-management skills and transition readiness

Next to the educational intervention, TWAH provided transitional skills training to improve the adolescents' autonomy by using a CHD-specific individual transition plan and formulating personalized goals. The adolescent's transition readiness assessment consisted of a discussion with the adolescent, the parents, the cardiologist, and the TC based on the adolescent's levels of maturity, disease-related knowledge, and transitional skills (based on the individual transition plan). An assessment tool like the TRAQ (Transition Readiness Assessment Questionnaire) (53)

was not used since that instrument is not disease-specific and thus falls short in discussing CHD-specific issues. In addition, the TRAQ focuses on medical self-management and, to a lesser extent, on the wider perspective of social participation (25). We could not quantitatively test the evolution of the level of transitional skills since the individual transition plan is a clinical tool but not a validated instrument for research. Nevertheless, we have the impression from clinical practice that the adolescents' autonomy improves after completing TWAH. Interestingly, a previous study found a positive link between disease-relating knowledge and self-management. Adolescents with better disease-related knowledge were more likely to communicate directly with healthcare providers than those with less knowledge (54). Likewise, the more independent adolescents with higher TRAQ scores had better disease-related knowledge than the less transition-ready adolescents (30).

Gaps in follow-up and organizational aspects

Remarkably, in the current study, none of the IG and CG adolescents showed gaps in follow-up. Although the study period was rather short, this is still interesting, especially compared to the previously reported 26% of CHD patients worldwide developing gaps in follow-up (6). The absence of these gaps in this study could be attributed to proactively making appointments, sending out reminders by mail and text message prior to the visits, the shared medical record used in the pediatric and adult clinic, both services being located on the same campus, and the relatively short traveling distance for patients (55). Additionally, all our adolescents were transferred via a joint transfer outpatient visit. This organizational intervention properly navigates patients to adult care and tends to ease transfer (52, 56).

Another organizational characteristic of TWAH is the continuation of the transition in adult care, meeting the recommendation of Moons and colleagues (12) to not stop with the transition of care after the transfer. As a result, adolescents get more time to adjust to the new healthcare environment. With this, TWAH differs from other recent multi-component transition programs for adolescents with CHD that do not continue the transition program after the transfer to adult care (22, 23).

Lastly, the TWAH transition coordinator is employed in adult care. The adolescents and parents can gradually get to know the new healthcare provider in the trusted pediatric healthcare environment. Furthermore, the TWAH transition coordinator remains the point of contact in adult care. Subsequently, another farewell from a trusted healthcare provider is unnecessary and potentially reassuring for adolescents and parents since they previously expressed hard feelings (52, 57, 58). From that perspective, appointing a transition coordinator from adult care is an advantage and a strength of TWAH.

Strengths and limitations

The study strengths are the pre-post-test design and the inclusion of a control group with comparable characteristics as the IG. Randomization was not applied for logistic and ethical reasons. The selection of the CG was partially based on the treating pediatric cardiologist. This could have introduced selection bias, although no major differences were found between the IG and CG characteristics. Nevertheless, IPTW was applied to equalize the sample characteristics, approaching the properties of a randomized controlled trial (41, 42, 59). Unlike the initial sample, all standardized mean differences between the IG and CG in the weighted sample were below 0.1,

the threshold indicating a more balanced distribution of relevant covariates (See supplementary material) (59). We must be aware that the IG and CG may have differed on other variables not included in the IPTW.

It needs to be noted that the CG could not participate in the pre-test, so insights into their initial levels of knowledge and HR-QoL are lacking. Another study limitation is the lack of data on parents' characteristics. Due to the unavailability of these data, the parents' transition experiences were linked to their children's characteristics in the regression analysis. The clinical implications of the results regarding the transition experiences need to be confirmed since several answer scales and the differences were relatively small. Additionally, the regression analysis revealed that TWAH was the primary determinant of the adolescents' positive transition experiences. We are conducting a qualitative study, providing more a more comprehensive understanding of the clinical implications of these results.

