



OPEN Nature-based interventions for individuals with rare skeletal disorders: evaluation of a 5-day sailing program on health-related quality of life

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Individuals with rare skeletal disorders like Multiple Osteochondromas and Ollier Disease often experience physical and psychological burdens. Adventure therapy, with activities like sailing in outdoor settings, promotes personal growth and psychological well-being, potentially improving health-related quality of life (HRQoL). This study aimed to evaluate the impact of a sailing program on health-related quality of life and participant satisfaction in individuals with Multiple Osteochondromas and Ollier Disease. A quasi-experimental one-group pre-post design was employed, with HRQoL assessed using the EQ-5D[®] instrument and participant satisfaction measured via a feedback survey. Data were collected before and after the five-day sailing program conducted in the Mediterranean Sea in 2022 and 2023, involving participants diagnosed with Multiple Osteochondromas and Ollier Disease. Statistical analyses were performed using the Wilcoxon signed-rank test and McNemar's test for paired data. A significance level of $p < 0.05$ and $p < 0.10$ was considered. A total of 25 participants, predominantly male (52%), with a median age of 16 years (ranking from 11 to 31), were included in the study. The sailing program had mixed impact on HRQoL. Specifically, individuals who were female ($p = 0.03$), aged 16 and older ($p = 0.04$), with higher educational attainment ($p = 0.10$) or stronger self-management ($p = 0.09$), resilience ($p = 0.01$) and self-engagement ($p = 0.09$) skills experienced enhanced HRQoL. Conversely, other participants exhibited an increase in self-care difficulties ($p = 0.02$) and a trend towards worsening pain/discomfort ($p = 0.38$). Overall satisfaction with the program was high, with 90% of participants expressing satisfaction.

This is the first study which examined HRQoL in Multiple Osteochondromas and Ollier Disease patients within an outdoor adventure therapy setting. Findings suggest that adventure therapy, integrated into healthcare strategies, may offer a valuable complement to conventional treatments for rare skeletal disorders. Future research, including randomized controlled trials, are necessary to confirm these results and develop robust interventions for improving the well-being in this population.

Keywords Adventure therapy, Sailing, Health-Related Quality of Life, Rare skeletal disorders, Natural environments, Outdoor

Individuals with rare diseases (RDs) face a set of psychological challenges, encompassing their own experiences to permeate their families and communities^{1,2}. The chronic, often severe, and frequently poorly understood nature of RDs results in a wide range of emotional, social, and psychological issues that can have profound and lasting effects on individuals' well-being³.

Isolation is a common experience for those with RDs, as they often lack peers and support systems that can relate to their experiences. Feelings of loneliness and stigmatization can arise as a result^{4,5}. The uncertainty surrounding RDs, including diagnosis, prognosis, and treatment options, can lead to significant anxiety⁶. The chronic nature of many RDs, along with the limitations they impose, can contribute to depression and a sense of

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helplessness and hopelessness. Moreover, RDs disrupt the lives of individuals, impacting their education, social activities, and prospects. For instance, educational hurdles may arise such as absenteeism and discrimination, leading to isolation and low self-esteem^{7,8}.

Multiple Osteochondromas (MO, MIM #133700 and #133701) and Ollier Disease (OD, MIM #166000) are rare, lifelong, genetically skeletal disorders with onset at birth. These conditions are distinguished by the presence of osteochondral outgrowths termed osteochondromas in MO and multiple benign intramedullary cartilage lesions known as enchondromas in OD. The number and size of these lesions escalate throughout childhood, plateauing post-closure of the growth plate^{9–11}. Common clinical symptoms include bone deformities, restricted motion, and pain. Malignant transformation into secondary peripheral chondrosarcoma is a significant concern, with 2–5% risk for MO and 10–40% for OD. Furthermore, surgical intervention is the main treatment of choice^{10–14}.

Due to the complications associated with these disorders, previous studies have demonstrated the pervasive impact of MO and OD on individuals' physical, psychological, social, and personal growth and well-being^{15–18}.

Involving individuals in their healthcare, including shared decision-making and developing healthy coping mechanisms, is crucial in adapting to psychological concerns and restoring their sense of control.

Adventure therapy (AT) is a nontraditional approach that harnesses the transformative power of outdoor experiences to promote personal growth, enhance psychological well-being, and facilitate resilience in a wide range of populations and conditions^{19–24}. AT can engage participants physically, emotionally, and mentally through carefully designed activities, such as rock climbing, hiking, rafting, and sailing^{25–28}. These activities allow participants to learn how to challenge their perceived limitations and develop valuable problem-solving and decision-making skills^{29,30}. This approach encourages individuals to step out of their comfort zones, fostering personal growth and self-discovery^{31–39}.

Sailing as a context for promoting self-development for therapeutic or rehabilitative interventions for marginalized groups, people with disabilities, or mental health problems has been recently reviewed⁴⁰. The Nave Italia project (<https://www.naveitalia.org/en>) distinguishes itself among adventure programs by seamlessly blending AT principles with a distinctive marine ambiance to offer individuals remarkable avenues for healing and personal growth⁴¹. Launched in 2007 by the Tender To Nave Italia Foundation (TTNIF), this project aims to craft an immersive and therapeutic experience for individuals grappling with cognitive impairments, sensory limitations, genetic conditions, such as MO and OD, and psychosocial challenges, employing AT principles to address these concerns^{42,43}.

