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A B S T R A C T

Objective: Transition education programs dedicated to adolescents and young adults with congenital heart disease (CHD) aim to facilitate transfer to adult cardiology and bring more autonomy to teenagers. This prospective controlled multicentre study analysed the factors influencing the participation in a transition education program.
Methods: CHD patients aged 13–25 y were offered to participate in the transition program. A multiple linear regression identified the explanatory factors for participation in the program.
Results: A total of 123 patients (mean age 19.6 ± 3.4 y) were included in the study, with 57 participants and 66 non-participants. Both groups showed similar socio-demographic and quality of life characteristics, low level of physical activity with muscular deconditioning and high exposure to risk behaviours (71% patients with ≥1 risk factor). Patients with complex CHD (OR = 4.1, P = 0.03), poor disease knowledge (OR = 0.3, P = 0.02), risk behaviours (body piercing, OR = 5.53, P = 0.01; alcohol, OR = 3.12, P = 0.06), and aged <20 y (OR = 0.29, P = 0.03), were more likely to join the program.
Conclusion: Many risk factors influencing the participation of adolescents and young adults with CHD in transition education programs are controllable.
Practice implication: Further randomized studies are necessary to evaluate the impact of transition education program on quality of life, successful transfer to adult centre and, ultimately, prognosis.

1. Introduction

Congenital heart diseases (CHD) are the leading cause of congenital diseases with an incidence of 0.8% at birth [1]. The evolution of surgical technics and anaesthesia strategies during the second part of the 20th century has dramatically improved the prognosis. Currently, more than 90% patients reach adulthood [2–4]. In the past decade, the adult population with a congenital heart disease (ACHD) has overpassed in numbers the paediatric population and each year, more than 200 patients on 100,000 live birth will integrate the adult care system [5]. Teenagers and young adults with a CHD may be reluctant to leave the paediatric environment, and frequently stop their follow-up soon after transfer to adult cardiology, being at risk to reintegrate the healthcare system when experiencing a complication [1,6].

The concept of “transition” has emerged as a response to this public health issue, aiming to provide structured patient therapeutic education, before transfer to adult care [7,8]. Indeed, structured transition education programs intend to bring more autonomy to teenagers, not only in the knowledge of their CHD, but also in every domain of their life: physical activity, psychological

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well-being, sexuality, graduate studies, professional carriers, risky behaviour, etc [9–11]. Several transition education programs are being experimented in European and Northern American countries, with heterogeneous practices [12]. However, while educational needs are now identified, the detailed structure and efficacy of such programs have scarcely been reported [13]. Clinical research in therapeutic education has been given recent interest among children and adult patients with CHD [14,15]. However, more research is needed to evaluate the impact of transition education programs in CHD, considering the existing heterogeneity in terms of disease severity, age (e.g. paediatric and adult patients) and type of education [16]. Moreover, such research should involve an evaluation of patient related outcomes, such as health-related quality of life assessment, throughout the transition education program. [15,17]

Based on our expertise on patient education in children with CHD [14,15], and following the current guidelines [7,8], we developed in 2015 a non-selective structured education transition program, dedicated to teenagers and young adults followed in all 3 tertiary care CHD centres from the Occitanie region, France. Transition education programs might be time consuming for healthcare providers, especially considering the high prevalence of the CHD population. Therefore, the creation of a single and common education program dedicated to all patients followed in the CHD referral centres from a large region is of interest.

After describing the structure of the program, this study aimed to determine which factors influenced the participation of adolescents and young adults with a CHD in this regional transition education program.

2. Materials and methods

2.1. Study design

This prospective observational cross-sectional multicentre study was carried out from January 2016 to January 2017 in three paediatric and congenital cardiology departments in France (University Hospital of Montpellier, University Hospital of Toulouse and Saint-Pierre Institute of Palavas-Les-Flots). These institutions represent the 3 tertiary care CHD referral centres in the Occitanie region (5.8 million inhabitants). Together, they created a unique and joint transition education program dedicated to all adolescents and young adults with a CHD living in this region.

2.2. Participants

Patients with any CHD, as defined by the international CHD classification [18], and aged from 13 to 25 years old, were prospectively recruited in all 3 centres during an outpatient visit. During the study period, all patients in this age group seen at the congenital cardiology consultation in the 3 CHD centres were offered to participate in one single and common transition education program. Participating patients (group A) could choose to be accompanied by any relative(s) of their choice (parents, siblings, partners, friends, etc.). If the patient did not accept, for whatever reason, to participate in the education program, they were offered to participate in the study as a control group (group B). The recruitment was made by the regular cardiologist, without any incentives methods or other specific tools than the medical information on the existence of the transition program.

Patients with a severe mental disorder were not eligible for this program and were referred to the national reference centre for developmental anomalies for specific care (Montpellier University Hospital, France).

