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ORIGINAL ARTICLE

Characterizing Decision-Making Surrounding Exercise in ARVC: Analysis of Decisional Conflict, Decisional Regret, and Shared Decision-Making

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BACKGROUND: Limiting high-intensity exercise is recommended for patients with arrhythmogenic right ventricular cardiomyopathy (ARVC) due to its association with penetrance, arrhythmias, and structural progression. Guidelines recommend shared decision-making (SDM) for exercise level, but there is little evidence regarding its impact. Therefore, we sought to evaluate the extent and implications of SDM for exercise, decisional conflict, and decisional regret in patients with ARVC and at-risk relatives.

METHODS: Adults diagnosed with ARVC or with positive genetic testing enrolled in the Johns Hopkins ARVC Registry were invited to complete a questionnaire that included exercise history and current exercise, SDM (SDM-Q-9), decisional conflict, and decisional regret.

RESULTS: The response rate was 64.8%. Two-thirds of participants (68.0%, n=121) reported clinically significant decisional conflict regarding exercise at diagnosis/genetic testing (DCS [decisional conflict scale]≥25), and half (55.1%, n=98) in the past year. Prevalence of decisional regret was also high with 55.3% (n=99) reporting moderate to severe decisional regret (DRS [decisional regret scale]≥25). The extent of SDM was highly variable ranging from no (0) to perfect (100) SDM (mean, 59.6±25.0). Those diagnosed in adolescence (≤age 21) reported significantly more SDM (P=0.013). Importantly, SDM was associated with less decisional conflict (ß=–0.66, R^2=0.567, P<0.01) and decisional regret (ß=–0.37, R^2=0.180, P<0.001) and no difference in vigorous intensity aerobic exercise in the 6 months after diagnosis/genetic testing or the past year (P=0.56; P=0.34, respectively).

CONCLUSIONS: SDM is associated with lower decisional conflict and decisional regret; and no difference in postdiagnosis exercise. Our data thus support SDM as the preferred model for exercise discussions for ARVC.

Key Words: arrhythmogenic right ventricular cardiomyopathy ■ exercise ■ genetic counseling ■ shared decision-making

Arrhythmogenic right ventricular cardiomyopathy (ARVC) is an inherited cardiovascular condition associated with frequent ventricular arrhythmias, cardiomyopathy, and increased risk of sudden cardiac death. Pathogenic variants in genes encoding the cardiac desmosome, a protein structure linking cardiac myocytes, are the most common genetic cause of ARVC.[1] Frequent, intense aerobic exercise is associated with worse cardiovascular outcomes in patients with ARVC and their at-risk relatives likely due to the resulting structural and functional abnormalities.[2,3] For those at risk for ARVC due to a pathogenic or likely pathogenic desmosomal variant, exercise is associated with increased penetrance and risk of sustained ventricular arrythmias.[2,4] For those diagnosed with ARVC, exercise is associated with higher arrhythmia burden, worse structural involvement, and heart failure.[5] Consequently, it is typically recommended that patients with

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Nonstandard Abbreviations and Acronyms

ARVC	arrhythmogenic right ventricular cardiomyopathy
SDM	shared decision-making
DCS	decisional conflict scale
DRS	decisional regret scale
GT	genetic testing

ARVC avoid most competitive sports and frequent high-intensity aerobic activity.[6]

Nonetheless, decisions surrounding exercise participation for patients with ARVC and at-risk relatives are complex. The ideal level of exercise for a specific patient is uncertain, may vary by genotype, and is based on an ever-evolving evidence base.[4] Patients must weigh the risks associated with exercise against the physical, psychological, and social benefits that exercise can bring. Many of those diagnosed with ARVC are highly active individuals for whom exercise restriction may be particularly challenging.[7,8]

In recognition of this complexity, guidelines recommend that exercise decisions for those with or at risk for ARVC follow a shared decision-making (SDM) model.[6] SDM is an increasingly popular model in medicine that aims to increase patient autonomy and engagement in medical decision-making. Although SDM has been defined inconsistently throughout the literature, broadly, there are 2 components to SDM: clarifying patient values and exchanging information about options and their risks and benefits.[9–11] The utility of SDM in exercise decision-making for people with inherited heart conditions is disputed. Some clinicians call for exercise decision-making to follow an SDM model for patients with inherited cardiomyopathy and arrhythmia syndromes.[12–14] Still, other clinicians refute the utility of SDM in these exercise decisions, with particular concern for young athletes, citing patient perceptions of SCD risk estimates as low and the motivation level to continue sports participation as reasons why SDM might not be a fitting model in this space.[15] While there are many opinions on the matter, there has been little work to describe what clinical support patients are receiving with regard to exercise decision-making, and almost none describing the decision-making process and outcomes of adolescent patients.