The participants in this transformative endeavor live on board a uniquely configured brig, which provides a safe and supportive environment. The Italian Navy crew, the TTNIF staff, and the participants form a cohesive unit, cohabiting on board. The project aims to enable individuals to overcome personal challenges, develop coping strategies, and foster a sense of empowerment, solidarity, and community through a range of sailing activities conducted during the voyage^{44–48}. Furthermore, the marine environment inspires a sense of wonder, calmness, and connection with nature, making it an ideal setting for personal reflection, emotional healing, and self-discovery²⁵.

The primary aim of this study is to evaluate changes in health-related quality of life (HRQoL) in individuals affected by MO and OD through their participation in the five-day sailing program within the Nave Italia project. The secondary aim is to assess participants' satisfaction with the five-day sailing program. We hypothesized that the program would have a positive impact on both outcomes.

Materials and methods

Study design

A quasi-experimental one-group pre-post study design was employed to appraise the HRQoL of individuals with MO and OD participating in the Nave Italia project⁴⁹. Data were collected at two time points: before the start of the five-day sailing program (T_0) and after the sailing program (T_1). Thereafter, it was recorded in Registry of Multiple Osteochondromas and Registry of Ollier Disease and Maffucci Syndrome, two standardized disease registries that store clinical, genetic, and family history, as well as quality of life data from patients with MO or OD located at IRCCS Istituto Ortopedico Rizzoli. The registries are web based and in compliance with national and European privacy regulations as well as medical informatics standards.

This process was managed by a clinical psychology research assistant with expertise in rare skeletal disorders affiliated to Rare Skeletal Disorders Department at IRCCS Istituto Ortopedico Rizzoli.

The five-day sailing program was conducted in the Mediterranean Sea onboard the TTNIF schooner brig for two editions: June 21–25, 2022, and July 25–29, 2023. The current study presents the findings from both editions of the program.

Ethical statement

This research was part of two ongoing studies approved by the Local Ethical Committee: Registry of Multiple Osteochondromas (protocol numbers 0021283/2013, NCT04133285) and the Registry of Ollier Disease and Maffucci Syndrome (protocol numbers 0022938/2016, NCT04134572)^{50,51}, and was conducted in accordance with the 1964 Declaration of Helsinki.

The written informed consent was obtained from all participants aged ≥ 18 years and from parents/guardians of those participants aged < 18 years before starting the sailing program.

Sailing program: Nave Italia project

TTNIF conducts an annual call for proposals, inviting non-profit organizations, schools, or institutions to submit project proposals. Selected projects gain access to Nave Italia brig as a setting for their educational activities, with the support of TTNIF staff (Supplementary Figure S1).

The Nave Italia, a 61-meter-long steel schooner brig with a masthead height of 44.60 m and a crew of Italian Navy officers and 21 personnel and boasts 11 cabins that accommodate up to 25 guests (Fig. 1).

The activities unfold during a five-day sailing voyage, seamlessly blending the routine of nautical life with dedicated onboard laboratories.

The Italian Association for MO and OD patients (Associazione Conto Alla Rovescia – ACAR Aps, <https://www.acar-aps.org/>) in collaboration with Rare Skeletal Disorders Department of IRCSS Istituto Ortopedico Rizzoli embarked on two engaging projects in 2022 and 2023 that promoted both environmental consciousness and artistic expression. The 2022 project, named “*Temerari si nasce*” (Born to brave), empowered individuals to transform discarded plastic waste into creatively produced sea turtles. The 2023 project, entitled “*Vela d’A-mare*” (Sail for love), fostered the collaborative choral painting (Supplementary Figure S2).

These projects, combined with sailing activities, promoted autonomy, increased solidarity among participants, and improved HRQoL. Each individual was assigned specific tasks and learned the principles of sailing, including hauling sails, steering the craft, and making nautical knots. Using safety ropes, even patients with limited mobility were able to climb the mast. Participants were responsible for several daily maintenance activities, such as organizing their berths, cleaning the bathrooms, and assisting with meal preparation (Fig. 2).

Participants

The participants comprised individuals affiliated with ACAR. To be eligible, subjects had to meet the following criteria: (a) age ≥ 10 years; (b) a clinical diagnosis of MO or OD; (c) no surgery within the preceding month prior to boarding; and (d) enrollment in Registry of Multiple Osteochondromas or Registry of Ollier Disease and Maffucci Syndrome and provision of informed consent. Exclusion criteria included: (a) age < 10 years; (b) diagnosis of other distinct, rare skeletal disease; (c) undergoing evaluation for differential diagnosis; (d) presence of medical conditions incompatible with the marine environment and sailing activities; (e) healthy individuals; and (f) declination to provide informed consent.

Furthermore, brigantine guest capacity has been reduced to 13 per boarding, in accordance with Italian Navy’s anti-COVID-19 regulations implemented in 2022 and 2023.



Fig. 1. Nave Italia schooner brig.