2.3. Transition education program

As required by health authorities in France, the education program, named as the “transition program of adolescents and young adults with a CHD to adulthood”, was expertized and received authorization to recruit patients from the all over the region on December 2014. The structure of the transition program was built as recommended by the current guidelines, and adapted to our practice in patient education [7]:

(1) *First educational outpatient visit (*1 h*):* after giving his/her informed consent (legal guardians for patients < 18 y) to participate in the education program, the patient underwent an individual interview with a health educator (e.g. a specialist nurse), in the purpose of making an educational diagnosis and to determine the educational objectives [20,21].
(2) *Group session (1 day):* dedicated to patients and their relatives. Organised over a full day (from 10 a.m. to 4 pm). A patient association delegate was present during the group session, along with 1 adult congenital cardiologist, 1 paediatric cardiologist and 2 health educators. Each group was made of 5–8 patients with similar age ranges (13–17 y or 18–25 y) and types of CHD (Tetralogy of Fallot, single ventricle, transposition of the great arteries, atrial septal defect, ventricular septal defect, etc.)

The program’s content was structured into four parts:

a. *Medical aspects:* a congenital cardiologist and one health educator (e.g. knowledge of the disease, type of repair, follow-up in adult cardiology, complications), and different topics were addressed (physical activities, quality of life, jobs, graduate studies, sexuality and contraception, driver’s license, etc.).
b. *“Living with a CHD”:* Then, patients were gathered in a small room, with two health educators and a congenital cardiologist, without their relatives. The aim was to improve self-advocacy in deciding to act both independently from parents and medical providers, and in interdependence with them. Many education tools were used, depending on each group (play cards, “wheel of fortune”, experience sharing group etc.), and used examples from everyday life in relation with their CHD (prophylaxis of endocarditis, sports, etc.) or with any other aspects of their life (sexuality, tattoos, body piercing, school, etc.). Simultaneously with the previous patients group session, relatives were gathered in another meeting room with a congenital cardiologist. Through informal exchanges, the main goal was to identify factors that could promote or inhibit the patient’s autonomy and answer the parents and or relatives’ questions.
c. *Administrative workshop:* with the patient association delegate and a medical secretary, to address the following themes: social security, making appointment with adult cardiology CHD professionals, insurance, bank loans, etc.
d. *Synthesis and individual interview*: Simultaneously with the previous session, healthcare professionals joined together to analyse each patient’s situation, from the educational point of view (medical report, questionnaires, questions raised during the group sessions, etc.), and ultimately drafted an individual synthesis for each patient. At the end of the day, the patient and his/her relative(s) underwent an individual interview with a cardiologist and a health educator, to establish a personalized educational report. When needed, additional healthcare and educational objectives were defined, such as psychological support, knowledge reinforcement, cardiac rehabilitation [19], anticoagulant self-monitoring [20], etc.

(3) *Transfer preparation outpatient visit:* approximately 6 months after the group session, the patient underwent a medical visit with both a paediatric cardiologist and an adult congenital cardiologist, to provide an individual feedback from the group session and to prepare the transfer in the adult care CHD centre.

2.4. Questionnaires

During the outpatient visit, before any education intervention, patients from both groups filled in the same questionnaires, under specialist nurse supervision:

(1) The level of knowledge was assessed by the "transition readiness assessment" instrument. We selected a 10-item questionnaire reflecting the basic knowledge of the disease and its management (treatment, health care system, etc.), based on the study from Uzark et al., which showed acceptable reliability and acceptability of the transition readiness assessment questionnaire in the CHD population [9].
(2) The level of physical activity was assessed by the Ricci and Gagnon score, composed by 8 items (total score <16 points: no activity; 17–32 points: moderate activity; 33–40 points: intensive activity) [22].
(3) The health related quality of life was assessed by the PedsQL questionnaire, using the appropriate version for adolescents (13–18 years) or young adults (18–25 years) [23,24]. This generic quality of life questionnaire, designed for sick and healthy children, underwent transcultural adaptation and psychometric validation [25]. The 23 questions are completed in 10 min and explore 4 dimensions: physical capacity, emotional state, relation to others and study/work. The scores range from 0 to 100 (100 corresponding to the better quality of life possible), overall and by dimension.

2.5. CHD outcomes

All medical characteristics were collected from the patient's medical report (number of cardiac surgery, cardiac comorbidities, and treatment). The severity of the CHD was classified into four groups, as previously described by Uzark et al. [24].

When available, the data from the last cardiopulmonary exercise test (CPET) were collected, if performed less than 3 months before participation in the transition program, following the same methodology as in our previous CPET studies [8]. The following CPET variables were measured: maximum oxygen uptake ($VO2_{max}$), maximum heart rate, maximum load, ventilatory anaerobic threshold (VAT) using Beaver's method [26], ventilation efficiency (VE/VCO2 slope with VE = slope x VCO2 + b), and pulse oximetry (SpO2). $VO2_{max}$ and VAT values were normalized in a percentage of the predicted $VO2_{max}$ using reference values for cycle ergometer test in the general population [27].

The exposure to comorbidity risks factors (tobacco, alcohol, body piercing and tattoo) was informed and assessed in a qualitative way (yes/no question).

2.6. Formal aspects

As required by the French Ministry of Health, the transition patient education program received agreement from the regional health authorities (ARS-LR-2014-2186). The study was conducted in compliance with the Good Clinical Practices protocol and Declaration of Helsinki principles, and was approved by the South Mediterranean IV Ethics Committee (2016-A01681-50). Informed consent was obtained from all parents or legal guardians.

