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Feasibility and safety of sailing based rehabilitation for rare skeletal disorders using wearable sensors and patient reported outcomes

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Rare skeletal disorders (RSDs) cause disability, pain, and reduced quality of life (QoL), with limited access to effective rehabilitation. This pilot study evaluated the feasibility, safety, and preliminary effects of a sailing-based rehabilitation program for adolescents with RSDs, integrating wearable inertial sensors and validated patient-reported outcomes. Eight adolescents (12–18 years) with RSDs participated in a five-day sailing intervention focused on physical activity, social interaction, and psychological reflection. Feasibility (recruitment, retention, adherence, acceptability) and safety (adverse events) were primary outcomes. Secondary measures included QoL (EuroQol 5-Dimension), psychological well-being (Pediatric Outcomes Data Collection Instrument, Rosenberg Self-Esteem Scale, Young Persons-Clinical Outcomes in Routine Evaluation, Tampa Scale of Kinesiophobia), and physical function assessed using inertial measurement units. All feasibility criteria met or exceeded: 100% retention and adherence, and high acceptability (mean score 3,75, SD = 0,46). No serious adverse events occurred. Significant benefits were found in proprioception ($p = 0.01$), postural control ($p = 0.01$), gait quality ($p = 0.04$), and upper limb function ($p = 0.02$). Trends toward improved QoL (EuroQol 5-Dimension Visual Analogue Scale, $p = 0.10$), happiness (Pediatric Outcomes Data Collection Instrument, $p = 0.06$), and reduced kinesiophobia (Tampa Scale of Kinesiophobia, $p = 0.03$) were observed. Some effects declined at follow-up. Sailing-based rehabilitation appears feasible, safe, and potentially effective for improving physical and psychological outcomes in RSDs. These findings support the development of larger controlled trials.

Trial registration: ClinicalTrials.gov NCT06397443; release date: May 2, 2024.

Keywords Feasibility and safety, Patient-reported outcomes, Rare skeletal disorders, Sailing-based rehabilitation, Wearable sensors, HRQoL

Rare diseases consist of a heterogeneous group of disorders involving approximately 263–446 million people worldwide, with a deep impact on individuals' lives[1]. Among these, rare skeletal disorders (RSDs) represent a group of conditions characterized by significant skeletal deformities, chronic pain, disability, and reduced

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quality of life[2]. Despite their extreme impact, effective rehabilitation options for RSD patients remain severely limited [3]. This unmet need exacerbates psychological distress and social isolation, with patients often reporting frustration, loss of independence, and difficulty navigating fragmented healthcare systems[4,5]. The lack of adapted rehabilitation services not only delays physical recovery but also contributes to a diminished overall quality of life[6].

Addressing these needs requires innovative and multidisciplinary approaches that integrate physical rehabilitation with psychological support, all within the framework of the International Classification of Functioning, Disability, and Health[7]. Sailing-based programs have emerged as an encouraging intervention with the potential to address the multifaceted needs of RSD patients. These programs leverage nature-based activities and group collaboration to foster physical and psychological health. Previous studies indicate that sailing interventions can reduce distress and enhance well-being in various populations, including cancer survivors and individuals with chronic conditions[8–10]. Furthermore, sailing-based programs provide an engaging environment that combines physical activity with social interaction, essential factors for improving participation and quality of life in individuals facing complex physical and emotional challenges[11].

Prior research on sailing-based rehabilitation have primarily examined psychosocial and quality of life outcomes in different clinical populations. Carta et al. (2014) reported improved well-being in individuals with severe mental disorders after a structured sailing program[8], while Mirandola et al. (2020) observed reduced distress and better quality of life in breast cancer survivors following a tailored sailing intervention[9].

Evidence from neurological and motor disorders also indicates rehabilitative benefits. Recio et al. (2013) showed that sailing supports social integration[12], and Aprile et al. (2016) reported neuromotor improvements[13]. Interventions combining virtual reality with sailing simulators in children with motor and balance impairments, including spinal cord injury, cerebral palsy, diplegia, apraxia, and dyspraxia, have been shown to produce significant improvements in postural control. These benefits arise from the integration of exteroceptive and proprioceptive stimuli, reinforcing the broader therapeutic value of sports engagement for individuals with disabilities in improving quality of life and participation [13].

More recently, Boarini et al. (2024) reported short-term health-related quality-of-life improvements after a five-day sailing program in individuals with RSDs[10]. The present study builds on and extends this body of work by focusing on adolescents with RSDs, integrating synchronized wearable sensor and pressure platform measurements to objectively assess proprioception, postural stability, gait, and upper-limb motor function, and prospectively evaluating feasibility and safety in conjunction with multidomain outcomes and a three-month follow-up period.

Advanced technologies like Inertial Measurement Units (IMUs) are increasingly utilized in rehabilitation research. IMUs provide reliable tools for quantifying complex movement patterns, such as mobility and posture, while carefully monitoring rehabilitation progress[14,15]. For individuals with RSDs, these systems permit precise evaluation of balance, gait, and upper limb function, domains often compromised by their condition. By integrating objective data from IMUs with validated Patient-Reported Outcomes (PROs), well suited to evaluate populations with musculoskeletal conditions, such as EuroQol 5-Dimension (EQ-5D), Pediatric Outcomes Data Collection Instrument (PODCI), Rosenberg Self-Esteem Scale (RSES), Young Persons-Clinical Outcomes in Routine Evaluation (YP-CORE), and Tampa Scale of Kinesiophobia (TSK), this study adopts a multidimensional approach to evaluating both the physical and psychological effects of rehabilitation[16–19].