TENDER TO NAVE ITALIA

PROGRAM ACTIVITY SCHEDULE

Five-day activity schedule	Tuesday	Wednesday	Thursday	Friday	Saturday
Morning activities (1st slot)	COVID-19 testing and reception	Hauling sails	General ship cleaning	Dedicated project laboratory	Farewell ceremony and disembarkation
Afternoon activities (2nd slot)	Boarding, cabin assignment, and handing out daily maintenance activities	Dedicated project laboratory	Dedicated project laboratory	T-shirt creation laboratory	
Afternoon activities (3rd slot)	Crew introduction and onboard safety training	Nautical knots laboratory	Climbing the mast	End-of-experience reflection group	
Evening activities	Games (organized by participants)	Music game	Stargazing workshop	Closing party	

Fig. 2. Five-day sailing program activity schedule.

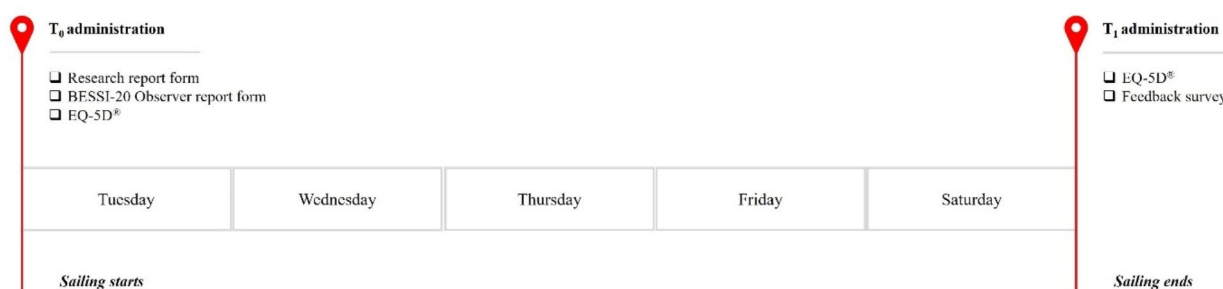


Fig. 3. Data collection timeline.

Measures

Socio-demographic and clinical characteristics

The socio-demographic and clinical characteristics considered were sex at birth, age at boarding (in years), education level, employment status, and diagnosis of MO or OD. This data was collected at T_0 (Fig. 3) using a research report form (Supplementary File 1).

Behavioral, emotional, and social skills

The behavioral, emotional, and social skills of each participant were assessed at T_0 using the validated Italian version of The Behavioral, Emotional, and Social Skills Inventory – Observer report form (BESSI-20)⁵² (Fig. 3).

The BESSI-20 Inventory is an observer report tool that evaluates five key skill domains: Self-Management (SM), Social Engagement (SE), Cooperation (CO), Emotional Resilience (ER), and Innovation (IN). Each item details a specific behavior pertinent to its corresponding domain, and individuals evaluate their ability to exhibit that behavior using a 5-point Likert scale ranging from 1 (Not at all well) to 5 (Extremely well). Scores for each domain range from 4 to 20, with higher scores indicating higher competence in the assessed skill⁵³.

The BESSI is a useful tool for professionals in various disciplines, including education, psychology, counseling, and social work, enabling them to assess social, emotional, and behavioral skills. It helps in identifying specific areas for intervention, personal development, and skill enhancement⁵⁴.

Health-related quality of life

HRQoL was assessed using the validated Italian version of the EQ-5D tools: (a) EQ-5D-Y for children < 16 years, and (b) EQ-5D-5 L for adults \geq 16 years^{55,56}.

The EQ-5D is a brief, generic instrument for measuring health-related quality of life and consists of two parts: A self-classifier and a visual analogue scale (EQ-VAS)^{57,58}.

The self-classifier consists of five items that assess problems in the following dimensions: mobility, self-care, usual activities, pain/discomfort, and anxiety/depression. Each dimension has 5-level options for adults and 3-level options for children. All individuals generate a unique health profile based on their responses, which ranges from the highest (11111) to lowest level of health for adults (55555) or children (33333).

An Index Value (IV), a summary score, was assigned to each health profile based on country-specific preferences. For adult populations, the Italian EQ-5D-5 L value set was employed. However, in accordance with EuroQol guidelines, the Spanish EQ-5D-Y value set was applied for pediatric populations due to the absence of the Italian pediatric value set^{59–61}. The IV ranges from 1 (perfect health) to 0 (absence of life/death). Values less than zero reflect health states “worse than death”. In this study, for all five dimensions, the levels ≥ 2 were grouped and thus dichotomized to “no problem” or “any problem”, according to EuroQol Group guidelines⁵⁹.

The EQ-VAS is a self-report measure of overall health status on a scale from 0 (“The worst health you can imagine”) to 100 (“The best health you can imagine”).

The IV and EQ-VAS scores served as the primary outcomes for this study. EQ-5D questionnaires were administered to participants at T_0 and T_1 to evaluate the impact of sailing program on HRQoL. See Fig. 3.

Participants satisfaction

At T_1 a feedback survey developed by the authors was administered to evaluate the participants’ satisfaction with the project (Fig. 3). The survey consisted of five items and statements were scored on a five-point Likert scale that generated categorical data ranging from 0 (very dissatisfied) to 4 (very satisfied), with higher scores reflecting higher levels of satisfaction (Supplementary File 2).