2.7. Statistical analysis

Patients' characteristics were presented using median and range (or mean and SD) for continuous variables and frequencies and proportions for categorical variables. Groups were compared using Student or Wilcoxon rank test for continuous variables and Chi-square or Fisher test for categorical ones. Statistical bilateral significance threshold was set at 5%.

A multiple linear regression was used to identify the explanatory factors for participation in the transition education program. The clinically relevant variables with a P-value ≤ 0.2 in the univariate analysis were included in the model. The final model was obtained using an upward selection based on the Akaïke information criterion (AIC) and with an exit threshold of 0.10. The normality of residues in the final model was tested using the Shapiro-Wilk test.

The statistical significance was set at 0.05 and analysed using SAS version 9 (SAS Institute, Cary, NC, USA).

3. Results

A total of 123 patients were included in the study, aged from 13 to 25 years (mean 19.6 ± 3.4 years, 40% female). They were 57 participants to the transition program (group A) and 66 non-participants (group B) (Table 1). Both groups presented with similar socio-demographic characteristics (Table 2). In particular, gender did not influence participation in the education transition program. However, there was a trend for a lower education level in the group B, with 34% of high school graduates against 55.5% in the group A (P = 0.06).

Both groups were similar in terms of living environment, distance to the educational centre, and transfer rate initiated to an adult CHD centre.

All remaining CHD variables (e.g. number of cardiac surgeries, medical treatment, cardiac and non-cardiac comorbidities) were similar in both groups and were not associated with the participation in the education program, apart from the severity of the CHD which was higher in the group A than in the group B (severity score of 3–4 in 84.2% vs. 60.6% patients, P = 0.02, respectively) (Table 2).

Exposure to risk behaviours (alcohol, tobacco, body piercing and tattoo) was significantly higher in the group A, with at least one risk factor for 84% of the patients, versus 55% in the group B (P = 0.02). Alcohol and body piercing were the two predominant risk exposures in univariate analysis. We found no difference in terms of level of exposure to risk behaviours between teenagers (<20 y) and young adults (≥20 y), with one risk behaviour or more for 67% vs. 70.5% (P = 0.50), respectively.

The level of knowledge was higher in the group B than in the group A (good knowledge in 64.3% vs. 22.7% patients, respectively, P = 0.013). The level of physical activity was higher in the group A than in the group B, with a mean total score of physical activity of 19.9 ± 6.2 vs. 15.9 ± 6.3 (P = 0.001), respectively. A total of 113 patients (92%) performed a CPET during the study period. CPET variables were similar in both groups, with a moderately impaired $VO2_{max}$ of 74% of theoretical values, overall. The mean VAT was moderately decreased to 21.2 ml/kg/min (e.g. 51% of the predicted $VO2_{max}$) and significantly lower in young adults (≥20 y) than teenagers (<20 y) (19.6 ml/kg/min vs. 22.6 ml/kg/min, P = 0.02, respectively).

The quality of life scores were not statistically different between group A and B, globally (78.9 ± 12.1 vs. 75.7 ± 16, P = 0.46, respectively), and within each dimension.

In multivariate analysis, the factors associated with the participation in the transition program were the severity of the CHD with an odd ratio (OR) of 4.16 (95% confidence interval (CI) [1.13–4.61], P = 0.03), and the presence of body piercing with an OR of 5.53 (95% CI [1.6–7.34], P = 0.01). There was a also trend in multivariate analysis for age < 20 years old (OR of 0.29, 95% CI [0.1–4.98], P = 0.03), and level of knowledge with (OR of 0.3, 95% CI [0.1–5.09], P = 0.02) (Table 3).

Table 1
Population characteristics.

Socio-demographic characteristics		
Average age (year, mean ± SD)		19.6 ± 3.4
Patient age groups, N (%)	≥ 20 years old	60 (49)
	< 20 years old	63 (51)
Gender, N (%)	Male	73 (60)
	Female	50 (40)
Height (cm, mean ± SD)		168 ± 10.8
Weight (kg, mean ± SD)		61.3 ± 12.1
BMI class, N (%)	Underweight	24 (19.7)
	Normal weight	86 (70.5)
	Overweight	8 (6.6)
	Obesity	4 (3.3)
Level of education, N (%)	College certificate	50 (42.4)
	Baccalaureate	52 (44.1)
	University	16 (13.6)
Living environment, N (%)	Rural	22 (17.9)
	Urban	101 (82.1)
Living with parents[a]	Yes	76 (67.3)
	No	37 (32.7)
Distance from home to educational centre (km, mean ± SD)		83.4 ± 109.3
Congenital heart disease, N (%)		
Severity score, N (%)	1	11 (8.9)
	2	24 (19.5)
	3	68 (55.3)
	4	20 (16.3)
CHD classification	Tetralogy of Fallot	30 (24.4)
	Pulmonary atresia with open septum	9 (7.3)
	Transposition of the great arteries	16 (13)
	Functionally univentricular heart	18 (14.6)
	Aortic coarctation	4 (3.3)
	Ventricular septal defect	6 (4.9)
	Atrial septal defect	7 (5.7)
	Aortic valve stenosis / Bicuspid aortic valve	10 (8.1)
	Other CHD	23 (18.7)
Cardiovascular events[a], N (%)		
Arrhythmia		40 (32)
Stroke		5 (4)
Coronary syndrome		4 (3.2)
Endocarditis		4 (3.2)
Pericarditis		3 (2.4)
Valvular comorbidities		17 (13.8)
Vascular comorbidities		10 (8.1)
Total		110 (89.4)
Other comorbidities, N (%)		
Asthma, allergy		14 (11.4)
Genetic syndrome		15 (12)
Orthopaedic anomaly		13 (10.5)
ENT and ophthalmic anomaly		11 (9)
Neuropsychiatric disease		15 (12)
Endocrinological disease		7 (5.6)
Uro-nephrology malformation		2 (3.3)
Digestive disorder		4 (1.6)
Other		4 (3.2)
Total		85 (69)
Risk behaviours, N (%)		
Alcohol consumption		62 (67.4)
Tobacco consumption		31 (33.3)
Piercing		27 (29.4)
Tattoo		20 (21.7)