This research addresses a central public health issue: the urgent need for effective rehabilitation strategies for individuals with RSDs[20]. By systematically evaluating the feasibility and safety of a structured sailing-based intervention, this study contributes to advancing medical knowledge and practice in rehabilitative care while exploring the integration of cutting-edge motion tracking technologies and psychological well-being. Furthermore, this study sets itself within the broader context of emerging trends in rehabilitation research that point up combining advanced technologies with supportive social environments. Such approaches have demonstrated promising results in improving outcomes for patients with rare diseases and other chronic conditions[21–23].

The primary aim of the present study is to evaluate the feasibility and safety of a sailing-based rehabilitation intervention for individuals with RSDs. A secondary aim is to explore potential changes in sensor-derived measures of proprioception, posture, gait, and upper limb function, as well as PROs. Our findings may inform scalable, patient-centered rehabilitation strategies customized to the unique needs of individuals with RSDs, contributing to improved outcomes and guiding future therapeutic approaches for this neglected population[24].

Methods

Study design

This single-center, non-randomized, longitudinal feasibility study was conducted from May 16th to September 13th, 2024, to evaluate feasibility, safety, and describe trends in potential efficacy of a sailing-based rehabilitative intervention for individuals with RSDs.

The study design was chosen to prioritize analysis of feasibility metrics while addressing the challenges of conducting research with small, and heterogeneous populations affected by RSDs. The intervention and assessments followed the CONSORT guidelines for feasibility trials (Appendix A)[25]. A visual summary of the study's timeline and participants assessment is provided in Fig. 1.

All data were managed using the REDCap electronic data storage tool hosted at Yale University. REDCap's real-time data validation, automated audit trails, and centralized management features ensured data integrity across multiple time points, particularly given the complexity of managing repeated measures in a longitudinal study[26].

Ethical approval was obtained from the Local Ethical Committee, Comitato Etico Area Vasta Emilia Centro (CE-AVEC), under protocol number 115/2024/Sper/IOR. The study was registered on ClinicalTrials.

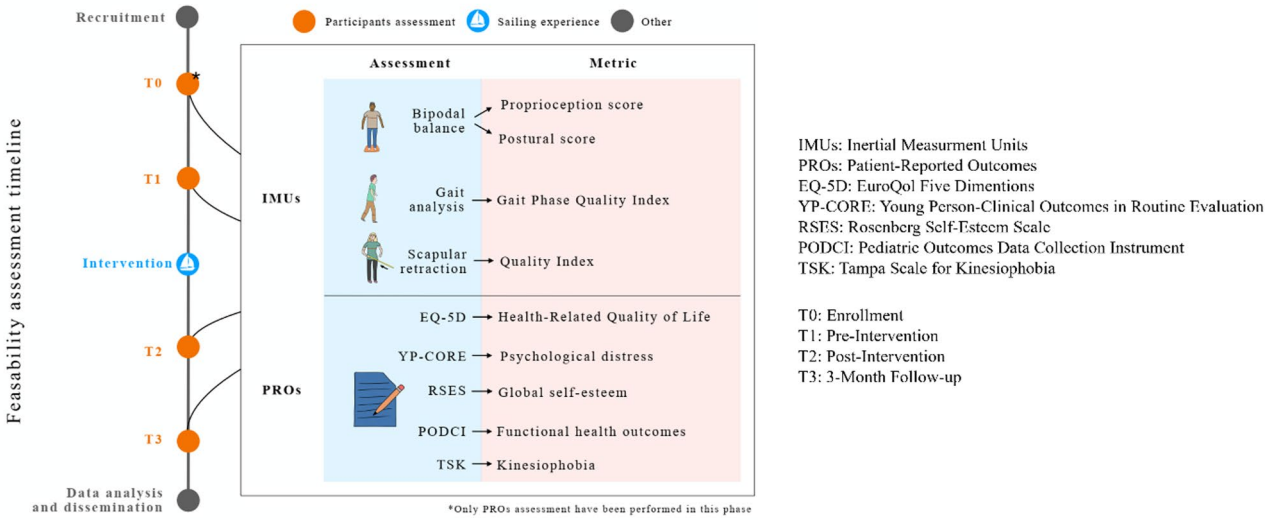

Fig. 1. Feasibility study timeline and participants assessment from recruitment to three-month follow-up.

gov (NCT06397443) and conducted in accordance with the 1964 Declaration of Helsinki and its subsequent amendments.

Setting

The study was conducted across three sites: enrollment (at T0) and three-month follow-up assessment (T3) were performed at IRCCS Istituto Ortopedico Rizzoli (IOR), national referral centre for RSDs, based in Bologna, Italy; sailing sessions were held at the port of Marina di Ravenna, Italy; and 'Palazzo Manzoni' in San Zaccaria (Ravenna, Italy) served as the accommodation site, hosting the pre- and post-intervention (T1 and T2) evaluations.