Statistical analysis

Continuous data were presented as mean, standard deviation, median, minimum and maximum, whereas categorical and ordinal data were described using numbers and percentages. The EQ-5D questionnaires and BESSI-20 scores were computed according to the developers’ guidelines^{52,53,58}.

Continuous outcome scores before and after the sailing program were compared using the Wilcoxon signed-rank test for paired data. Binary outcomes at the two time points were compared using the exact McNemar test for paired data. Stratified analyses were performed according to sex (male/female), year (2022/2023), high and low BESSI-20 five domain scores, age, and education level. The observed medians were used to determine the high (\geq median) and low ($<$ median) scores.

Satisfaction levels were measured using ordinal variables on a Likert scale, and comparisons between participant responses were conducted using single or pairwise Wilcoxon signed-rank tests to assess differences when stratifying by age, sex, and education level.

A significance level of $p < 0.05$ and $p < 0.10$ was considered. Statistical analyses were performed using R 3.6.3 software (The R Foundation for Statistical Computing, Wien).

Results

Participants characteristics

A cohort of 25 individuals participated in the five-day sailing program, twelve in 2022 and thirteen in 2023; 52% were male. The median age was 16 years ($SD \pm 6$; ranking from 11 to 31), with a predominant manifestation of MO (92%) over OD (8%). The majority of participants reported current student status (76%), with a significant proportion (40%) attending upper secondary school.

Overall, the BESSI-20 revealed weaknesses in the ER domain, with the lowest mean score of 12 ($SD \pm 3.6$; ranking from 5 to 17). Conversely, the IN skills emerged as the leading domain, with a mean score of 16 ($SD \pm 4.0$; ranking from 7 to 20). The SM and CO domains showed similar incidence, while SE domain score was closely linked with the ER domain.

All the descriptive statistics are reported in Table 1.

Five-day sailing program impact on HRQoL

Table 2 summarizes the participants’ HRQoL scores at two designated time points and the change resulting from the five-day sailing program.

Overall, 24 out of 25 participants reported difficulties in at least one dimension of the EQ-5D instrument before the sailing program. Pain/discomfort was the most common and distressing concern, affecting 84% of the cohort. Anxiety/depression and difficulties with usual activities displayed similar prevalence (63%), while mobility issues were reported by 56% of participants. In contrast, self-care challenges were not perceived as a substantial issue for the overwhelming majority (96%) of individuals. The EQ-5D IV score had an average of 0.81 ($SD \pm 0.10$), with values ranking from 0.65 to 1. The mean EQ-VAS score was 76 ($SD \pm 12$), ranking from 40 to 92.

As a result of sailing program the assessment revealed an increased number of participants reporting difficulties in the self-care (4 vs. 32%, $p = 0.02$) and, although not statistically significant, a worsening in pain/discomfort dimension (84 vs. 96%, $p = 0.38$). Particularly, individuals with lower SM, CO, SE, and ER as well as male sex, reported a deterioration in the self-care dimension ($p = 0.03$, $p = 0.06$, $p = 0.03$, $p = 0.03$, and $p = 0.06$, respectively) after the program. No significant changes were observed in the remaining domains. All findings are listed in Supplementary Table 1.

No significant change has been observed in the IV and VAS scores for the aggregate dataset ($p = 0.47$ and $p = 0.47$, respectively). However, when stratifying by age, sex and skills the two scores showed evident differences.

Characteristics	Overall (N = 25)
Age (years)	
Mean (SD)	18 (\pm 6.0)
Median (min-max)	16 (11–31)
Sex, n (%)	
Male	13 (52.0%)
Female	12 (48.0%)
Diseases, n (%)	
Multiple Osteochondromas	23 (92.0%)
Ollier Disease	2 (8.0%)
Education level, n (%)	
Lower secondary education	8 (32.0%)
Upper secondary education	10 (40.0%)
Bachelor's or equivalent	7 (28.0%)
Employment status, n (%)	
Employed for wages	4 (16.0%)
Out of work and looking for work	2 (8.0%)
Student	19 (76.0%)
Year of attendance, n (%)	
2022	12 (48.0%)
2023	13 (52.0%)
BESSI-20 domains	
Self-Management	
Mean (SD)	15 (\pm 3.6)
Median (min-max)	15 (8–19)
Innovation	
Mean (SD)	16 (\pm 4.0)
Median (min-max)	17 (7–20)
Cooperation	
Mean (SD)	15 (\pm 3.4)
Median (min-max)	16 (16–20)
Social Engagement	
Mean (SD)	13 (\pm 3.7)
Median (min-max)	14 (5–19)
Emotional Resilience	
Mean (SD)	12 (\pm 3.6)
Median (min-max)	12 (5–17)
SD: standard deviation; min: minimum; max: maximum	

Table 1. Participants characteristics.