Values are mean ± SD or N (%).
SD, standard deviation; BMI, Body Mass Index; CHD, congenital heart disease; ENT, ear, nose, and throat disease.
[a] A patient can have multiple events.

Table 2
Patients in the transition program (participants) compared to patients with regular follow-up (non-participants).

		Transition N = 57	No transition N = 66	P-value
Average age (cm, mean ± SD)		19.1 ± 2.7	20.1 ± 3.9	0.1
Gender, N (%)	Male	31 (54.4)	42 (63.6)	0.2
	Female	26 (45.6)	24 (36.4)	
Height (cm, mean ± SD)		168.5 ± 12.8	168 ± 8.9	0.4
Weight (kg, mean ± SD)		61.9 ± 13.7	60.9 ± 10.7	0.8
BMI class, N (%)	Underweight	12 (21.4)	12 (18.2)	0.6
	Normal weight	38 (67.9)	48 (72.7)	
	Overweight	3 (5.4)	1 (1.5)	
	Obesity	3 (5.4)	5 (7.6)	
Level of education, N (%)	College certificate	17 (31.5)	33 (51.6)	0.06
	Baccalaureate	30 (55.6)	22 (34.4)	
	University	7 (13)	9 (14.1)	
Living environment, N (%)	Rural	10 (17.5)	12 (18.2)	0.9
	Urban	47 (82.5)	54 (81.8)	
Living with parents N (%)		36 (72)	40 (63)	0.34
Distance from home to educational centre (km, mean ± SD)		90 ± 80.6	77.5 ± 129.4	0.1
Effective transfer in ACHD centre, N (%)		30 (52.6)	37 (56)	0.7
Severity score, N (%)	1	2 (3.5)	9 (13.6)	**0.02**
	2	7 (12.3)	17 (25.8)	
	3	39 (68.4)	29 (43.9)	
	4	9 (15.8)	11 (16.7)	
Heart surgeries, N (%)	0	7 (12.3)	11 (16.7)	0.7
	1	27 (47.4)	34 (51.5)	
	2	14 (24.6)	14 (21.2)	
	>2	9 (15.8)	7 (10.6)	
Patient under medication, N (%)		21 (36.8)	22 (33.3)	0.6
Emergency cardiac consultations, N (%)		9 (21)	14 (21.9)	0.9
Non-cardiac comorbidities, N (%)		28 (50)	22 (33.3)	0.06
Cardiac comorbidities, N (%)		28 (50)	44 (66.7)	0.06
$VO2_{max}$ (ml/kg/min, mean ± SD)		29.7 ± 7.6	30.2 ± 7.9	0.6
% predicted $VO2_{max}$ (%, mean ± SD)		74.1 ± 16.2	74.6 ± 17.4	0.9
VAT (ml/kg/min, mean ± SD)		21.1 ± 5.9	21.2 ± 7.3	0.7
% predicted VAT (%, mean ± SD)		51.3 ± 12.7	50.8 ± 13.3	0.8
VE/VCO2 slope (%, mean ± SD)		31.3 ± 6.6	32.2 ± 5.9	0.3
FVC (%, mean ± SD)		88.5 ± 17.1	85.2 ± 16	0.4
FEV1 (%, mean ± SD)		86.3 ± 16.7	83.9 ± 16.9	0.5
FEV1/FVC (%, mean ± SD)		83.7 ± 8.7	83.9 ± 8.9	0.8

Values are mean ± SD or N (%). Significant P-values are marked in bold.
BMI, body mass index; ACHD, adult congenital heart disease; $VO2_{max}$, maximum oxygen uptake; VAT, ventilatory anaerobic threshold; VE, minute ventilation; VCO2, CO2 production; FVC, forced vital capacity; FEV1, forced expiratory volume in 1 s.

Table 3
Factors associated with the participation in the transition program.

Effect	Odds ratio	95% CI		P-value
Patients over 20 years	0.29	0.1	4.98	**0.03**
Severe heart disease (class 3 and 4)	4.16	1.13	4.61	**0.03**
Alcohol consumption	3.12	0.97	3.67	0.06
Piercing	5.53	1.6	7.34	**0.01**
Correct knowledge of the pathology	0.3	0.1	5.09	**0.02**

CI, confidence interval.
Significant P-values are marked in bold.