Participants' recruitment

Participants were recruited and screened from April 10th to May 10th, 2024, using a combined approach, including direct engagement during outpatient visits at the Department of Rare Skeletal Disorders at IOR and through outreach to the national rare disease community, with social media initiatives and patient advocacy networks, in collaboration with UNIAMO-Federazione Italiana Malattie Rare (UNIAMO-FIMR), the Italian umbrella patients advocacy organization, who played an essential role in the study's conceptualization, planning, execution, and dissemination.

Eligibility criteria included: (a) age of 12 years or older; (b) a confirmed diagnosis of a rare skeletal disorder; (c) the ability to engage in physical activities with adaptations; and (d) no surgical interventions within the six months before enrollment. These criteria were selected to ensure a balance between homogeneity for meaningful analyses and practical inclusivity given the rarity of the conditions.

Exclusion criteria included: (a) ongoing evaluation for a differential diagnosis; (b) recent surgical interventions (< 6 months); (c) fractures or musculoskeletal injuries (< 12 months); (d) conditions contraindicating marine environments or sailing activities; (e) absence of a rare skeletal disorder (i.e., healthy individuals); or (f) refused to provide informed consent.

Written informed consent was obtained from participants aged 18 years or older, and parental/guardian consent was secured for minors.

Sample size

Consistent with FDA Rare Disease Guidance (2023), a sample size of eight participants was selected to prioritize feasibility outcomes (e.g., 85% power to detect adherence rates > 75% at $\alpha = 0.10$)[27]. This approach aligns with recommendations for pilot studies in rare populations. Small cohorts are necessary in early-phase studies due to limited recruitment capacity and patient heterogeneity; as noted by Mitani et al. (2020) and Videnovic et al. (2023), small sample sizes are scientifically and ethically justified, providing reliable feasibility data to inform later for larger, confirmatory studies[28,29].

Intervention

The sailing-based intervention (Fig. 2) was conducted through a collaboration between IOR research team, Euleria srl Società Benefit, and Marinando Ravenna ODV, a charitable organization based in Marina di Ravenna. This organization advocates sailing and navigation as effective means of promoting social integration, emphasizing the values of perseverance, discipline, respect for others, inclusivity, and active participation. The

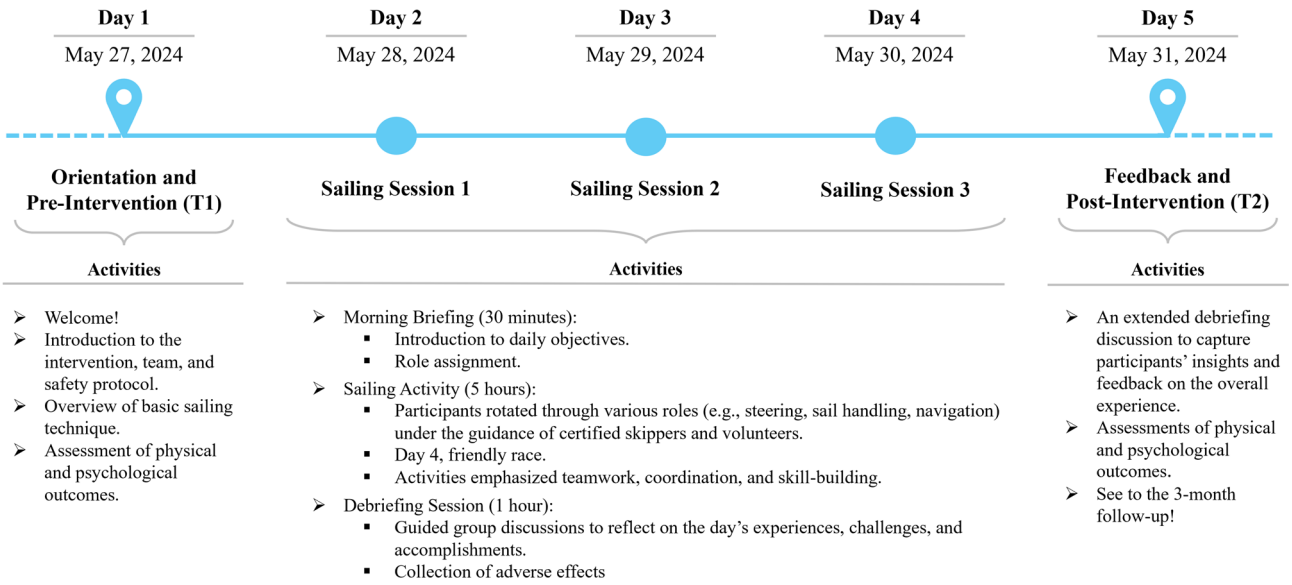

Fig. 2. Timeline and activities of the sailing-based rehabilitative intervention. The timeline and structure of the sailing-based rehabilitation intervention are summarized in this figure. The program covered five days, starting with an orientation session and a pre-intervention assessment (T1) on Day 1. Over the next three days (Days 2–4), participants engaged in daily sailing sessions, which included morning briefings to set objectives, practical sailing activities, and debriefing sessions to reflect on the experiences. On the final day (Day 5), a feedback session and post-intervention assessment (T2) were conducted to gather participants' perspectives and to evaluate both physical and psychological outcomes. A follow-up at three months was also planned to assess the sustainability of the intervention's effects.

sailing sessions took place at the 'Marinara' port in Marina di Ravenna, with each session lasting approximately five hours. Sessions were scheduled from the morning to the afternoon, starting with a brief overview of the daily activities, followed by the division of participants into two groups of equal sizes.