Decisional conflict and decisional regret are psychosocial outcomes of decision-making. Both decisional conflict and decisional regret have been associated with poor psychosocial and medical outcomes. Decisional conflict conceptualizes feelings of uncertainty, lack of support, and lack of knowledge that can come with making a complex decision.[16] It has been associated with delaying medical decisions, lower physician satisfaction, fretting, nervousness, and increased decisional regret.[17–19] Decisional regret conceptualizes the extent to which a person retrospectively considers the decision they made to have been the best decision for them. Importantly, this can refer to either the decision that was made—the content—or the way the decision happened—the process (ie, did the person feel supported, did the person have all the information they needed at the time of decision-making).[20] Decisional regret related to medical decisions has been associated with decreased role and social functioning, increased physical pain, lower quality of life, and increased depression and anxiety.[18,21,22]

SDM has been associated with decreased decisional conflict and decisional regret, as well as increased adherence to decisions in some populations.[17,23–26] However, in contrast to much of the existing medical decision-making literature, exercise decision-making happens throughout the lifespan, rather than at a single decision-making time or time period (such as for a surgical decision or treatment of a time-limited disease). It is uncertain whether the predicted benefits of SDM would be applicable to exercise decision-making for ARVC. Furthermore, the appropriateness of SDM application in adolescents is debated because while they are capable of making many decisions independently, there are concerns about their ability to fully comprehend risk.[27] This is of concern for adolescents with ARVC because the risks associated with ARVC are serious and potentially irreversible.

In summary, exercise decisions are difficult for those with ARVC, and SDM is recommended, but there has been no study of either the extent of SDM for exercise decision-making or its consequences. Therefore, via a cross-sectional questionnaire administered to adults in the Johns Hopkins ARVC registry, we sought to describe exercise decision-making and to analyze associations between SDM and decisional outcomes. Our aims were to (1) measure the extent to which SDM for exercise is occurring, (2) characterize which patients are most likely to engage in exercise SDM with a particular focus on adolescent patients and athletes, and (3) determine how SDM is associated with decisional conflict, decisional regret, and adherence in patients with ARVC and genetically at-risk relatives.

METHODS

The data that support the findings of this study may be available as a limited data set from the corresponding author upon reasonable request.

This study was approved by a Johns Hopkins School of Medicine Institutional Review Board and participants provided written informed consent.

Methods are available as Supplemental data (Supplemental Methods).

RESULTS

Study Population

A total of 316 invitations were sent, and 205 individuals completed the questionnaire, resulting in a response rate

of 64.8%. Of the 205 responses, 2 were removed because they did not self-report a clinical diagnosis of ARVC or positive GT for ARVC, and 9 had been diagnosed more than 11 years ago. This left 194 responses for analysis.

The demographic and exercise history of the population are summarized in Tables 1 and 2. The average age of the population at the time of questionnaire was 43.9±15.0 years with men and women equally represented. The population was overwhelmingly White (92.9%). Most of our population had a clinical diagnosis of ARVC (76.7%, n=148). Consistent with this, most had an ICD at the last follow-up (59.4%, n=111), and 39.4% (n=54) had presented with a sustained ventricular arrhythmia.

Table 1. Demographic and Clinical Characteristics (n=194)*

	Range	N (%) or mean±SD
Gender† (No. of female)		105 (54.1)
Age	18–82	43.9±15.0
Race/ethnicity		
White		171 (92.9)
Black		2 (1.1)
Latinx		5 (2.7)
Asian		1 (0.5)
Middle Eastern		3 (1.1)
Other		2 (1.1)
Age at time of diagnosis or GT	10–75	38.6±15.2
Age categories		
No. of diagnosed 18 or younger		20 (10.6)
No. of diagnosed 21 or younger		29 (15.3)
No. of diagnosed 25 or younger		40 (21.2)
Years since diagnosis	0–11	5.0±2.9
ARVC status (number with ARVC diagnosis)		148 (76.7)
ICD at last follow-up		111 (59.4)
Sustained ventricular arrhythmia at presentation		54 (39.4)
Lived alone at time of diagnosis		14 (7.2)
Lived alone at time of questionnaire		23 (11.9)
Education level		
Some high school		2 (1.0)
Completed high school/GED		9 (4.6)
Some college		25 (12.9)
Completed college		71 (36.6)
Some graduate school		15 (7.7)
Completed graduate school		72 (37.1)
Relationship status		
Single		41 (20.6)
Married or partnered		151 (79.4)
Genotype		
PKP2 variant		80 (41.2)
DSP variant		34 (17.5)
Other variant		35 (18.0)
No variant identified		36 (18.6)

ARVC indicates arrhythmogenic right ventricular cardiomyopathy; GED, General Equivalency Diploma; GT, genetic testing; and ICD, implantable cardioverter defibrillator.