Adolescents with a CHD of simple complexity were not included in TWAH since the intensive TWAH trajectory cannot be performed during the less frequent follow-up this patient group needs. Nevertheless, these adolescents are transferred via a joint transfer outpatient visit, and they, nowadays, receive information during the post-transfer visit to emphasize life-long cardiac follow-up and prevent gaps in follow-up.

This study is a single-center study with a relatively small sample size. We included the vast majority of the population in TWAH (28 of the 32 adolescents expected to be eligible in the four-year-long inclusion period). These aspects strengthen the generalizability. An intervention bias cannot be ruled out due to the long inclusion and intervention period. However, no methodological, personnel or organizational changes occurred during this period, reducing that risk. Our center has specific organizational characteristics, with separated pediatric and adult outpatient clinics on the

same campus and a collaborative culture between both teams for several decades. Our results may not reflect the effectiveness of TWAH in hospitals with different organizational configurations.

Unfortunately, we could not cover all transition-related outcomes in this study, like self-management, clinical outcomes, and cost-effectiveness. The latter is underinvestigated but necessary to convince all stakeholders of the importance of TPs.

4.2 Conclusion

Implementing the multi-component and person-tailored TP TWAH substantially improved adolescents' disease-related knowledge and transition experiences and may lead to a more successful transition. This study expands on the current scientific knowledge regarding the effectiveness of TPs for adolescents with CHD, which is necessary to inspire healthcare professionals and convince stakeholders in transitional care for adolescents with chronic conditions.

4.3 Practice implications

The core components of TWAH are providing a person-tailored education program and transition skills training, starting in pediatric care and continuing in adult care. In addition, the following organizational interventions are included: (1) a transition coordinator as leader and guide, (2) a general transition flowchart, and (3) a joint transfer outpatient visit, attended by the pediatric and adult cardiologist, and the transition coordinator. The TWAH design can be used as a basis in settings with adolescents suffering from chronic conditions. Considering the specific disease-

related needs and the local organizational configurations with the available (human) resources is necessary.

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SUPPLEMENTAL MATERIAL

The step-by-step plan below was followed as suggested by Moons et al. (41) and Joffe et al. (60) to apply a thorough inverse Probability Treatment Weighting (IPTW).

Step 1: The primary effect of interest

The average treatment effect in the population (ATE) was chosen as the primary effect of interest since we wanted to investigate the effect of the transition program TWAH in the adult CHD population. In ATE, both the IG and CG are weighted (41).

Step 2: Selection of clinical and demographic characteristics included in the propensity score calculation

Age

Gender

Boy

Girl

CHD complexity*

Moderate

Severe

Medication intake

Yes

No

Number of cardiac surgeries

Devices

Yes

No

Number of comorbidities

College level

Vocational

Technical

** Classified according to Baumgartner et al.³*

Step 3: Estimation of propensity scores

- 1) Modeling binary logistic regression analysis
 - a. Dependent variables: treatment assignment (intervention or control group)
 - b. Independent variables: selected clinical and demographic characteristics (step 2)
- 2) Calculation of probabilities of treatment assignment (p)

Step 4: Calculation of weights (w)

The weights were calculated by inverting the propensity scores (p) using the formulas below (42):

$$W_{intervention\ group} = \frac{1}{p} \quad W_{control\ group} = \frac{1}{1-p}$$

Step 5: Analyzing standardized mean differences in the unweighted and weighted sample

	Unweighted sample			Weighted sample		
	Intervention	Control	SMD	Intervention	Control	SMD
<i>Patient characteristics</i>						
<i>n</i>	28	53		81	81	
Age (in years)						
<i>T0</i>	14.5 ±1.4					
<i>T1</i>	17.7 ±1.3	17.9 ±1.3	-0.173	17.9 ±1.3	17.9 ±1.3	0.040
Female	11 (39%)	19 (36%)	0.072	32 (40%)	31 (38%)	0.025
Vocational high school	15 (54%)	27 (51%)	0.054	40 (49%)	40 (50%)	-0.012
<i>Medical characteristics</i>						
Disease-complexity*						
<i>Moderate</i>	16 (57%)	39 (74%)		54 (67%)	55 (67%)	
<i>Severe</i>	12 (43%)	14 (26%)	0.352	27 (33%)	26 (32%)	0.025
No. of cardiac surgeries	1.8±1.1	1.3±0.9	0.468	1.5±1.0	1.5±1.0	0.006
Medication **	13 (46%)	16 (30%)	0.338	28 (35%)	28 (35%)	-0.008
Cardiac implantable electronic device	1 (4%)	1 (6%)	-0.100	4 (5%)	4 (5%)	0.000
No. of comorbidities	0.4±0.6	0.3±0.6	0.212	0.3±0.5	0.3±0.5	0.088