Over a five-day period, participants engaged in three sailing sessions, each lasting five hours. The first and last days were dedicated to physical and psychological data collection, while the intermediate days included sailing activities combined with structured debriefing meetings. Participants were accommodated at the 'Palazzo Manzoni', which served as the site for daily debriefing activities, and hosted the full program on the first and last day. On the first day (May 27th, 2024), participants attended an explanatory session, where they were introduced to the Marinando skippers and volunteers, received basic sailing instruction, and were briefed on safety procedures. On the final day (May 31st, 2024), participants took part in an extended debriefing discussion, offering detailed feedback on their emotional, cognitive, and experiential responses to the intervention. This session lasted approximately two hours.

Two 10-meter sloop-rigged sailing yachts (Oasis 33s model), "Gaia" and "Rolling Black", adapted to accommodate individuals with disabilities and special needs, were used for the sailing sessions (Fig. S1). These boats were skippered by Marinando-certified instructors. Volunteers from Marinando Ravenna ODV supported the participants throughout the activities. Participants actively rotated through various roles on the boat (such as steering, adjusting sails, and assisting with navigation), under the guidance of the skipper. The final session included a friendly race between the two boats. Each sailing session concluded with a one-hour debriefing session conducted by clinical psychologist research assistant, allowing participants to reflect on their experiences, discuss challenges faced, and provide feedback.

In case of adverse weather conditions, contingency plans were implemented to ensure both the safety and engagement of participants. Activities were either rescheduled or adapted while maintaining the treatment aims of the program.

Daily transportation between 'Palazzo Manzoni' and the 'Marinara' port was thoughtfully arranged by the 'Pubblica Assistenza Paolina' of Imola (Bologna, Italy), a public assistance service, ensuring continuous logistical support and participant safety throughout the program.

The intervention adhered to the Template for Intervention Description and Replication (TIDieR, Appendix B), ensuring detailed documentation for replicability[30].

Measures

Demographic and clinical characteristics

Baseline demographic and clinical characteristics, including age, gender, educational level, participation in sports, disease type, anthropometric measurements (height, weight, and BMI), physical impairments, and mobility status, were collected during the enrollment visit at IOR (T0). All assessments were conducted by the physical medicine and rehabilitation team specializing in RSDs, using a study-specific Research Report Form (Appendix C).

Primary outcomes: feasibility and safety

Feasibility was evaluated following Bowen et al. (2009), which fits to this study's context[31]. Five core parameters were assessed: recruitment, retention, compliance, adherence, and acceptability, with thresholds based on existing literature[27,32].

Recruitment was the proportion of eligible participants approached and who consented, with a threshold of ≥80%. Retention reflected participants completing the intervention and assessments, targeting ≥75%. Compliance was measured by attendance, with satisfactory compliance set at ≥75% of sessions. Adherence was defined as total sailing hours completed relative to the planned program, with ≥75% completion deemed satisfactory. Acceptability was assessed post-intervention (T2) using a 5-point Likert scale evaluating agreement with the statement, "I have been happy with the things I have done," from 0 ("Not at all") to 4 ("Most or all the time"), with a mean score ≥3 indicating acceptability.

Safety was monitored after each sailing session using a checklist documenting adverse events, duration, premature discontinuation, and medications. A standardized approach tracked symptom progression and new adverse effects.

This multidimensional assessment comprehensively evaluated the study's feasibility across key metrics.

Secondary outcomes: health-related quality of life (HRQoL), psychological well-being, and physical functioning

HRQoL and psychological well-being were assessed at all time points (T0 to T3) using validated Italian versions of standardized instruments[33–37]. A clinical psychology research assistant specializing in RSDs and affiliated with IOR conducted the assessments with paper-based forms.

The EQ-5D is a standardized generic instrument for assessing HRQoL. This study employed the EQ-5D-5 L version for adults (≥ 16 years), following EuroQol guidelines, which allow use from age 12[38]. The questionnaire evaluates five dimensions: mobility, self-care, usual activities, pain/discomfort, and anxiety/depression, each rated on five severity levels. A summary Index Value (IV) was assigned using the Italian EQ-5D-5 L value set[40]. Health profiles range from the best (11111) to the worst (55555), with IV scores from 1 (perfect health) to 0 (death-equivalent), and negative values for "worse than death." Participants also rated their health on a visual analog scale (VAS) from 0 to 100, with higher scores indicating better overall health status.

The PODCI assesses functional health outcomes in pediatric and adolescent musculoskeletal conditions[41]. It includes 86-item across six scales: Upper Extremity, Transfer and Basic Mobility, Sports and Physical Function, Pain/Comfort, Happiness, and Global Function. Scores range from 0 to 100, with higher values indicating better function. It provides a comprehensive measure of physical ability, pain, and psychosocial satisfaction.

The RSES measures global self-esteem in individuals aged ≥ 12 years[42]. It includes 10 items rated on a 4-point Likert scale, assessing positive (e.g., "I feel I am a person of worth") and negative self-esteem (e.g., "At times, I think I am no good at all"). Scores range from 10 to 40, with higher values indicating greater self-esteem.