*Some items were left blank by some participants. Percentages reflect the proportion of those who answered the items (ie, those who did not answer were excluded).

†Gender options included male, female, and nonbinary/third gender. No participants responded that they identified as nonbinary/third gender.

Table 2. Exercise History*

	Range	N (%) or mean±SD
Ever participated in competitive sports		143 (77.7)
Athlete identity		
Identified as an athlete at the time of diagnosis		134 (69.8)
Identified as an active individual at the time of diagnosis		179 (93.7)
Currently identifies as an athlete		27 (15.5)
Currently identifies as an active individual		107 (60.8)
Vigorous activity		
Engaged in vigorous activity in the year before diagnosis		124 (63.9)
Engaged in vigorous activity in the 6 mo after diagnosis		16 (8.2)
Engaged in vigorous activity in the year before study completion		13 (6.7)
Hours spent doing vigorous activity per week		
In the year before diagnosis	0.0 to 44.3	4.9±7.2
In the 6 mo after diagnosis	0.0 to 15.4	0.5±1.9
In the year before study completion	0.0 to 6.7	0.2±0.8

*Some items were left blank by some participants. Percentages reflect the proportion of those who answered the items (ie, those who did not answer were excluded).

Exercise Decision-Making

As shown in Table 2, the population was particularly athletic. More than three-quarters (77.7%, n=143) reported participating in a competitive sport at some time during their life, and 69.8% reported that they viewed themselves as athletes in the year before they were diagnosed. Nearly all participants (93.7%, n=179) viewed themselves as active individuals in the year before diagnosis. Overall, participants were highly engaged in vigorous activity before diagnosis or GT. In the year before diagnosis or GT, 63.9% (n=124) of participants participated in some level of regular vigorous activity and participants averaged 4.9±7.2 hours per week at vigorous intensity exercise (median, 2.8; interquartile range, 6.5).

Participants had overwhelmingly decreased exercise since their ARVC diagnosis or GT. Nearly all (94.6%, n=175) reported that they had decreased their exercise because of their ARVC diagnosis or GT. Only 1 (0.5%) participant reported increased exercise since diagnosis, and 4.9% (n=9) reported that they had not changed their exercise since diagnosis or GT. After diagnosis or GT, self-reported vigorous activity level also decreased greatly. In the 6 months after their diagnosis or GT, 8.2% (n=16) of participants participated in vigorous activity. In the year before study completion, 6.7% (n=13) of participants

participated in vigorous activity. In the 6 months after diagnosis or GT, participants averaged 0.5±1.9 hours per week of vigorous activity with the median, first quartile, and third quartile all equal to 0.0. In the year before study completion, the average time spent on vigorous activities was 0.2±0.8 hours per week, again with the median, first quartile, and third quartile again equal to 0.0.

Shared Decision-Making

The distributions of SDM scores for adults and adolescents (≤age 21 at diagnosis/GT) are shown in Figure 1. The average score on the SDM-Q-9, reflecting exercise decision-making at diagnosis/GT was 59.64±25.0. Scores ranged from no SDM (SDM-Q-9=0) to perfect SDM (SDM-Q-9=100). Generally, participants reported high SDM on items related to exchange of information (ie, my provider made it clear that a decision needed to be made or my provider helped me understand all of the information) and lower scores on items that reflected partnering or considering participant opinion (ie, my provider asked me which option I prefer or my provider and I selected an option together). SDM-Q-9 mean item scores are presented in Table S2. Table 3 summarizes the association of extent of SDM regarding exercise with demographic, clinical, and exercise/athlete characteristics.