4. Discussion and conclusion

4.1. Discussion

This multicentre prospective study provided relevant data on factors influencing the participation of adolescents and young adults with CHD in a regional transition education program, from a cohort of 123 patients. To our knowledge, this study addressed, for the first time in the CHD population, the factors associated with participation and non-participation to an education program, using an observational prospective study design. Considering the high risk of loss to follow-up during transfer from paediatric to adult care in this population, our results may be of great interest for healthcare professionals working on transition education programs.

Patients with complex CHD are likely to experience more complications in adulthood and such education programs may participate in improving the prognosis [28]. Indeed, this transition program intends to promote patients' autonomy, help them acquiring a better knowledge of their health condition, and provide solutions to include their CHD in everyday life. This is in line with previous studies from *Uzark et al.*, showing the direct link between level of knowledge, self-efficacy and quality of life [9]. Nevertheless, many studies stated that patients with less severe CHD were also at high risk of loss to follow-up [29–31]. Therefore, this transition program was also dedicated to patients with non-complex CHD, such as atrial and ventricular septal defects. Indeed, *Iversen et al.* showed that these patients lost to follow-up usually described their health perception as "as good as the general population" [32]. Therefore transition education programs find their place in helping these teenagers and young adults with CHD to understand the value of medical follow-up in adulthood.

Another aim of the transition program was to avoid the exposure to controllable risk factors in patients with CHD, and limit cardiac and non-cardiac comorbidities in this population. In this study, overall, nearly 10% patients were concerned by overweight or obesity and the level of physical activity was rather low. This impaired cardiovascular health has been previously evaluated up to 68% of youth and 73% of adults with CHD [33].

The low level of physical activity had no impact on patients' participation in the transition program. In this cohort, more than 90% of the patients had undergone CPET before participation in the transition program and their $VO2_{max}$ was moderately impaired (74% of predicted values). Interestingly, similar CPET results were found in patients who did not participate in the transition program, despite a lower level of CHD severity. In this cohort, overall, the VAT was moderately decreased to nearly 50% of the predicted $VO2_{max}$ and this decrease was even more marked in young adults (>20 years old). That means that these patients are at risk of muscular deconditioning, from an early age. We recently showed that $VO2_{max}$ in adolescents with CHD correlated with their quality of life and decreased with age faster than controls, especially for the most complex CHD [34]. Moreover, we also found that muscular deconditioning affected three times more children with CHD than controls, even those with simple CHD [35]. Indeed, adolescents with CHD are often hovered over by their parents, stigmatized by their teachers, and eventually remain on the "sidelines" [17,36]. In this context, education programs dedicated to this population should participate in promoting self-confidence to the adolescent, reassuring his or her family, and motivating them to engage the young patient in physical activity. Therefore, education messages need to encourage physical activity, and fight against the consequences of a sedentary lifestyle. As a result, the promotion of physical activity in the transition program has become our priority, in order to improve the prognosis of this young population. Furthermore, this transition program promotes cardiac rehabilitation in these young patients, aiming to slow the decline of oxygen uptake in adulthood, include physical activity in their daily life and improve their quality of life.

This study highlighted the existence of a significant level of exposure to other risk behaviours such as tobacco and alcohol, especially in the educated group. Interestingly, these risk behaviours were similar to those reported in the general population (e.g. 60% of regular alcohol consumption and 30% of smokers in adolescents and young adults) [37]. Yet, a recent Canadian study reported that tobacco exposition in a similar young CHD population represented only 12–14% of the cohort [33]. However, we need to take in account that the smoking rate is lower in Canada than in France [38]. Youth alcohol use should be taken into consideration, as even a low consumption of alcohol may result in early vascular lesions [39]. Moreover, a substantial number of patients with CHD are at risk of cardiac re-interventions during early adulthood (e.g. cardiac surgery or catheterization). Therefore, such education programs need to focus on limiting the exposition of these patients to situation at risks of postoperative complications (endocarditis, thrombosis, arrhythmia, etc.) [40].

In this study, risk behaviours were present early in age, from adolescence, as in similar studies [41]. Adolescents living with a chronic disease represent a vulnerable group; therefore, prevention messages have to be early delivered to patients and their families. Moreover, the presence of such risk factors might be predictive of transfer failure, as shown by Reid et al. [30] When starting the transition program, our education team was not aware of the existence of such a high level of risk behaviours, and, after this study, decided to reinforce prevention messages.

Patients with a low level of knowledge of their disease were, in this study, more prone to participate in the education program. Nevertheless, it is difficult to identity what or who, between the patients themselves, their relatives, or their paediatric or congenital cardiologist, precisely influenced this participation. Indeed, no psychological quality analysis was performed in this study. Similarly, the study from *Ladouceur et al.*, observed that therapeutic education including basic knowledge led to a better understanding of the disease 11 months after the educative action, as compared to a non-educated group [13]. Considering that the level of basic knowledge in this population was associated with their level of quality of life [9], this transition education program purposely dedicated a significant amount of time on patient-centred theoretical course of basic knowledge.

Interestingly, teenagers were more likely to participate in the transition program than young adults over 20 years old. Patients over 20 years old might find it difficult to take a day off from work or school to participate in a group session. Such patients might therefore be more interested in individual education sessions, during their routine follow-up. From a general perspective, transition has to be considered as a process that will ultimately lead to the transfer to adult cardiology [12]. The process has to be engaged as soon as possible to ensure the success of the transfer. In this education program, the mean age of patients transfer was 20 years old, as commonly reported [7,8]. We consider that age should not be the main element of decision-making to transfer. Indeed, 13 out of 60 young adults were still followed in paediatric cardiology at the time of the study. This is in line with the study from *Hilderson et al.* in which 50 centres out of 51 in Europe and America did not consider age as a major element of transfer decision [12].