Continuous variables are represented as mean ± standard deviation

Dichotomous variables are represented as n(%)

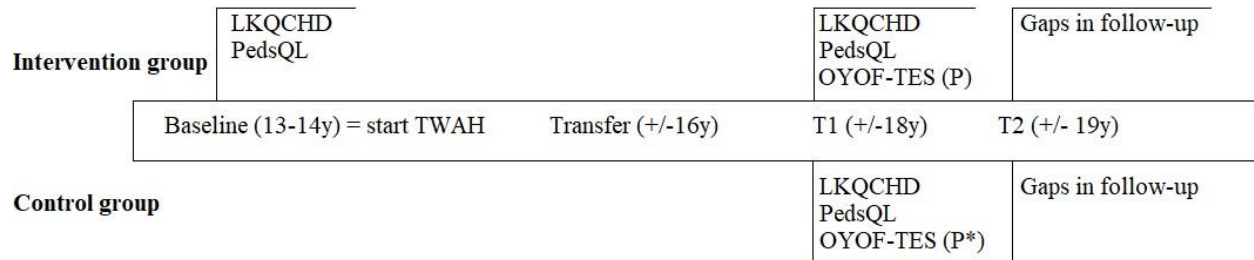
SMD = standardized mean difference between intervention and control group

** Classified according to Baumgartner et al. (3) - ** all (non)cardiac medication noted in file*

Step 6: Estimation of the effect of TWAH by comparing the intervention and control group in the weighted sample

LIST OF FIGURES AND LEGENDS

Figure 1: Participant data collection timeline



Legend

*Parents recruited if transferred between December 2017-February 2020

T1 = program completion (intervention)/2nd visit in adult care (control)

T2 = visit following T1

LKQCHD = Leuven Knowledge Questionnaire for Congenital Heart Disease

PedsQL = Pediatric Quality of Life Inventory

OYOF-TES (P) = On Your Own Feet Transfer Experiences Scale (Parents)

Figure 2: The TWAH communication-paper

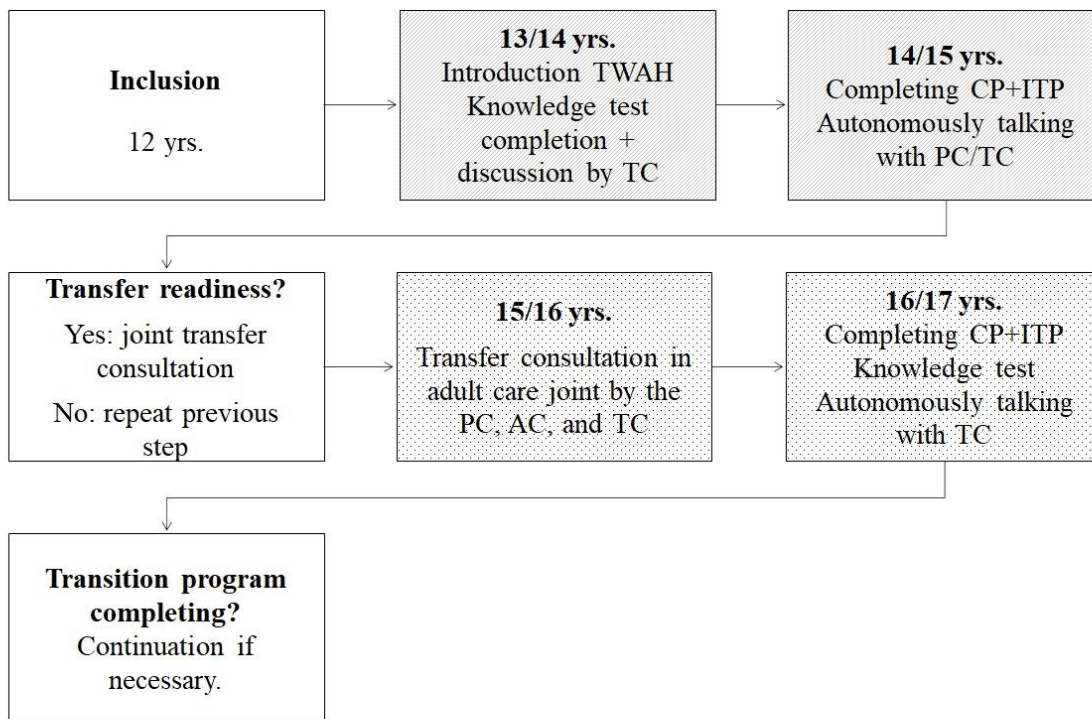
I want to talk about (yes/no)			
My heart disease	Operations	Medication	Care (skin, teeth...)
Education	Leisure and sports	Relationships and sex	Something else....