Younger age at diagnosis was associated with higher levels of SDM. The association of younger age at diagnosis with more SDM was evident both when comparing SDM in adolescent (diagnosis or GT ≤age 21) versus adult patients (diagnosis or GT >21 years; difference in means, −12.8, P=0.013 [95% CI, −22.8 to −2.9]) and when modeling age linearly (ß=−0.42, P<0.001 [95% CI, −0.65 to −0.18]). The relationship between SDM and being diagnosed or tested during adolescence as compared with adulthood strengthened when the age category was instead defined as diagnosis or GT at 18 or younger (difference in means, −16.4, P=0.007 [95% CI, −28.2 to −4.6]). Notably, time since diagnosis was

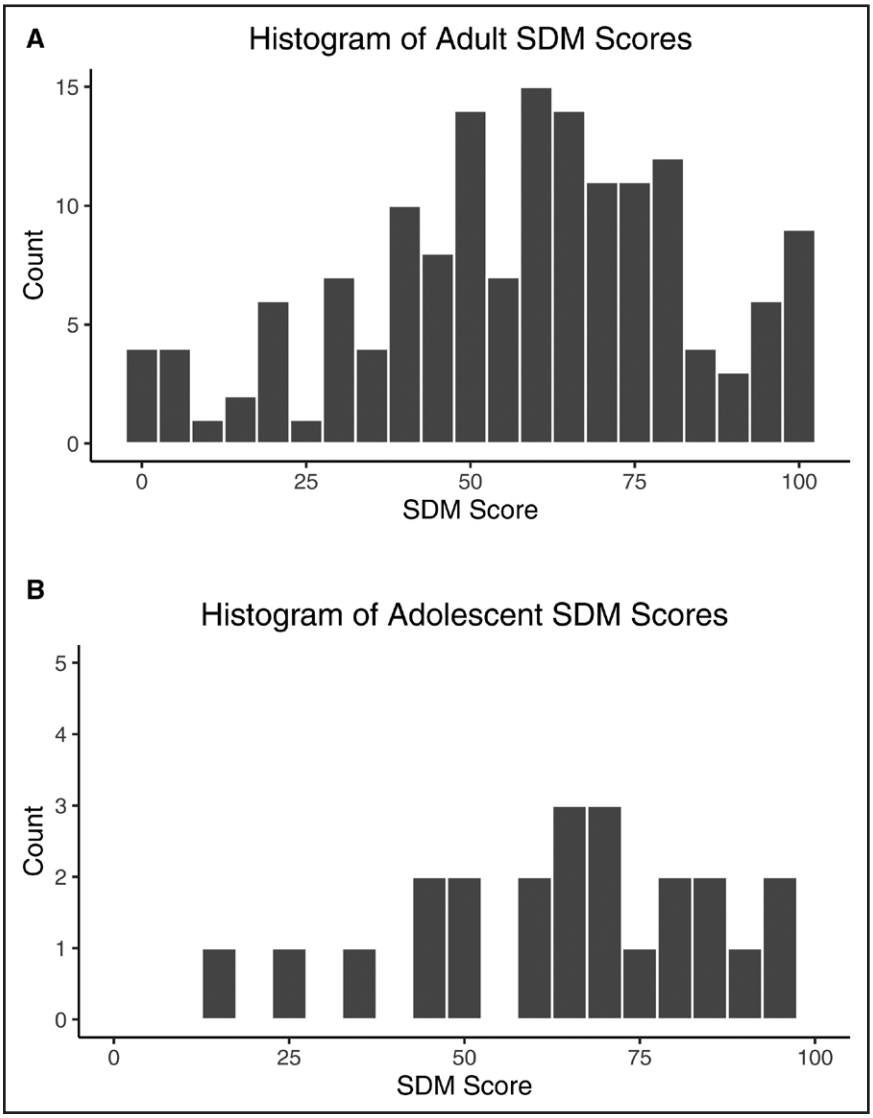

Figure 1. Histograms of shared decision-making scale score distribution.
A, Histogram of adult shared decision-making questionnaire (SDM-Q-9) scores (those with diagnosis/genetic testing at age 22 or later); (**B**) histogram of adolescent SDM-Q-9 scores (those diagnosed at age 21 or earlier).

Table 3. Summary of SDM Scores

	N	Mean	Difference in means	95% CI	*P* value
Gender					
Male	85	61.2	−3.0	(−4.34 to 10.32)	0.422
Female	95	58.2			
Age categories					
Diagnosed 21 or younger	28	70.6	−12.8	(−22.8 to −2.9)	0.013
Diagnosed 22 or older	153	57.8			
ARVC status					
Diagnosed with ARVC	141	59.6	−0.9	(0.84 to 17.22)	0.841
Genetically at risk for ARVC	41	58.7			
Exercise history					
Had played a competitive sport	142	60.4	−3.5	(−12.42 to 5.32)	0.431
Had never played a competitive sport	40	56.9			
Viewed self as athlete in the 6 mo before diagnosis	128	61.2	−5.0	(13.02 to 3.00)	0.219
Did not view self as athlete in the 6 mo before diagnosis	54	56.2			
Viewed self as active in the 6 mo before diagnosis	170	59.3	2.1	(−13.33 to 17.52)	0.789
Did not view self as active in the 6 mo before diagnosis	11	61.4			
Vigorous activity in the 6 mo before diagnosis	123	60.9	−4.0	(−11.83 to 3.73)	0.305
No vigorous activity in the 6 mo before diagnosis	60	56.9			
Clinical history					
ICD at last follow-up	104	58.8	1.3	(−6.23 to 8.80)	0.736
No ICD at last follow-up	—	60.0			
Had sustained VT at presentation	53	55.8	9.0	(0.84 to 17.22)	0.031
No sustained VT at presentation	76	64.8			
Genotype					
No variant	34	67.2			0.036
PKP2 variant	77	61.6	−5.6	(−4.34 to 15.5)	
DSP variant	30	51.3	−15.9	(−28.0 to −3.7)	
Other variant	35	54.2	−13.0	(−24.7 to −1.3)	