The YP-CORE assesses psychological distress and well-being in youth[16]. It consists of 10 items rated on a 5-point Likert scale (0–4), covering emotional well-being, interpersonal functioning, and coping. Scores range from 0 to 40, with higher values indicating greater distress.

The TSK evaluates fear of movement or re-injury due to pain[43]. The 13-item Italian version, rated on a 4-point Likert scale (1–4), measures harm and activity avoidance. Scores range from 13 to 52, with higher values indicating greater kinesiophobia. It is validated for musculoskeletal and chronic pain populations[37].

Physical functioning was assessed from T1 to T3 using objective measures developed in collaboration with biomedical engineers and bioinformatician specializing in RSDs, affiliated with IOR and Euleria Health, a Benefit Company specializing in digital rehabilitation solutions. All assessments were supervised by the physical medicine and rehabilitation team and conducted according to a standardized physical functioning exercises protocol (Appendix D).

The IMUs are wearable, non-invasive devices that provide objective data on movement patterns, including mobility, proprioception, and posture[44]. They offer quantitative measurements that, along with self-reported outcomes, enable a comprehensive assessment of physical improvement. In this study, five IMUs and a pressure board were integrated with dedicated software (Euleria Lab, Euleria srl, Rovereto, Italy). Both systems communicated with the software via Bluetooth Low Energy (BLE), and operated at a 60 Hz sampling frequency, enabling synchronized data acquisition and real-time visualization (Fig. 3). The IMU system has an angular resolution of 0.1° and a dynamic accuracy within ± 2°, thereby meeting the threshold recommended for clinically relevant motion analysis, particularly in monitoring upper-limb movements[45,46]. The IMUs were positioned on specific anatomical landmarks with adjustable elastic bands, as detailed below. Calibration was performed before each assessment, requiring participants to stand still for three seconds while wearing the IMUs, and, when specified, standing on the pressure platform.

Three primary assessments evaluated mobility using IMUs:

(a) Bipodal balance: a single IMU at the sternum level was used to estimate postural stability and the Center of Mass (COM), while the Centre of Pressure (COP) trajectory was derived from the pressure board data as participants stood for 30 s. The test was conducted with both eyes open and closed to assess proprioception, with the eyes-closed condition evaluating reliance on vestibular, and somatosensory inputs in the absence of visual feedback.
(b) Gait analysis: five sensors placed on the feet, tibias, and trunk measured the gait cycle during straight walking. Gait phases were compared to standard references to detect asymmetrical or anomalies.
(c) Arm exercises: participants performed bilateral and unilateral scapular retraction (isometric and repetitive) and overhead lateral shifts while wearing five IMUs on the trunk, wrists, and humeri. These controlled the Euleria Lab exergame and collected kinematic data.

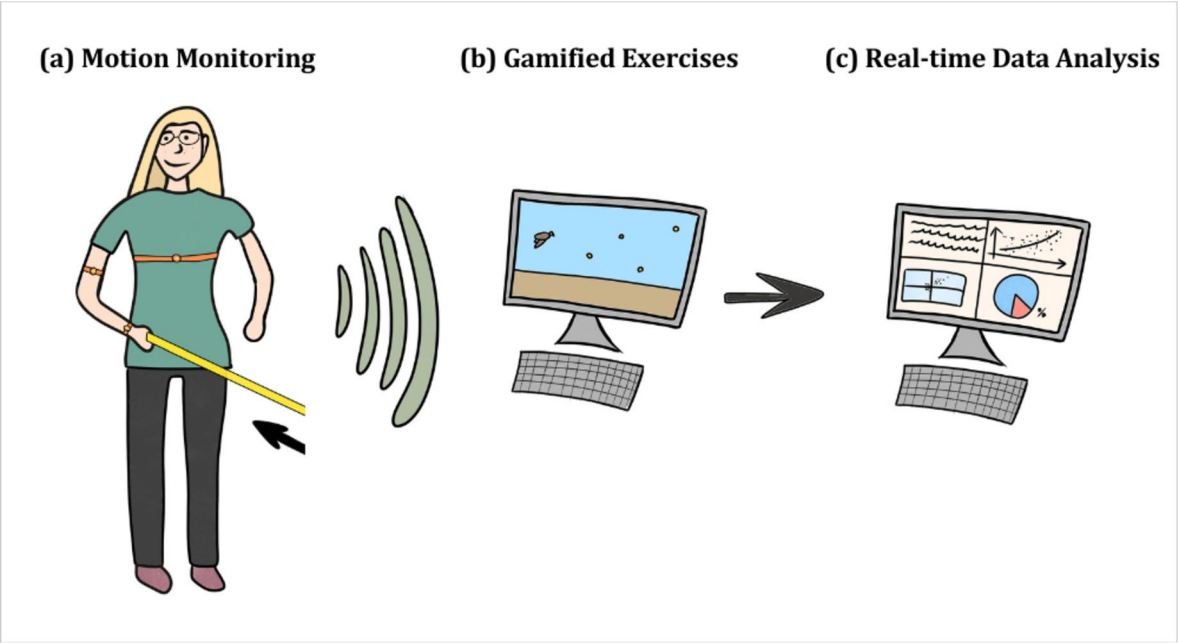

Fig. 3. Workflow for IMU-based motion monitoring, gamification, and data analysis. This figure presents a sensor-based telerehabilitation workflow designed to enhance rehabilitation outcomes. The system incorporates three key components: (**a**) motion monitoring, where wearable sensors capture real-time motion data during patient movements; (**b**) gamified exercises, employing interactive tasks to promote patient adherence and motivation; and (**c**) real-time data analysis, delivering immediate feedback and generating clinically relevant insights to support data-driven decision-making for personalized rehabilitation interventions.