I want to talk about this with:			
My cardiologist	A psychologist	My nurse	Somebody else....

Figure 3: The TWAH individual transition plan

My heart disease
I can recognize the warning signs of my heart I know how to handle an acute medical situation
The future care
I can ask questions during the consultation I see the doctor unaccompanied during (a part of) the consultation I keep track of my next appointment and can make an appointment myself
Medication
I ask for a prescription when necessary and go to the pharmacy myself I make sure I take my medication myself
Social
I know what to do when I'm away from home for a longer time I talk to my friends about my heart disease
Answer scale
Not at all - Just a little/sometimes - Yes, about - Absolutely yes <i>Optional: I want to work on this</i>


Figure 4: Flowchart of Transition With A Heart




Legend

TWAH = Transition with a Heart - TC = Transition Coordinator - PC = Pediatric Cardiologist

AC = Adult Cardiologist - CP = Communication-Paper - ITP = Individual Transition Plan

 = process step

 = pediatric care


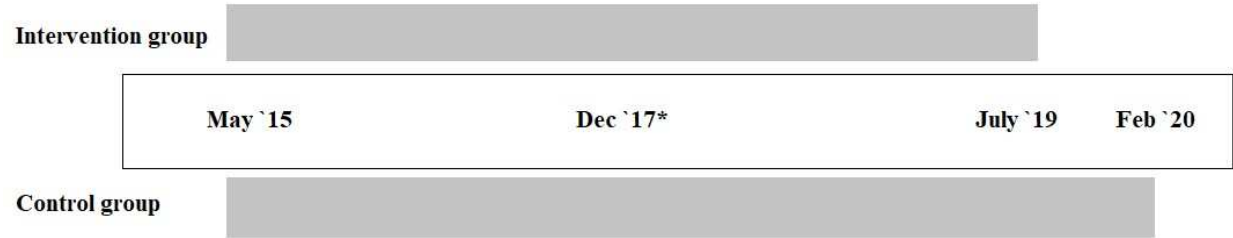

 = adult care

Figure 5: Inclusion timeline



Legend

*Parents recruited as control group if transferred from December 2017

 = inclusion period

TABLES AND LEGENDS

Table 1: Sample characteristics (unweighted sample)

Table 1: Sample characteristics (unweighted sample)

	Intervention	Control
<i>Patient characteristics</i>		
<i>n</i>	28	53
Age (in years)		
<i>T0</i>	14.5±1.4	
<i>T1</i>	17.7±1.3	17.9 ±1.3
Female	11 (39%)	19 (36%)
Vocational high school	15 (54%)	27 (51%)
Number transition outpatient visits	4.4±0.9	
<i>Medical characteristics</i>		
Disease-complexity*		
<i>Moderate</i>	16 (57%)	39 (74%)
<i>Severe</i>	12 (43%)	14 (26%)
No. of cardiac surgeries	1.8±1.1	1.3±0.9
Medication **	13 (46%)	16 (30%)
Cardiac implantable electronic device	1 (4%)	1 (6%)
No. of comorbidities	0.4±0.6	0.3±0.6