ARVC indicates arrhythmogenic right ventricular cardiomyopathy; SDM, shared decision-making; and VT, ventricular arrhythmia.

not associated with extent of SDM (ß=−0.62±0.67, *P*=0.352 [95% CI, −0.72 to 1.96]). When comparing adults with a clinical diagnosis to adolescents with a clinical diagnosis (excluding those with genetic risk only), the trend of adolescents reporting more SDM than adults was maintained but at a level that was not statistically significant (difference in means, −8.2, *P*=0.18 [95% CI, −3.9 to 20.4]). Overall, there was no difference in SDM in clinically diagnosed versus genetically at-risk relatives (Table S3).

Genotype had a limited association with SDM (*P*=0.036 across groups), with those with *DSP* variants tending to have lower levels of SDM (shown in Figure S3). When *PKP2* variants, *DSP* variants, and other variants were added to a linear regression using gene elusive participants as the reference category, only *DSP* variants and other variants had a significant association with SDM ($ß_{DSP}$=−15.856, *P*=0.011 [95% CI, −28.039 to −3.673]; $ß_{other\ variant}$=−12.967, *P*=0.030 [95% CI, −24.679 to −1.256]).

In contrast, athletic history, participation, and identity were not associated with extent of SDM. There was a slight trend in most exercise history categories toward those who were more active or athletic reporting more SDM, but it was insignificant for every variable analyzed. Likewise, clinical and demographic variables were largely not associated with SDM. The exception to this was seen among patients who had experienced a sustained ventricular arrhythmia before or at the time of diagnosis. This clinical presentation was associated with significantly less SDM (difference in means, −9.03, *P*=0.013 [95% CI, 0.840–17.22]).

When age at diagnosis, whether the participant presented with a sustained ventricular arrhythmia and genotype were added to a multivariable linear model, age, having a *DSP* variant, or having a variant in the other category were significantly associated with SDM ($ß_{age}$=−0.333, *P*=0.009 [95% CI, −0.580 to −0.086]; $ß_{DSP}$=−15.696, *P*=0.011 [95% CI, −27.712 to −3.680]; $ß_{other\ variant}$=−12.199, *P*=0.043 [95% CI, −23.985 to −0.414]). Having a sustained ventricular arrhythmia at

diagnosis and having a *PKP2* variant were not significantly associated with SDM in this model ($ß_{VT\ at\ pres}$=−0.007, *P*=0.098 [95% CI, −0.016 to 0.001]; $ß_{PKP2}$=−6.495, *P*=0.210 [95% CI, −16.694 to 3.704]).

Decisional Conflict and Decisional Regret

Overall, the population had significant levels of decisional conflict and decisional regret regarding exercise decision-making. Two-thirds (68.0%, n=121) of participants reported experiencing clinically significant decisional conflict in the 6 months following diagnosis or GT. In the year before study completion, 55.1% (n=98) of participants were experiencing clinically significant decisional conflict. Similarly, while 16.8% (n=30) of participants experienced no decisional regret, 27.9% (n=50) experienced mild decisional regret, and 55.3% (n=99) experienced moderate to severe decisional regret with regard to the decisions they made about exercise in the 6 months after diagnosis. The population levels of SDM-Q-9, DCS (decisional conflict scale), and DRS (decisional regret scale) scores are summarized in Table 4. Decisional conflict subscale summary data are presented in Table S1.