Performance was quantified using:

I. In-game score: The "Arm Quality Index" (AQI), calculated within Euleria Lab software, measured adherence to optimal movement trajectories.
II. Raw and derived scores: Additional metrics included the Gait Phase Quality Index (GPQI), Proprioception Scores, and Postural Scores.

Statistical analysis

Descriptive statistics, including median, minimum and maximum, mean, standard deviations, and frequencies, were used to illustrate the study population and summarize feasibility, and safety outcomes.

HRQoL and psychological well-being outcomes were calculated according to the developers' guidelines.

Physical functioning outcomes were assessed using specific quantitative metrics designed to evaluate proprioception, postural control, gait and arm quality, which are critical for understanding the impact of the intervention.

The Proprioception Score quantifies proprioceptive control by integrating trunk and COP variability. Trunk position was recorded using a sternum IMU, while COP position was measured via a pressure board. Both signals were synchronized at 60 Hz using Euleria Lab software (Euleria srl, Rovereto, Italy).

The score was calculated as:

$$Proprioception\ Score = \frac{1}{\sigma_t + \sigma_{COP}} \times 100$$

where σ_t and σ_{COP} represent the standard deviations of the distribution of the trunk and COP positions, respectively. Lower variability of trunk and COP indicates higher proprioceptive control, reflected by a higher Proprioception Score[47,48].

The Posture Score assesses posture control by evaluating the alignment between trunk and COP. Trunk position was recorded via the sternum IMU, and COP via the pressure board, with both signals synchronized at 60 Hz.

The score was calculated as:

$$Postural\ Score = \frac{1}{\overline{D}_{(t-COP)}} \times 100$$

where $\overline{D}_{(t-COP)}$ is the average Euclidean distance between trunk and COP positions. Smaller distances indicate closer trunk-COP alignment, corresponding to higher posture control and a higher Posture Score[47,48].

The AQI quantifies the performance of the patient during the gaming experience, and it expressed as a percentage.

The GPQI total was calculated using the formula:

$$GPQI_{tot} = \sqrt{\sum_{i=1}^{4}(P_{Ri} - r_i)^2 + \sum_{i=1}^{4}(P_{Li} - r_i)^2}$$

where P represents the observed proportion of time spent in each phase for both the right (R) and left (L) foot and the reference value r for each ith phase of the gait cycle. The reference values (r) correspond to normative gait timing in healthy subjects: heel strike 7%, full stance 39%, heel-off 16%, and swing 38%. The squared differences between the observed and reference values are summed across the four phases for both feet. The square root of this sum yields the GPQI, which reflects how the individual's gait deviates from the normal pattern across all phases of the gait cycle. Higher GPQI values indicate greater deviation from normal gait, capturing both inter-limb asymmetries and overall phase timing differences.

Changes over time were analyzed using the Wilcoxon signed-rank test, selected for its robustness with small sample sizes and non-normal data distributions. Statistical significance was set at $p < 0.05$ and $p < 0.10$, recognizing the exploratory nature of this feasibility study and the inherent limitations of statistical power in small sample sizes. In addition, a significance of $0.10 < p < 0.15$ was applied to highlight potential trends warranting further investigation. Data analysis was performed using R software version 3.6.3 (The R Foundation for Statistical Computing, Wien).

Results

Participant demographics and clinical presentation

A total of eight participants were enrolled in the study (Fig. S2). The cohort was equally distributed by gender (50% male, 50% female), with a mean age of 14 years (SD = 2), ranging from 12 to 18 years. Five out of eight participants (62,5%) had completed middle school, and 87,5% were active in sports. Diagnoses included Multiple Osteochondromas (50%), Ollier Disease (25%), and Osteogenesis Imperfecta (25%). Clinical assessment indicated that 62,5% of participants had physical impairments, including skeletal deformities, functional limitations, or both. Notably, all participants were independent in mobility, as clinically verified.

Table 1 provides a summary of the participants' characteristics.

Feasibility and safety

The feasibility of the study was evaluated across five parameters: recruitment, retention, compliance, adherence, and acceptability (Table S1).

Recruitment reached 80% of the eligible individuals approached (8/10), meeting the ≥ 80% threshold (Fig. 3). Retention was 100% (8/8), exceeding the ≥ 75% threshold. Compliance and adherence were 100% (8/8), surpassing the ≥ 75% threshold for each. Acceptability was assessed post-intervention using a study-specific 5-point Likert scale, with an average score of 3,75 (SD = 0,46), exceeding the threshold of ≥ 3.

The high scores across all domains indicate that the study was highly feasible, with all predefined criteria met or exceeded.

No serious adverse effects were reported. Among the nine predefined categories of mild adverse effects monitored during the sailing-based intervention, only two were reported. Nausea/dizziness occurred in four out of eight participants (50%), predominantly after the second sailing session, whereas muscle soreness was reported by one out of eight participants (12,5%) after each of the three sessions. All adverse events were mild, transient, and self-limiting, requiring neither medication nor medical evaluation, and no participant discontinued the study due to adverse effects. No other categories of adverse events (muscle cramps, thermal discomfort, general fatigue, post-activity drowsiness, bruising/abrasions, temporary joint stiffness, or lower back/neck discomfort) were observed (Fig. 4). Complete results are available in Table S2.