Specifically, participants with a lower SM, IN, CO, SE, and ER exhibited a significantly reduced IV score, as well as those with younger age ($p=0.04$, $p=0.05$, $p=0.04$, $p=0.01$, and $p=0.09$, respectively). Conversely, individuals with higher ER, SE, and SM displayed an increased IV score ($p=0.01$, $p=0.09$, and $p=0.09$, respectively).

As for VAS score, similar differences have been noticed in participants with age > 16 years, higher educational attainment, or female sex ($p=0.04$, $p=0.10$, and $p=0.03$, respectively). In addition, individuals with higher SM and ER showed an improved perception of overall health ($p=0.05$ and $p=0.03$, respectively).

The complete findings are presented in Table 3.

Despite the absence of statistical significance, Fig. 4 highlights a set of findings which may be of interest for larger-scale investigations pertaining to the improvement or deterioration of HRQoL. Specifically, participants with lower educational attainment, CO, or male sex were observed to relate to reduced IV scores. Additionally, male subjects experienced increased pain as a result of taking part of the sailing program. Furthermore, individuals with higher SE enhanced VAS scores. Finally, participants with higher ER improved the anxiety/depression dimension.

Participants' satisfaction

Figure 5 presents the comprehensive participants satisfaction ratings after the sailing program. Upon program completion, over 90% of individuals reported being “satisfied” or “very satisfied” across all investigated items.

HRQoL	Before sailing program (N = 25)	After sailing program (N = 25)	p-value
Mobility			
No problems	11 (44.0%)	11 (44.0%)	1
Any problems †	14 (56.0%)	14 (56.0%)	
Self-Care			
No problems	24 (96.0%)	17 (68.0%)	0.02**
Any problems	1 (4.0%)	8 (32.0%)	
Usual Activities			
No problems	9 (36.0%)	10 (40.0%)	1
Any problems	16 (64.0%)	15 (60.0%)	
Pain/Discomfort			
No problems	4 (16.0%)	1 (4.0%)	0.38
Any problems	21 (84.0%)	24 (96.0%)	
Anxiety/Depression			
No problems	9 (36.0%)	10 (40.0%)	1
Any problems	16 (64.0%)	15 (60.0%)	
Any Dimension			
No problems	1 (4.0%)	1 (4.0%)	1
Any problems	24 (96.0%)	24 (96.0%)	
EQ-5D Index Value			
Mean (SD)	0.82 (0.10)	0.78 (0.15)	0.47
Median (min-max)	0.81 (0.65-1)	0.86 (0.51-1)	
EQ-VAS			
Mean (SD)	76 (12)	77 (14)	0.47
Median (min-max)	76 (40-92)	78 (41-99)	

Table 2. Participants' HRQoL before and after sailing program. SD: standard deviation; Min: minimum; Max: maximum. ** $p < 0.05$; * $p < 0.10$ Dimensions data are expressed as n (%). †All levels ≥ 2 was collapsed.

However, a small subset (4%) expressed dissatisfaction with the fulfillment of their expectations and needs, while 4–8% remained neutral in their evaluation of expectations, sailing environment, daily activities, and companionship.

Specifically, participants younger than 16 years exhibited lower satisfaction scores compared to the oldest participants in terms of expectations, needs, and sail environment ($p = 0.09$, $p = 0.09$, $p = 0.09$, and $p = 0.01$, respectively). Moreover, individuals with lower secondary education referred reduced satisfaction with the sailing environment compared to their upper secondary counterparts ($p = 0.07$). Lastly, individuals with a bachelor's or equivalent reported greater satisfaction with daily activities than those in the upper secondary group ($p = 0.04$). Sex did not influence satisfaction ratings (Supplementary File 3).

Discussion

This is the first study to explore HRQoL among individuals with rare skeletal disorders in an outdoor setting, specifically through a structured 5-day sailing program based on AT principles. The findings suggest that female, aged 16 and older, with higher educational attainment or stronger self-management, resilience and self-engagement skills experienced enhanced HRQoL. The sailing program demonstrated potential benefit by strengthening self-management, social engagement, and emotional resilience skills in individuals with MO and OD. These results are consistent with previous reports highlighting the positive impact of similar interventions on chronic conditions^{25,28,30,32,35,37–39}.

The unique design of this study, integrating a sailing program in a natural Mediterranean environment, offers valuable insights away from conventional healthcare settings as outlined by Bowen and Neill (2013). The group-based delivery and the involvement of a dedicated and highly motivated multidisciplinary team likely contributed to its emotional impact and social benefits³⁶. The program's adventurous nature, combined with supportive environment and shared experiences with peers facilitated increased social support, reduced feeling of isolation, and fostered bonds promoting a sense of belonging and acceptance^{33–35}.

It is important to note that this study focused on a specific patient population aged 11–31 years who may have experienced psychosocial challenges such as loneliness, low self-esteem, physical limitations, and pain impacting daily activities and social interaction^{15–18}. Participation in sailing activities or similar outdoor settings with individuals facing analogous challenges might address these needs and offer opportunities for skill development tailored to individual independence within a safe environment^{25,35,40}. Nevertheless, randomized controlled trials are required before any causal conclusions can be drawn. Furthermore, adventurous activities offered opportunities for joy, excitement, and fulfillment, potentially improving HRQoL by providing a break from routine and promoting personal growth and enjoyment^{36–39}. These improvements could positively impact mental health, relationships, and overall well-being.