Association of SDM with Decisional Conflict and Decisional Regret

As shown in Figure 2, SDM had significant, negative linear relationships with both decisional conflict (both in the 6 months after diagnosis and currently) and decisional regret. In other words, a higher SDM-Q-9 score (more SDM) was associated with lower DCS and DRS scores. SDM-Q-9 scores at diagnosis or GT had the strongest association with DCS scores in the 6 months after diagnosis or GT (Figure 2A; ß=−0.66, R^2=0.567, *P*<0.001 [95% CI, −0.75 to −0.58]). The association between SDM-Q-9 and DCS scores in the year before study completion was weaker but maintained the same direction of the effect (Figure 2B; ß=−0.41, R^2=0.247, *P*<0.001 [95% CI, −0.49 to −0.26]). SDM-Q-9 score was significantly, yet more weakly associated with DRS score (Figure 2C; ß=−0.37, R^2=0.180, *P*<0.001 [95% CI, −0.52 to −0.30]). DRS scores were more strongly associated with DCS scores in the 6 months after diagnosis, with higher DCS scores associated with higher DRS scores (Figure 2D; ß=0.64, R^2=0.397, *P*<0.001 [95% CI, −0.52 to −0.75]). This showed that those who had higher decisional conflict in the 6 months after they were diagnosed or tested tended to have higher decisional regret regarding the decisions they made about exercise during that time. The direction of these relationships was maintained when the data was stratified into those with diagnosis or GT at age 21 or younger and those with diagnosis or GT at age 22 and older (see Figures S1 and S2).

Table 4. SDM-Q-9, DCS, and DRS Summary*

	N	Mean±SD median (IQR) or %
Shared decision-making (at the time of diagnosis/GT; mean±SD)	183	59.6±24.3
Decisional conflict in the 6 mo after diagnosis /GT		
DCS whole scale score (mean±SD)	178	34.3±22.7
Proportion with clinically significant decisional conflict (DCS≥25) (%)	121	68.0
Decisional conflict in the year before study completion		
DCS whole scale score (mean±SD)	178	27.3±21.3
Proportion with clinically significant decisional conflict (DCS≥25) (%)	98	55.1
Decisional regret in the 6 mo after diagnosis/GT		
Decisional regret whole scale score (median [IQR])	179	25 (35)
Proportion with no decisional regret (DRS=0) (%)	30	16.8
Proportion with mild decisional regret (0<DRS<25) (%)	50	27.9
Proportion with moderate to severe decisional regret (DRS≥25) (%)	99	55.3

DCS indicates decisional conflict scale; DRS, decisional regret scale; GT, genetic testing; IQR, interquartile range; and SDM, shared decision-making.

*Some items were left blank by some participants. Percentages reflect the proportion of those who answered the items (ie, those who did not answer were excluded).

SDM and Adherence to Exercise Guidelines

SDM did not seem to be associated with adherence to exercise guidelines. Participants who engaged in any vigorous activity did not have significantly different SDM-Q-9 scores than those who did not participate in vigorous activity in the 6 months after diagnosis (mean $SDM_{no\ vigorous\ activity}$=59.90±25.57, mean $SDM_{vigorous\ activity}$=56.11±18.97, *P*=0.56]) or in year before study completion (mean $SDM_{no\ vigorous\ activity}$=59.08±25.13, mean $SDM_{vigorous\ activity}$=65.98±23.85, *P*=0.34]).

DISCUSSION

In this study, we characterized decision-making for exercise among people with ARVC and at-risk relatives with the goals of evaluating the extent and implications of SDM for the decision made, decisional conflict, and decisional regret. We found that participants report a highly variable extent of SDM for exercise, with younger participants more likely to report having engaged in SDM. While participants reported decreasing exercise significantly after diagnosis, they expressed high levels of decisional conflict and decisional regret with respect to making a decision about how much to exercise. Importantly, SDM was associated with less decisional conflict and decisional regret. Adherence to exercise guidelines was high regardless of extent of SDM. Our findings therefore suggest that an SDM approach to exercise decision-making