Parents must be considered as main actors of the transition process, and should not be left aside. The medical history of their children is part of their own life, and, as we recently showed, most parents have experienced psychological trauma [42,43]. Giving autonomy to their child may generate tension and anxiety in parents. Therefore, transition educational programs should also support parents during transition and transfer process to ensure its success.

Running an education program is very time consuming. Every session required the presence of two cardiologists, two specialist nurses and one patient association delegate. The originality of this program is based on its unique, regional and joint structure between 3 tertiary care CHD centres. Therefore, all paediatric and adult congenital cardiologists from all 3 institutions were particularly motivated to include their patients and participate themselves in this common education program. The support from the patient association also played an important role. The role of transition care manager may be of great interest in the CHD population [44]. In 2018, French health authorities have labelled a Master's degree in advanced practice nursing, and implemented a specific sub-specialty on chronic cardiac conditions. Therefore, the results of this study were presented to our regional healthcare authorities (e.g. "Agence Régionale de Santé Occitanie") and our institution decided to train and recruit an advanced practice nurse as transition care manager".

Interestingly, the distance from the patients' home and the transition education centre did not impact their participation. The median distance was even higher in the educated group (66 km) than in the non-educated group (33 km). In France, the social security reimburses the patient's costs to participate in labelled education programs.

4.2. Study limitations

This study included patients from 3 tertiary care centres and may not represent CHD in the general population. Indeed, complex CHD, such as the transposition of the great arteries, was over-represented (47% vs. 20% in the French national CHD registry), and simple CHD, such as ventricular septal, was under-represented (7% vs. 52% in the national registry) [45]. However, genetic anomaly was as represented in this cohort as in the national registry (12% vs. 14%).

The information about the existence of the transition program was first given by the paediatric or congenital cardiologist during the outpatient visit. Therefore, the actual participation in the

program depended on the way this information was provided. Indeed, the oral information provided by each physician might have been heterogeneous and no specific information tool was used when patients were offered to participate in the education program. After this study, a video (https://www.youtube.com/watch?v=OJ67oIm0Wv8) was created to harmonize this information.

The results come from a non-randomized study. The real impact of the transition program needs to be determined with a higher level of evidence. Therefore, we recently started a randomized multicentre controlled trial about the impact of this regional transition education program on the quality of life of teenagers and young adults with CHD (https://clinicaltrials.gov/show/NCT03005626). Moreover, this study will include a psychological quality analysis.

4.3. Conclusion

This multicentre observational study from a cohort of 123 patients with a CHD provided relevant information on the factors influencing their participation in a regional transition education program, dedicated to adolescents and young adults followed in three referral CHD centres. Patients with complex CHDs, patients with a low level of knowledge of their disease, patients with risk behaviours, and adolescents were more likely to participate in the transition education program. However, educated and non-educated patients were similar in many characteristics, such as a low level of physical activity, and the program appeared to be rather non-selective.

4.4. Practice implications

Further randomized studies are necessary to evaluate the impact of transition education program on quality of life, successful transfer to ACHD centre and, ultimately, prognosis.

Author contributions

Conceived and designed the experiments: OW and PAm. Performed the experiments: OW, PAm, CB, PAc, YD, NS and SG. Analysed the data: HA, KL, MCP and CS. Wrote the paper: OW, HA and PA. Database searches, data extraction, and input to final review: HA and KL.

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References

[1] C.A. Warnes, R. Liberthson, G.K. Danielson, A. Dore, L. Harris, J.I. Hoffman, et al., Task force 1: the changing profile of congenital heart disease in adult life, J. Am. Coll. Cardiol. 37 (2001) 1170–1175.

[2] L.G. Spector, J.S. Menk, J.H. Knight, C. McCracken, A.S. Thomas, J.M. Vinocur, et al., Trends in long-term mortality after congenital heart surgery, J. Am. Coll. Cardiol. 71 (2018) 2434–2446.

[3] P. Moons, L. Bovijn, W. Budts, A. Belmans, M. Gewillig, Temporal trends in survival to adulthood among patients born with congenital heart disease from 1970 to 1992 in Belgium, Circulation 122 (2010) 2264–2272.

[4] Z. Mandalenakis, A. Rosengren, K. Skoglund, G. Lappas, P. Eriksson, M. Dellborg, Survivorship in children and young adults with congenital heart disease in Sweden, JAMA Intern. Med. 177 (2017) 224–230.

[5] C. Wren, J.J. O'sullivan, Survival with congenital heart disease and need for follow up in adult life, Heart 85 (2001) 438–443.

[6] A.J. Marelli, A.S. Mackie, R. Ionescu-Ittu, E. Rahme, L. Pilote, Congenital heart disease in the general population: changing prevalence and age distribution, Circulation 115 (2006) 163–172.

[7] C. Sable, E. Foster, K. Uzark, K. Bjornsen, M.M. Canobbio, H.M. Connolly, et al., Best practices in managing transition to adulthood for adolescents with congenital heart disease: the transition process and medical and psychosocial issues: a scientific statement from the american heart association, Circulation 123 (2011) 1454–1485.