EQ-5D Scores	Stratification groups	Before sailing program				After sailing program				p-value
		Mean	SD	Min	Max	Mean	SD	Min	Max	
Index Value	Aggregate	0.81	0.10	0.65	1	0.78	0.15	0.51	1	0.47
	Age (< 16 years)	0.77	0.11	0.65	0.91	0.67	0.16	0.51	0.89	0.09*
	Age (≥ 16 years)	0.84	0.09	0.69	1	0.86	0.08	0.63	1	0.43
	Male	0.85	0.08	0.71	1	0.77	0.16	0.51	0.90	0.11
	Female	0.77	0.11	0.65	0.95	0.79	0.15	0.51	1	0.31
	2022	0.84	0.11	0.65	1	0.78	0.18	0.51	1	0.23
	2023	0.79	0.09	0.65	0.95	0.78	0.13	0.51	0.95	0.73
	Education level (< Secondary)	0.74	0.09	0.65	0.89	0.66	0.15	0.51	0.89	0.17
	Education level (≥ Secondary)	0.85	0.09	0.69	1	0.84	0.11	0.51	1	0.83
	SM ^a (< 15)	0.77	0.12	0.65	1	0.66	0.15	0.51	0.89	0.04**
	SM (≥ 15)	0.85	0.07	0.74	0.95	0.88	0.05	0.81	1	0.09*
	IN ^b (< 17)	0.79	0.14	0.65	1	0.70	0.15	0.51	0.89	0.05**
	IN (≥ 17)	0.83	0.08	0.69	0.95	0.82	0.14	0.51	1	0.47
	CO ^c (< 16)	0.81	0.12	0.65	1	0.71	0.17	0.51	0.89	0.05**
	CO (≥ 16)	0.82	0.09	0.69	0.95	0.84	0.11	0.63	1	0.15
	SE ^d (< 14)	0.79	0.13	0.65	1	0.68	0.16	0.51	0.89	0.04**
	SE (≥ 14)	0.83	0.08	0.71	0.95	0.86	0.08	0.65	1	0.09*
	ER ^e (< 12)	0.80	0.13	0.65	1	0.67	0.15	0.51	0.89	0.01**
	ER (≥ 12)	0.83	0.07	0.74	0.95	0.88	0.05	0.81	1	0.01**
EQ-VAS	Aggregate	76	12	40	92	77	14	41	99	0.47
	Age (< 16 years)	75	12	50	90	71	15	41	99	0.44
	Age (≥ 16 years)	77	12	40	92	81	12	45	95	0.04**
	Male	80	8	65	92	77	12	59	99	0.44
	Female	72	14	40	90	77	17	41	95	0.05**
	2022	74	15	40	90	76	14	45	99	0.50
	2023	77	8	65	92	78	14	41	95	0.67
	Education level (< Secondary)	74	13	50	90	71	17	41	99	0.57
	Education level (≥ Secondary)	77	12	40	92	80	12	45	95	0.10*
	SM (< 15)	71	16	40	90	69	16	41	99	0.53
	SM (≥ 15)	79	7	72	92	84	7	70	95	0.05**
	IN (< 17)	71	12	50	86	70	16	41	99	0.89
	IN (≥ 17)	78	12	40	92	80	12	45	95	0.24
	CO (< 16)	76	12	50	92	75	14	41	99	1
	CO (≥ 16)	76	12	40	90	79	14	45	95	0.21
	SE (< 14)	72	16	40	92	69	15	41	88	0.76
	SE (≥ 14)	79	6	72	90	83	11	59	99	0.15
	ER (< 12)	73	16	40	92	69	16	41	99	0.53
	ER (≥ 12)	79	6	72	90	84	7	70	95	0.03**

Table 3. Before and after sailing program changes in EQ-5D Index Value and VAS scores stratified by age, sex, year of attendance, education level, and BESSI-20. SD: standard deviation; Min: minimum; Max maximum. ** $p < 0.05$; * $p < 0.10$. ^aSM: Self-Management Skills Domain; ^bIN: Innovation Skills Domain; ^cCO: Cooperation Skills Domain; ^dSE: Social Engagement Skills Domain; ^eER: Emotional Resilience Skills Domain.

Considering these features, it is reasonable to suggest that for MO and OD individuals, the sailing program may represent a medium to reinforce the participants' personal and social skills and consequently increase their HRQoL. Notably, improvements in HRQoL were observed, with baseline differences in IV and VAS scores compared to the general healthy Italian population, this difference decreased slightly after the sailing experience⁶⁰. Regarding the five dimensions, our results showed an association within the self-care area, both in terms of increased and decreased evaluation by participants at the end of the sailing program. A possible reason for this ambivalence is that consistent with Brown's thinking, adventure therapy facilitates stepping out of one's comfort zone, promoting reflection and discussion, and pushing participants to explore their own experiences and boundaries³¹. Interestingly, there were manifestations between reduced anxiety/depression and higher scores in resilience skills from baseline to the end of the program. These findings suggest that a 5-day sailing program may be sufficient to improve the overall health status of MO and OD individuals, and potentially requires a longer duration to impact each specific dimension.