Legend

Continuous variables are represented as mean ± standard deviation

Dichotomous variables are represented as n(%)

* Classified according to Baumgartner et al.³ - ** all (non)cardiac medication noted in file

Table 2: Pre-posttest of TWAH participants from baseline to program completion (T1)

	Baseline	T1	Paired differences			
	Mean (SD)	Mean (SD)	MD (SD)	99% CI	t	p-value ^a
Disease-related knowledge^c						
Total score	59.8 (13.0)	75.7 (12.6)	15.9 (10.5)		8.05	<0.01
Adequate knowledge score* n(%)	1 (3.6)	16 (57.1)				<0.01 ^b
Quality of Life^d						
<i>Cardiac module</i>						
Heart Problems and treatment	75.8 (18.5)	75.6 (21.5)	-0.1 (16.4)		-0.04	0.97
Treatment II	90.9 (7.0)	90.0 (9.7)	-0.9 (6.6)		-0.45	0.66
Perceived Physical Appearance	87.3 (14.5)	85.8 (15.1)	-1.5 (18.1)		-0.44	0.66
Treatment Anxiety	91.2 (13.2)	92.8 (9.3)	1.6 (14.8)		0.57	0.57
Cognitive Problems	77.1 (22.5)	79.6 (22.2)	2.5 (25.9)		0.49	0.63
Communication	81.4 (13.2)	77.9 (23.3)	-3.5 (22.5)		-0.80	0.43
<i>General module</i>						
Physical Functioning	82.3 (14.1)	85.4 (13.6)	3.1 (14.4)		1.12	0.27
Emotional Functioning	82.3 (12.5)	82.5 (15.0)	0.2 (12.6)		0.09	0.93
Social Functioning	86.3 (16.4)	88.4 (14.7)	2.1 (12.7)		0.90	0.38
School Functioning	76.3 (16.8)	83.0 (13.8)	6.8 (12.9)		2.79	<0.01
Psychosocial Health Summary score	81.5 (12.5)	84.6 (12.2)	3.1 (9.5)		1.74	0.09
Total Score	81.9 (12.0)	84.9 (11.6)	3.0 (9.9)		1.59	0.12

Legend

^a Paired t-test unless indicated otherwise – ^b McNemar test - ^c range= 0-100 - ^d range= 0-100

* if LKQCHD score >80%

Table 3: Comparison between adolescents and parents from the intervention and control group after transition