[8] Endorsed by the Association for European Paediatric Cardiology (AEPC), Authors/Task Force Members, H. Baumgartner, P. Bonhoeffer, N.M.S. De Groot, F. de Haan, et al., ESC guidelines for the management of grown-up congenital heart disease (new version 2010): the task force on the management of grown-up congenital heart disease of the European Society of Cardiology (ESC), Eur. Heart J. 31 (2010) 2915–2957.

[9] K. Uzark, C. Smith, J. Donohue, S. Yu, K. Afton, M. Norris, et al., Assessment of transition readiness in adolescents and young adults with heart disease, J. Pediatr. 167 (2015) 1233–1238.

[10] P. Moceri, E. Goossens, S. Hascoet, C. Checler, B. Bonello, E. Ferrari, et al., From adolescents to adults with congenital heart disease: the role of transition, Eur. J. Pediatr. 174 (2015) 847–854.

[11] D.V. Schidlow, S.B. Fiel, Life beyond pediatrics: transition of chronically Ill adolescents from pediatric to adult health care systems, Med. Clin. North Am. 74 (1990) 1113–1120.

[12] D. Hilderson, A.S. Saidi, K. Van Deyk, A. Verstappen, A.H. Kovacs, S.M. Fernandes, et al., Attitude toward and current practice of transfer and transition of adolescents with congenital heart disease in the United States of America and Europe, Pediatr. Cardiol. 30 (2009) 786–793.

[13] M. Ladouceur, J. Calderon, M. Traore, R. Cheurfi, C. Pagnon, D. Khraiche, et al., Educational needs of adolescents with congenital heart disease: impact of a transition intervention programme, Arch. Cardiovasc. Dis. 110 (2017) 317–324.

[14] P. Amedro, F. Bajolle, H. Bertet, R. Cheurfi, D. Lasne, E. Nogue, et al., Quality of life in children participating in a non-selective INR self-monitoring VKA-education programme, Arch. Cardiovasc. Dis. 111 (2018) 180–188.

[15] P. Amedro, A. Gavotto, A. Legendre, K. Lavastre, C. Bredy, G. De La Villeon, et al., Impact of a centre and home-based cardiac rehabilitation program on the quality of life of teenagers and young adults with congenital heart disease: the QUALI-REHAB study rationale, design and methods, Int. J. Cardiol. 283 (2019) 112–118.

[16] J. Nicolarsen, Transition of adolescents and young adults with congenital heart disease: challenges, progress, and future improvements, Pediatr. Ann. 46 (2017) e224–8.

[17] P. Amedro, R. Dorka, S. Moniotte, S. Guillaumont, A. Fraisse, B. Kreitmann, et al., Quality of life of children with congenital heart diseases: a multicenter controlled cross-sectional study, Pediatr. Cardiol. 36 (2015) 1588–1601.

[18] L. Houyel, B. Khoshnood, R.H. Anderson, N. Lelong, A.-C. Thieulin, F. Goffinet, et al., Population-based evaluation of a suggested anatomic and clinical classification of congenital heart defects based on the International Paediatric and Congenital Cardiac Code, Orphanet J. Rare Dis. 6 (2011) 64.

[19] P. Amedro, F. Bajolle, H. Bertet, Quality of Life in Children Participating in a Non-Selective INR Self-monitoring VKA-Education Programme, (2018) .

[20] P.-Y. Traynard, Le diagnostic éducatif. Une bonne idée pour évoluer, Médecine Mal Métaboliques. 4 (2010) 31–37.

[21] H. Mosnier-Pudar, Some soughts on the educational diagnosis, Médecine Mal Métaboliques. 7 (2013) 465–469.

[22] A. Vuillemin, G. Denis, F. Guillemin, C. Jeandel, A review of evaluation questionnaires for physical activity, Rev. Epidemiol. Sante Publique 46 (1998) 49–55.

[23] J.W. Varni, M. Seid, P.S. Kurtin, PedsQL 4.0: reliability and validity of the Pediatric Quality of Life Inventory version 4.0 generic core scales in healthy and patient populations, Med. Care 39 (2001) 800–812.

[24] K. Uzark, K. Jones, T.M. Burwinkle, J.W. Varni, The Pediatric Quality of Life Inventory™ in children with heart disease, Prog. Pediatr. Cardiol. 18 (2003) 141–149.

[25] S. Tessier, A. Vuillemin, J.-L. Lemelle, S. Briançon, Propriétés psychométriques du questionnaire générique français « Pediatric Quality of Life Inventory Version 4.0» (PedsQLTM 4.0), Rev. Eur. Psychol. Appliquée Eur. Rev. Appl. Psychol. 59 (2009) 291–300.

[26] W.L. Beaver, K. Wasserman, B.J. Whipp, A new method for detecting anaerobic threshold by gas exchange, J. Appl. Physiol. 1986 (60) (1985) 2020–2027.

[27] N.L. Jones, L. Makrides, C. Hitchcock, T. Chypchar, N. McCartney, Normal standards for an incremental progressive cycle ergometer test, Am. Rev. Respir. Dis. 131 (1985) 700–708.