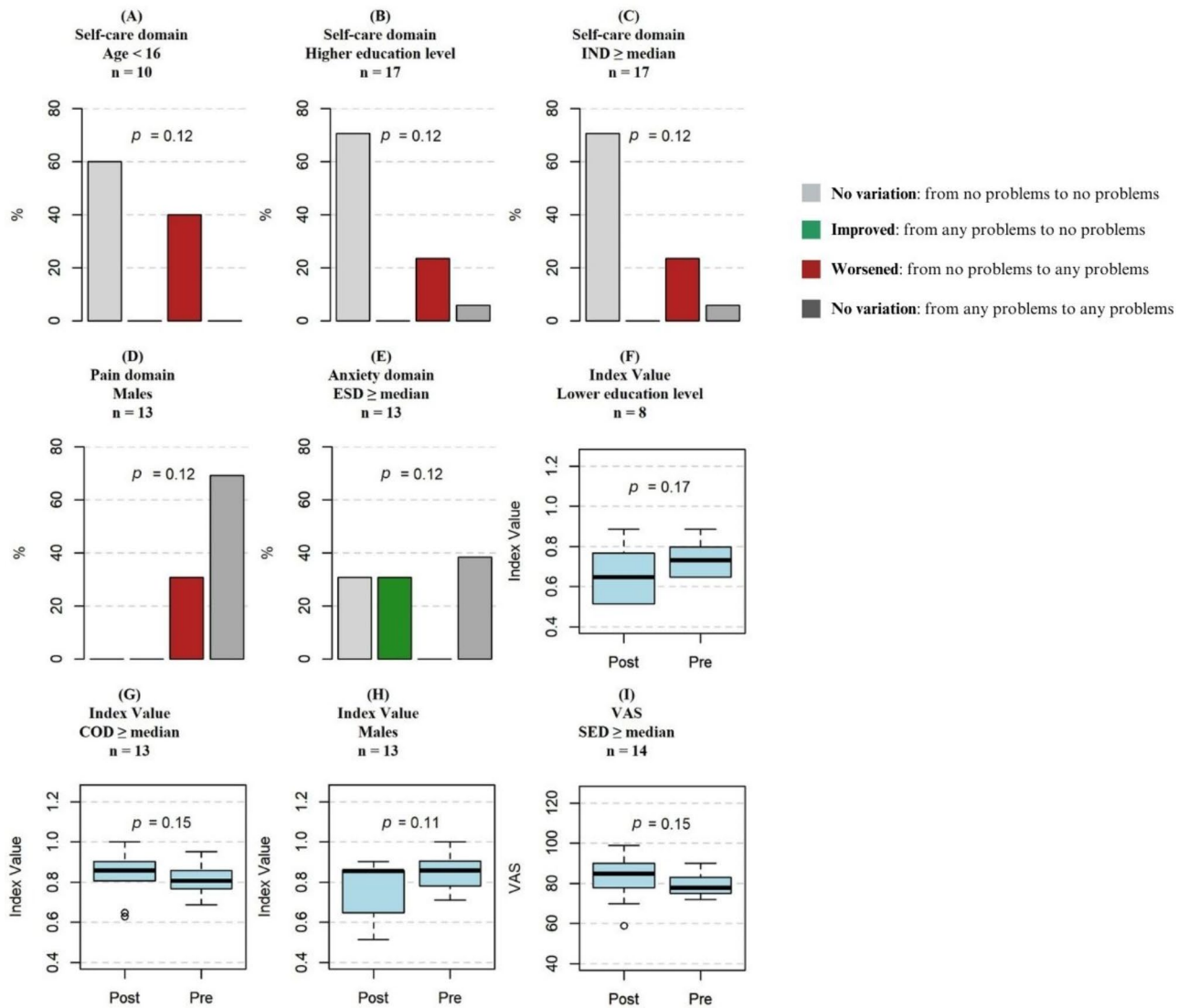


Fig. 4. Interesting trends in HRQoL.

This study’s strengths lie in its innovative approach and robust methodology, including validated assessment tools and a pre-post design. The novelty of applying a sailing program based on adventure therapy principles in a natural setting is particularly noteworthy. These factors reinforce the credibility and relevance of the findings, suggesting that adventure therapy programs can be effectively integrated into clinical treatment plans for individuals with rare skeletal disorders. The positive outcomes observed advocate for the broader application of such interventions in clinical practice. From a practical perspective, healthcare professionals are encouraged to consider incorporating these types of activities into their treatment plans, tailoring them to meet the specific needs and abilities of their patients. The implications for clinical practice extend to the enhancement of patient autonomy, competence, and social inclusion.

However, this study has several limitations. Firstly, due to the Italian Navy anti-COVID-19 regulation, the sample size was small, limiting the generalizability of the findings to a wider population of individuals with rare skeletal disorders. Secondly, the absence of a control group precludes definitive conclusions regarding the causal effect of the sailing program on HRQoL. Future studies should address these limitations by employing larger, randomized controlled trials to provide more robust evidence for the efficacy of sailing programs in this population. Additionally, future research should focus on the long-term effects of nature-based interventions. Specific line of research could include longitudinal studies to confirm whether the benefits observed in this study are sustained over time, could investigate potential moderators and mediators of treatment response, identify which types of activities are most effective, and further refine the therapeutic use of adventure-based interventions. Finally, there is a need to adapt and evaluate these interventions in other populations, particularly those with different chronic conditions or disabilities. This would provide a broader application of the findings and enhance the generalisability of adventure therapy as a therapeutic approach.