	Intervention	Control	
	Mean (SD)	Mean (SD)	p-value ^a
Disease-related knowledge^c			
Total score*	75.9 (12.3)	62.1 (12.5)	<0.01
Adequate knowledge score (>80%) n(%)	44 (54.3)	8 (10.1)	<0.01 ^b
Poor knowledge (<50%) n(%)	4 (5.0)	15 (19.0)	<0.01 ^b
Quality of Life^d			
<i>Cardiac module</i>			
Heart Problems and treatment	75.7 (21.3)	76.9 (18.7)	0.70
Treatment II	92.2 (9.1)	93.2 (7.4)	0.63
Perceived Physical Appearance	85.4 (14.7)	82.0 (22.8)	0.29
Treatment Anxiety	93.9 (8.4)	85.3 (20.6)	<0.01
Cognitive Problems	80.1 (21.8)	74.2 (17.7)	0.08
Communication	75.1 (25.0)	78.2 (25.3)	0.46
<i>General module</i>			
Physical Functioning	84.8 (13.6)	80.7 (20.9)	0.16
Emotional Functioning	82.8 (15.3)	75.7 (20.2)	0.02
Social Functioning	89.4 (13.6)	83.4 (19.5)	0.02
School Functioning	82.8 (13.2)	76.1 (16.7)	<0.01
Psychosocial Health Score	85.0 (11.7)	78.2 (17.0)	<0.01
Total Score	84.9 (11.1)	79.1 (17.1)	0.01
Transition experience^e			
Perceived alignment and collaboration ^f			
<i>Adolescents</i>	48.2 (5.2)	45.0 (4.6)	<0.01
<i>Parents</i>	47.5 (5.3)	47.0 (6.3)	0.70
Experienced preparation ^g			
<i>Adolescents</i>	30.6 (4.5)	28.9 (3.9)	0.02
<i>Parents</i>	30.0 (4.5)	29.7 (4.8)	0.79
Transition experience (OYOF-TES total score) ^h			
<i>Adolescents</i>	78.5 (9.4)	73.8 (7.8)	<0.01
<i>Parents</i>	77.5 (8.8)	76.8 (11.1)	0.75
Transfer satisfaction ⁱ			
<i>Adolescents</i>	9.0 (0.7)	8.2 (1.2)	<0.01
<i>Parents</i>	8.6 (1.0)	8.8 (1.1)	0.38
Confidence in the pediatric cardiologist ⁱ			
<i>Adolescents</i>	9.6 (0.7)	9.3 (0.8)	0.02
<i>Parents</i>	9.5 (0.8)	9.5 (0.5)	0.63
Confidence in the adult cardiologist ⁱ			
<i>Adolescents</i>	9.1 (0.9)	8.7 (1.1)	<0.01
<i>Parents</i>	8.8 (1.3)	8.9 (1.1)	0.70

Gaps in follow-up (n)

0

0

Legend

^a Independent t-test unless indicated otherwise

^b Fisher's exact test

^c Leuven Knowledge Questionnaire for Congenital Heart Disease (LKQCHD) range= 0-100

^d Pediatric Quality of Life Inventory (PedsQL) range= 0-100

'On Your Own Feet Transfer Experiences Scale' (OYOF-TES) - ^f Range= 11-55 - ^g Range=7-35 - ^h Range=18-90 - ⁱ Range=0-10

SD= standard deviation

Table 4 (part 1): Double robust multivariable regression analysis on the secondary outcome variables*