Exploratory impact on HRQoL, psychological well-being, and physical functioning

HRQoL and psychological well-being

The sailing intervention was associated with positive benefits in HRQoL and psychological well-being (Table 2).

Notably, the overall health status (EQ-5D VAS) improved post-intervention ($p = 0.04$), suggesting a better perception of quality of life. Positive changes were observed in happiness (PODCI-Happiness scale, $p = 0.06$), and psychological distress showed a positive trend (YP-CORE, $p = 0.15$), indicating a potential short-term benefit in emotional well-being. However, most improvements were not sustained at three-month follow-up, with most scores showing a decline compared to post-intervention levels.

A sustained reduction in fear of movement (T2 vs. T3, $p = 0.10$) was observed at three months, with the Harm subscale showing a decrease over time (T0 vs. T3, $p = 0.03$). These results suggest that the intervention improved participants' confidence in engaging in physical activities, a critical outcome for long-term behavioral changes. Additionally, trends toward positive changes were observed in the Global Functioning and Pain/Comfort scales (PODCI) post-intervention ($p = 0.15$ for both), although these effects diminished over time. These results

Demographics and Clinical Presentation	Total ($N=8$)
Gender (n, %)	
Male	4 (50,0%)
Female	4 (50,0%)
Age (year)	
Mean (SD)	14 (2)
Median (min-max)	13 (12–18)
Educational level (n, %)	
Middle school/Junior high school	5 (62,5%)
High school	3 (37,5%)
Participating in sport (n, %)	
No	1 (12,5%)
Yes	7 (87,5%)
Disease (n, %)	
Multiple osteochondromas	4 (50,0%)
Ollier disease	2 (25,0%)
Osteogenesis Imperfecta	2 (25,0%)
Anthropometrics (median, min-max)	
Height (cm)	155 (137–180)
Weight (kg)	47,00 (29,50–83,70)
BMI (kg/m^2)	18,04 (15,05–25,83)
Physical impairments (n, %)	
Skeletal deformities	1 (12,5%)
Functional limitations	1 (12,5%)
Skeletal deformities & functional limitations	3 (37,5%)
Not evident physical impairments	3 (37,5%)
Mobility (n, %)	
No helper	8 (100,0%)
Any helper	0 (0%)

Table 1. Participant demographics and clinical presentation. SD: Standard Deviation; Min-Max: Minimum-Maximum.

highlight the transient nature of most intervention-related improvements, while pointing up the importance for ongoing support to maintain long-term benefits.

Complete results are available in Table S3.

Physical functioning

Proprioception and posture In the Eyes Open exercise, marginal improvements in proprioception were observed immediately by post-intervention, becoming statistically significant at the three-month follow-up (T1 vs. T3, $p=0.01$) (Fig. 5a, left panel). Postural stability improvements were significant across all conditions (Fig. 5a, right panel). Participants exhibited immediate benefits following the intervention, with sustained effects at the three-month follow-up (T1 vs. T2, $p=0.01$; T1 vs. T3, $p=0.01$, respectively). Heatmap analysis of balance metrics (Fig. 5b) revealed a slight and progressive reduction in COP displacement particularly in the mediolateral direction and trunk sway in the anteroposterior direction, indicating improved postural control.

Similar patterns emerged in the Eyes Closed exercise, where proprioception reaching statistical significance at the three-month follow-up (T1 vs. T3, $p=0.01$) (Fig. 6a, left panel). Postural scores demonstrated a positive trend post-intervention that was maintained at T3 (T1 vs. T3, $p=0.02$) (Fig. 6a, right panel). Heatmap analyses (Fig. 6b) highlighted consistent reductions in COP sway and trunk displacement across both anteroposterior and mediolateral directions during static balance tests, underscoring improved sensorimotor integration and postural stability in the absence of visual input. These findings suggest that the intervention effectively enhances proprioceptive function and balance control independently of visual compensation, with sustained benefits evident at follow-up.

Gait phase quality index and arm quality index No significant difference in total GPQI was observed between the pre- and post-intervention assessments (T1 vs. T2, $p=0.38$). However, a significant decrease in total GPQI was detected over time (T1 vs. T3, $p=0.04$) (Fig. 7a).