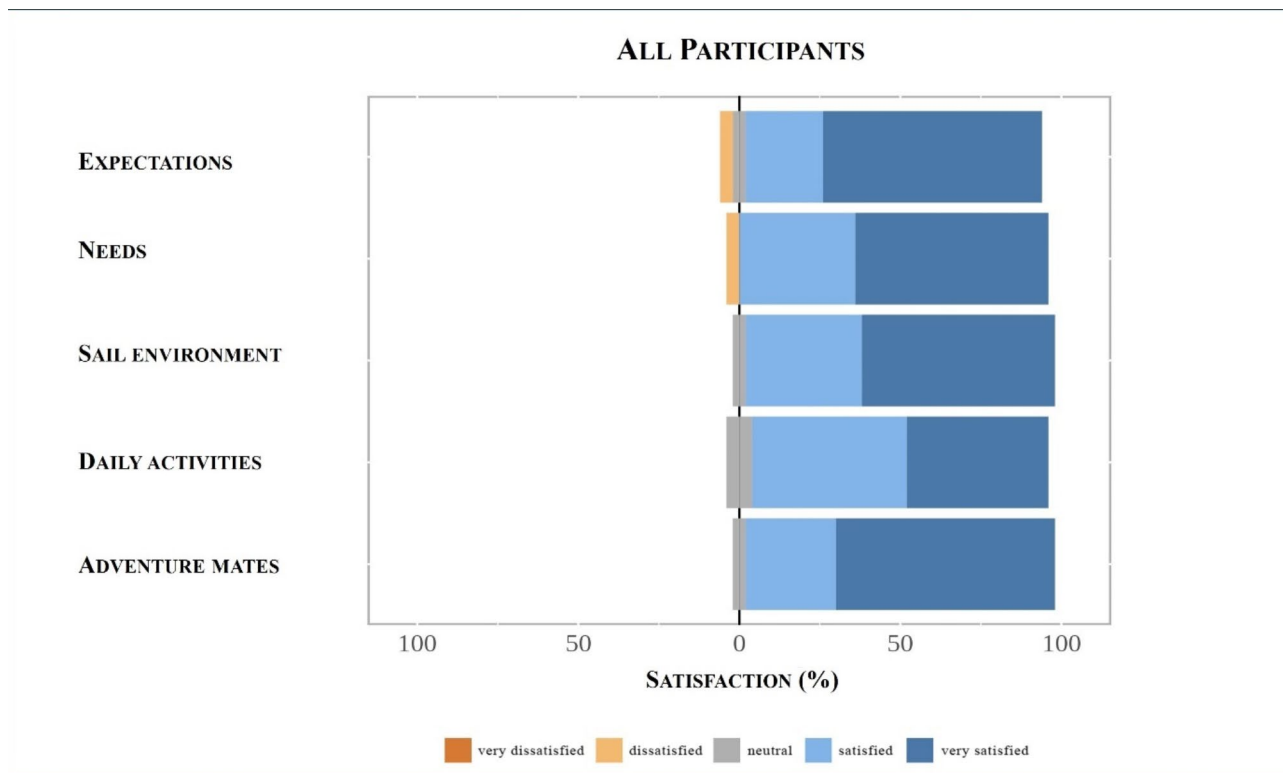


Fig. 5. Overall participants satisfaction Likert plot.

The present study suggests that the sailing program may improve HRQoL in individuals with MO and OD. Emerged findings contribute to the growing body of evidence supporting the efficacy of non-conventional therapeutic interventions in managing rare skeletal disorders. The positive impact of the sailing program on HRQoL aligns with theoretical frameworks suggesting the benefits of nature-based activities in fostering autonomy, competence, and relatedness. The research here reported supports the integration of adventure therapy as a complementary approach to conventional medical treatments in managing chronic conditions, offering a holistic perspective on improving well-being.

Data availability

The data supporting the conclusions of this manuscript will be made available by the corresponding author on a reasonable request. The data are not publicly available due to national privacy regulations.

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Author contributions

MB: Conceptualization; Data curation; Formal analysis; Investigation; Methodology; Project administration; Supervision; Validation; Visualization; Writing – original draft; Writing – review & editing. AR: Conceptualization; Funding acquisition; Methodology; Writing – review & editing. FB: Formal analysis; Methodology; Software; Validation; Visualization; Writing – original draft; Writing – review & editing. EG, SF, MCIF: Conceptualization; Methodology; Writing – review & editing. DS: Conceptualization; Formal analysis; Software; Visualization; Methodology; Writing – review & editing. PCF: Methodology; Resources; Writing – review & editing. LS: Conceptualization; Funding acquisition; Resources; Writing – review & editing. All authors critically reviewed and approved the final manuscript.

Competing interests

The authors declare no competing interests.

Additional information

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