	Group (project)			Age			CHD complexity (severe)**		
	β	[99%CI]	<i>p</i>	β	[99%CI]	<i>p</i>	β	[99%CI]	<i>p</i>
Quality of Life^a									
<i>Cardiac module</i>									
Heart problems and treatment	-0.6	[-9.5; 8.4]	0.87	-2.4	[-6.1; 1.3]	0.10	-22.2	[-33.6; -10.8]	<0.01
Treatment II	0.3	[-4.0; 4.7]	0.85	-1.1	[-3.1; 0.8]	0.12	-11.1	[-15.8; -6.4]	<0.01
Perceived physical appearance	3.4	[-4.4; 11.3]	0.26	0.6	[-2.9; 4.0]	0.67	-5.0	[-14.9; 4.9]	0.19
Treatment anxiety	9.0	[3.4; 14.6]	<0.01	-1.0	[-3.5; 1.5]	0.30	-5.2	[-12.3; 1.9]	0.06
Cognitive problems	6.1	[-2.4; 14.6]	0.06	-1.4	[-5.2; 2.4]	0.34	-18.7	[-29.7; -7.7]	<0.01
Communication	0.4	[-10.6; 11.4]	0.92	-3.3	[-8.1; 1.6]	0.08	-19.2	[-33.5; -5.0]	<0.01
<i>General module</i>									
Physical functioning	4.2	[-2.9; 11.3]	0.12	-2.2	[-5.1; 0.7]	0.05	-16.3	[-25.5; -7.1]	<0.01
Emotional functioning	8.0	[0.9; 15.2]	<0.01	-4.4	[-7.4; -1.4]	<0.01	-3.9	[-13.0; 5.2]	0.27
Social functioning	5.7	[-0.7; 12.0]	0.02	2.0	[-0.6; 4.6]	0.05	-15.4	[23.5; -7.4]	<0.01
School functioning	6.6	[0.0; 13.1]	0.01	-0.1	[-2.8; 2.6]	0.93	-7.2	[-15.5; 1.2]	0.03
Psychosocial Health Score	6.9	[1.0; 12.7]	<0.01	-0.8	[-3.2; 1.6]	0.38	-8.8	[-16.2; -1.4]	<0.01
Total Score	5.8	[0.0; 11.6]	0.01	-1.3	[-3.7; 1.1]	0.15	-11.7	[-19.1; -4.3]	<0.01
Transition experience^b									
Perceived alignment and collaboration									
<i>Adolescents</i>	3.6	[1.4; 5.8]	<0.01	0.4	[-0.5; 1.3]	0.22	-2.2	[-5.0; 0.6]	0.05
<i>Parents</i>	1.8	[-2.0; 5.5]	0.23	0.8	[-0.6; 2.3]	0.12	-4.4	[-8.5; -0.4]	0.01
Experienced preparation									
<i>Adolescents</i>	2.1	[0.2; 4.0]	<0.01	0.8	[0.0; 1.6]	0.01	-3.6	[-6.1; -1.2]	<0.01
<i>Parents</i>	1.9	[-0.8; 4.7]	0.07	2.5	[1.4; 3.5]	<0.01	-6.6	[-9.4; -3.7]	<0.01
Transition experience (OYOF-TES total score)									
<i>Adolescents</i>	5.5	[1.5; 9.5]	<0.01	1.0	[-0.6; 2.7]	0.10	-6.1	[-11.3; -0.9]	<0.01
<i>Parents</i>	3.8	[-2.7; 10.2]	0.13	3.3	[0.9; 5.7]	<0.01	-11.0	[-17.7; -4.3]	<0.01
Transfer satisfaction									
<i>Adolescents</i>	0.8	[0.4; 1.3]	<0.01	-0.1	[-0.2; 0.1]	0.47	-0.3	[-0.8; 0.3]	0.21
<i>Parents</i>	-0.1	[-0.8; 0.6]	0.71	-0.1	[-0.4; 0.2]	0.30	-0.5	[-1.3; 0.3]	0.09
Confidence in the pediatric cardiologist									
<i>Adolescents</i>	0.3	[0.0; 0.6]	0.02	0.0	[-0.2; 0.1]	0.40	-0.7	[-1.1; -0.3]	<0.01
<i>Parents</i>	-0.2	[-0.7; 0.3]	0.39	-0.2	[-0.4; 0.0]	<0.01	-1.2	[-1.7; -0.6]	<0.01
Confidence in the adult cardiologist									
<i>Adolescents</i>	0.4	[0.0; 0.9]	0.01	-0.1	[-0.3; 0.1]	0.23	-0.5	[-1.1; 0.0]	0.01
<i>Parents</i>	0.2	[-0.8; 1.2]	0.59	0.5	[0.1; 0.9]	<0.01	-0.4	[-1.4; 0.7]	0.36

Table 4 (part 2): Double robust multivariable regression analysis on the secondary outcome variables*