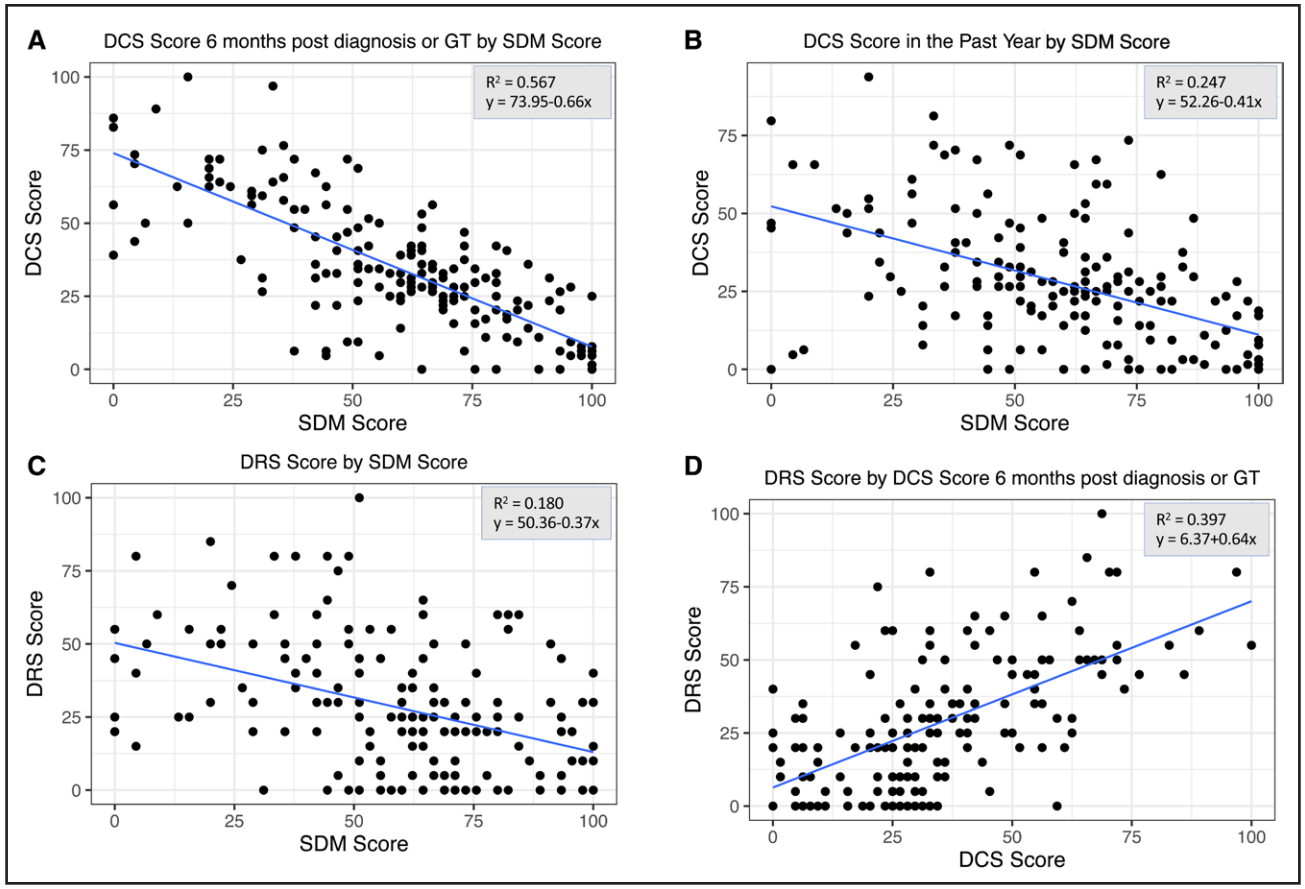

Figure 2. Association of shared decision-making (SDM-Q-9 score) with decisional conflict scale (DCS) and decisional regret scale (DRS) scores.

A, Scatterplot of DCS score at 6 mo after diagnosis/genetic testing associated with SDM-Q-9 scores. **B**, DCS score in the year before study completion associated with SDM score. **C**, DRS score associated with SDM score. **D**, DRS score associated with DCS score 6 mo after diagnosis/genetic testing.

will likely benefit patients with ARVC and possibly others with or at risk for inherited heart diseases who must make choices about exercise because of disease-related recommendations.

SDM is recommended in guidelines for exercise decision-making for ARVC because of its known associations with positive outcomes of decision-making, such as decreased decisional conflict and decisional regret.[6] While SDM is effective and preferable in theory, with regard to exercise decision-making for those with ARVC, it is complicated because the decision is ongoing throughout the lifespan, adverse outcomes can be life-threatening, and there has been little study surrounding its efficacy and implementation. We found that SDM is happening to some extent but with high variability. Participants reported anywhere from no SDM to perfect SDM regarding exercise. Generally, participants reported high SDM on items related to the exchange of information and lower scores on items that reflected partnering or considering patient opinion. This suggests that providers may, in general, sufficiently educate their patients on the risks and benefits of exercise with ARVC, but not specifically make space for patients to share their values and preferences or work through what might be the best decision for them.