[28] C. Karsenty, P. Maury, N. Blot-Souletie, M. Ladouceur, B. Leobon, V. Senac, et al., The medical history of adults with complex congenital heart disease affects

their social development and professional activity, Arch. Cardiovasc. Dis. 108 (2015) 589–597.
[29] E. Yeung, J. Kay, G.E. Roosevelt, M. Brandon, A.T. Yetman, Lapse of care as a predictor for morbidity in adults with congenital heart disease, Int. J. Cardiol. 125 (2008) 62–65.
[30] G.J. Reid, M.J. Irvine, B.W. McCrindle, R. Sananes, P.G. Ritvo, S.C. Siu, et al., Prevalence and correlates of successful transfer from pediatric to adult health care among a cohort of young adults with complex congenital heart defects, Pediatrics 113 (2004) e197–205.
[31] S. Kikkenborg Berg, P.U. Pedersen, Perception of general health in adults with congenital heart disease who no longer attend medical follow-up, Eur. J. Cardiovasc. Nurs. 7 (2008) 264–268.
[32] K. Iversen, N.G. Vejlstrup, L. Sondergaard, O.W. Nielsen, Screening of adults with congenital cardiac disease lost for follow-up, Cardiol. Young 17 (2007) 601–608.
[33] K.C. Harris, C. Voss, K. Rankin, B. Aminzadah, R. Gardner, A.S. Mackie, Modifiable cardiovascular risk factors in adolescents and adults with congenital heart disease, Congenit. Heart Dis. 13 (2018) 563–570.
[34] P. Amedro, M.C. Picot, S. Moniotte, R. Dorka, H. Bertet, S. Guillaumont, et al., Correlation between cardio-pulmonary exercise test variables and health-related quality of life among children with congenital heart diseases, Int. J. Cardiol. 203 (2016) 1052–1060.
[35] P. Amedro, A. Gavotto, S. Guillaumont, H. Bertet, M. Vincenti, G. De La Villeon, et al., Cardiopulmonary fitness in children with congenital heart diseases versus healthy children, Heart Br. Card. Soc. 104 (2018) 1026–1036.
[36] F. Moola, C. Fusco, J.A. Kirsh, The perceptions of caregivers toward physical activity and health in youth with congenital heart disease, Qual. Health Res. 21 (2011) 278–291.
[37] Julie-Émilie Adès, François Beck, Anne-Claire Brisacier, Agnès Cadet-Taïrou, Cristina D.íaz Gómez, Michel Gandilhon, Gauduchon Thibault, Éric Janssen, Olivier Le Nézet, Aurélie Lermenier-Jeannet, Magali Martinez, Maitena Milhet, Carine Mutatayi, Thomas Néfau, Ngantcha Marcus, Ivana Obradovic, Christophe Palle, Caroline Protais, Stanislas, Spilka de l'O.F.D.T, ainsique João Matias, André Noor, Jeunes et addictions. Observatoire français des drogues et des toxicomanies, (2016) .
[38] M. Ng, M.K. Freeman, T.D. Fleming, M. Robinson, L. Dwyer-Lindgren, B. Thomson, et al., Smoking prevalence and cigarette consumption in 187 countries, 1980–2012, JAMA 311 (2014) 183–192.
[39] M. Charakida, G. Georgiopoulos, F. Dangardt, S.T. Chiesa, A.D. Hughes, A. Rapala, et al., Early vascular damage from smoking and alcohol in teenage years: the ALSPAC study, Eur. Heart J. 40 (2019) 345–353.
[40] C.E. Mascio, S.K. Pasquali, J.P. Jacobs, M.L. Jacobs, E.H. Austin, Outcomes in adult congenital heart surgery: analysis of the Society of Thoracic Surgeons database, J. Thorac. Cardiovasc. Surg. 142 (2011) 1090–1097.
[41] T. Santos, M. Ferreira, M.C. Simões, M.C. Machado, M.G. de Matos, Chronic condition and risk behaviours in Portuguese adolescents, Glob. J. Health Sci. 6 (2014) 227–236.
[42] O. Werner, F. El Louali, V. Fouilloux, P. Amedro, C. Ovaert, Parental anxiety before invasive cardiac procedure in children with congenital heart disease: contributing factors and consequences, Congenit. Heart Dis. (2019), doi:http://dx.doi.org/10.1111/chd.12777 [Epub ahead of print].
[43] M. Skreden, H. Skari, U.F. Malt, G. Haugen, A.H. Pripp, A. Faugli, et al., Long-term parental psychological distress among parents of children with a malformation-a prospective longitudinal study, Am. J. Med. Genet. A 152A (2010) 2193–2202.
[44] M.M. Ciccone, A. Aquilino, F. Cortese, P. Scicchitano, M. Sassara, E. Mola, et al., Feasibility and effectiveness of a disease and care management model in the primary health care system for patients with heart failure and diabetes (Project Leonardo), Vasc. Health Risk Manag. 6 (2010) 297–305.
[45] B. Khoshnood, N. Lelong, L. Houyel, A.-C. Thieulin, J.-M. Jouannic, S. Magnier, et al., Prevalence, timing of diagnosis and mortality of newborns with congenital heart defects: a population-based study, Heart Br. Card. Soc. 98 (2012) 1667–1673.