Sex (boy) School level (vocational) No. Cardiac surgeries

	β	[99%CI]	<i>p</i>	β	[99%CI]	<i>p</i>	β	[99%CI]	<i>p</i>
Quality of Life^a									
<i>Cardiac module</i>									
Heart problems and treatment	7.3	[-2.5; 17.1]	0.05	3.3	[-5.9; 12.4]	0.36	-1.9	[-9.5; 5.7]	0.51
Treatment II	-5.4	[-10.6; -0.3]	<0.01	-3.0	[-8.0; 2.0]	0.11	1.3	[-1.5; 4.2]	0.22
Perceived physical appearance	12.9	[4.3; 21.5]	<0.01	7.2	[-1.4; 15.8]	0.03	-2.6	[-9.2; 4.0]	0.31
Treatment anxiety	4.4	[-1.8; 10.5]	0.07	9.2	[3.0; 15.3]	<0.01	1.2	[-3.5; 5.9]	0.52
Cognitive problems	-0.1	[-9.4; 9.3]	0.99	15.1	[5.7; 24.5]	<0.01	6.9	[-0.3; 14.2]	0.01
Communication	15.9	[3.8; 28.0]	<0.01	5.2	[-6.9; 17.4]	0.26	-9.8	[-19.2; -0.4]	<0.01
<i>General module</i>									
Physical functioning	10.1	[2.4; 17.8]	<0.01	6.5	[-0.7; 13.8]	0.02	-0.1	[-6.3; 6.0]	0.96
Emotional functioning	0.8	[-7.1; 8.6]	0.80	5.3	[-2.0; 12.6]	0.06	5.6	[-0.5; 11.7]	0.02
Social functioning	0.5	[-6.4; 7.4]	0.84	6.8	[0.3; 13.2]	<0.01	0.2	[-5.2; 5.6]	0.92
School functioning	-3.5	[-10.6; 3.7]	0.21	5.2	[-1.5; 11.8]	0.05	1.2	[-4.4; 6.8]	0.58
Psychosocial Health Score	-0.8	[-7.1; 5.6]	0.75	5.8	[-0.1; 11.8]	0.01	2.3	[-2.6; 7.3]	0.22
Total Score	3.2	[-3.1; 9.4]	0.19	6.0	[0.1; 11.8]	0.01	1.6	[-3.3; 6.6]	0.39
Transition experience^b									
Perceived alignment and collaboration									
<i>Adolescents</i>	0.3	[-2.1; 2.7]	0.75	1.8	[0.5; 4.0]	0.04	1.1	[-0.7; 3.0]	0.12
<i>Parents</i>	1.9	[-1.6; 5.4]	0.15	5.3	[2.3; 8.3]	<0.01	2.5	[-0.1; 5.1]	0.01
Experienced preparation									
<i>Adolescents</i>	1.1	[-0.9; 3.1]	0.16	0.5	[-1.3; 2.4]	0.46	0.4	[-1.3; 2.0]	0.55
<i>Parents</i>	3.8	[1.3; 6.3]	<0.01	2.8	[0.6; 4.9]	<0.01	-1.1	[-2.9; 0.7]	0.11
Transition experience (OYOF-TES total score)									
<i>Adolescents</i>	1.9	[-2.5; 6.2]	0.26	2.4	[-1.6; 6.4]	0.11	1.5	[-1.9; 5.0]	0.24
<i>Parents</i>	5.7	[-0.1; 11.5]	0.01	8.1	[3.1; 13.1]	<0.01	1.4	[-2.9; 5.7]	0.39
Transfer satisfaction									
<i>Adolescents</i>	0.3	[-0.2; 0.8]	0.12	-0.3	[-0.7; 0.2]	0.13	0.0	[-0.3; 0.4]	0.78
<i>Parents</i>	0.0	[-0.6; 0.7]	0.92	0.2	[-0.3; 0.8]	0.29	0.3	[-0.1; 0.8]	0.06
Confidence in the pediatric cardiologist									
<i>Adolescents</i>	-0.2	[-0.6; 0.1]	0.07	0.4	[0.0; 0.7]	<0.01	0.5	[0.2; 0.8]	<0.01
<i>Parents</i>	0.1	[-0.3; 0.6]	0.51	-0.4	[-0.8; 0.0]	0.01	0.4	[0.1; 0.8]	<0.01
Confidence in the adult cardiologist									
<i>Adolescents</i>	0.6	[0.1; 1.1]	<0.01	0.0	[-0.5; 0.4]	0.79	-0.2	[-0.6; 0.1]	0.10
<i>Parents</i>	0.5	[-0.4; 1.4]	0.12	1.1	[0.3; 1.9]	<0.01	0.1	[-0.6; 0.8]	0.74

Legend

* Propensity scores were included in the regression analysis but not shown as it has no clinical implication

- ** According to Baumgartner et al³

^a Pediatric Quality of Life Inventory (PedsQL)

^bOn Your Own Feet Transfer Experiences Scale' (OYOF-TES)

β =Regression coefficient - CI= 99% Confidence Interval - Bold= significant result