Additionally, we found that SDM is not happening at the same level for everyone. Most demographic and clinical variables were unrelated to extent of SDM reported. However, a few variables did have significant associations with SDM. Unsurprisingly, having a sustained ventricular arrhythmia at presentation was associated with significantly less SDM. While the reason for this association is uncertain, one could speculate that both the higher risk for recurrent ventricular arrhythmia and the emergent presentation could play a role. Genotype was also associated with SDM, with gene elusive patients reporting the most SDM and those with *DSP* or other variants (including *DSG2*, *DCS2*, *TMEM43*, *PLN*, *LMNA*, *TTN*, and *FLNC*) significantly less. In multivariable analysis, older age and having a *DSP* or "other" variant were independently associated with less SDM. While the reason for this association with genotype was not explored, it may reflect the relative strength of the evidence for the association of exercise with outcomes in gene elusive

and *PKP2* ARVC relative to other genotypes. More unexpectedly, we found that those who were diagnosed in childhood, adolescence, or young adulthood reported significantly more SDM than those diagnosed at older ages. While more research is necessary to determine why this is the case, there are a few possible explanations. First, it is possible that adult cardiologists practice differently than pediatric cardiologists. Furthermore, we know that provider preferences and lifestyle impact the exercise recommendations they make, and that provider gender and cultural background are associated with communication style.[28–30] Another possible explanation is that, while the SDM-Q-9 addresses specifically the decision happening between a patient and provider, participants were reflecting on their decision-making process as a whole, including others who may have been involved in the process. Children and adolescents often make medical decisions with involvement of their parents or other family members, so it is possible that they experienced more robust SDM and more support from their families that was reflected in their SDM-Q-9 scores. Notably, athletes reported similar SDM-Q-9 scores to nonathletes. This was surprising because those who are particularly athletic are often considered more likely to be nonadherent with exercise guidelines, therefore we hypothesized they may be less likely to be engaged in SDM.[7]

Perhaps most impactfully, we found that higher levels of SDM were associated with lower decisional conflict and decisional regret. This is important because DCS and DRS scores were relatively high and both have been associated with poor psychosocial and medical outcomes.

While SDM was associated with lower decisional conflict and decisional regret, it was not associated with adherence to exercise guidelines. This suggests that those patients who were engaged in SDM were not more likely to disregard exercise guidelines, at least in this population. This is in line with the existing literature on SDM and adherence, which has overwhelmingly linked SDM to either increased adherence or found no difference in adherence based on SDM, depending on the population.[23,25,26,31–33] This finding is significant because some clinicians refute the utility of SDM in exercise decision-making for those with inherited heart disease, arguing that it could lead patients to exercise against recommendations.[15] With all of this in mind, it is clear that decisional conflict and decisional regret are significant problems in this population and that following an SDM model is associated with less decisional conflict and decisional regret without being associated with less adherence to guidelines.

Clinical Implications

While SDM for exercise decision-making has been recommended for inherited heart disease, data has been unavailable on the efficacy of SDM for this complex and ongoing decision. The results of this study suggest that SDM may be the preferable model of decision-making for people with ARVC who are considering exercise modifications. Importantly, this study provides evidence that indeed SDM is associated with more positive decisional outcomes for patients with ARVC and at-risk relatives without being associated with less adherence to exercise guidelines. These findings have implications for the care of ARVC families and possibly more broadly for discussions of exercise in inherited heart disease clinics. Specifically, based on our findings, it seems likely that SDM for exercise will benefit patients with ARVC and families by reducing decisional conflict and decisional regret. Importantly, we saw no evidence high SDM was associated with poorer adherence to guidelines related to avoiding competitive sports or frequent vigorous aerobic exercise. It is also worth noting that multidisciplinary heart disease clinics are well placed to engage in SDM for exercise. Cardiology providers are familiar with and capable of implementing SDM. For example, the decision to implant an ICD often follows an SDM model.[34–36] In summary, exercise decision-making for those with ARVC is a lifelong discussion. This data does not suggest abdicating professional responsibility to advise patients but rather highlights that including patient voices in the discussion around exercise might lead to better long-term outcomes.

Limitations

It should be acknowledged that the cross-sectional nature of the study prevents us from establishing directionality of the relationships discussed. The population of this study was recruited through the Johns Hopkins ARVC registry, which may not be representative of all people with ARVC. The retrospective nature of the study introduces limitations on the ability of participants to accurately recall their experiences of exercise decision-making around the time they were diagnosed. Future studies could explore exercise decision-making using a prospective approach to reduce this bias. We acknowledge that our data are a limited representation of the nuanced exercise histories of these individuals. Our population reported high exercise guideline adherence (almost no participants reported engaging in vigorous aerobic activity after diagnosis), which limited our ability to analyze the effect of SDM on adherence. Additionally, decisional conflict and decisional regret are concepts that can represent a broad range of experiences, and we did not measure their nuances in this population.

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Supplemental Material

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