The intervention significantly improved upper limb function, particularly in repetitive and isometric movements involving the scapular region. For repeated bi-scapular retractions, increased AQI was noted

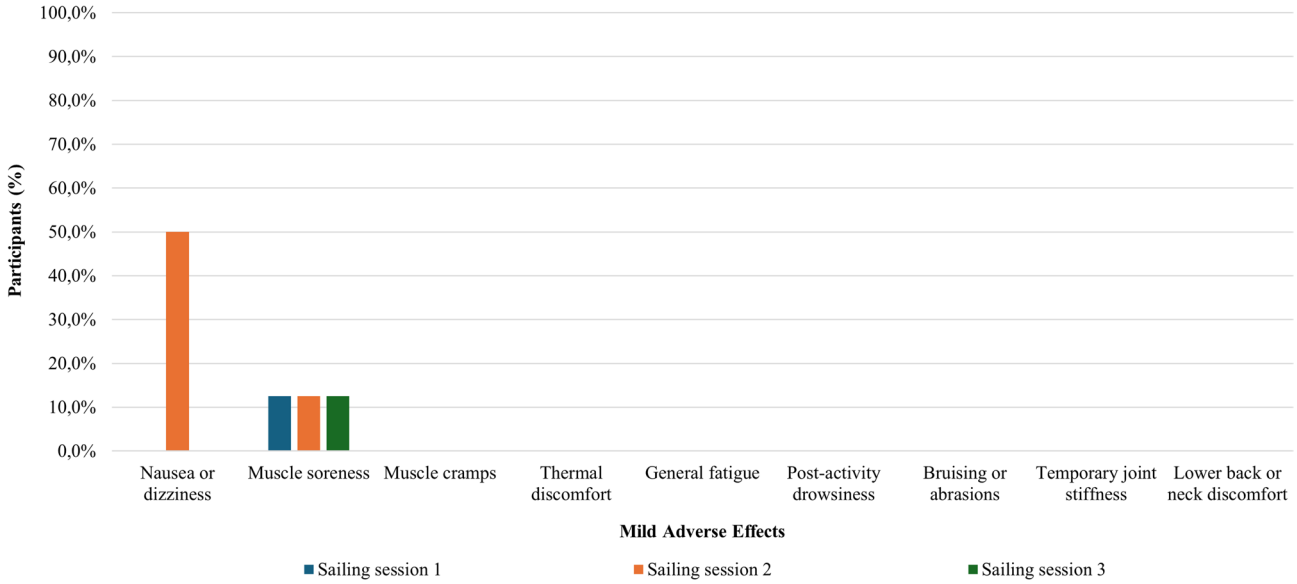

Fig. 4. Mild adverse effects reported by participants across the sailing-based intervention ($N = 8$). The figure illustrates the incidence of mild adverse effects across the three sailing sessions during the sailing-based rehabilitation intervention. The most frequently reported adverse effect was nausea or dizziness, occurring in approximately 50% of participants during the second sailing session. Muscle soreness was reported by a smaller proportion of participants across all sessions, with no reports of muscle cramps, thermal discomfort, general fatigue, post-activity drowsiness, bruising or abrasions, temporary joint stiffness, or lower back or neck discomfort.

HRQoL & psychological well-being scales	Median score (min-max)								*p* value[a]	
	Enrollment (T0)		Pre-intervention (T1)		Post-intervention (T2)		3-month follow-up (T3)			
EQ-5D										
VAS - Global Health	84.00	(50.00–99.00)	90.50	(45.00-100)	93.50	(70.00-100)	83.50	(50.00–99.00)	T0 vs. T2, $p = 0.04$** T1 vs. T2, $p = 0.10$*	↑ ↑
PODCI										
Happiness	85.00	(55.00-100)	82.50	(40.00–95)	92.50	(40.00-100)	80.00	(55.00-100)	T1 vs. T2, $p = 0.06$*	↑
Pain/Comfort	81.11	(46.11–100)	71.39	(47.78–100)	83.61	(49.44–100)	67.22	(38.33–93.33)	T0 vs. T2, $p = 0.15$	↔
Global Functioning	84.68	(69.65–100)	86.71	(71.79–98.96)	86.40	(79.91–100)	85.73	(77.10–97.50)	T0 vs. T2, $p = 0.15$ T1 vs. T2, $p = 0.15$	↔ ↔
YP-CORE										
Psychological Distress	0.85	(0.10 – 2.70)			0.45	(0.10 – 1.60)	1.25	(0.60 – 2.30)	T0 vs. T2, $p = 0.15$ T2 vs. T3, $p = 0.06$*	↔ ↓
TSK										
Harm Subscale	11.00	(8–18)			12.00	(10–17)	8.00	(5–11)	T0 vs. T3, $p = 0.03$** T2 vs. T3, $p = 0.02$**	↑ ↑
Total Fear of Movement	21.50	(15–35)			22.50	(19–30)	18.50	(12–28)	T2 vs. T3, $p = 0.10$*	↑

Table 2. Sailing-based intervention's significant impact on HRQoL and psychological well-being ($N = 8$).
[a] Comparisons of continuous variables were performed using the Wilcoxon signed-rank test. ** $p < 0.05$; * $p < 0.10$. $0.10 < p < 0.15$: promising trends. ↑: improvement; ↓: worsening; ↔: promising trend. EQ-5D: EuroQol 5-Dimension questionnaire; PODCI: Pediatric Outcomes Data Collection Instrument; YP-CORE: Young Person's Clinical Outcomes in Routine Evaluation; TSK: Tampa Scale of Kinesiophobia.

post-intervention (T1 vs. T2, $p = 0.02$) (Fig. 7b). In isometric bi-scapular retraction, a positive trend of AQI was observed between pre- and post- intervention, although not significant ($0.10 < p < 0.15$) (Fig. 7c).

Detailed results of GPQI, and AQI scores over time can be found in Fig. S3.

Discussion

RSDs represent a significant challenge in rehabilitation medicine, given their overwhelming impact on physical function, psychological well-being, and overall quality of life. Despite progresses in genetic diagnosis and

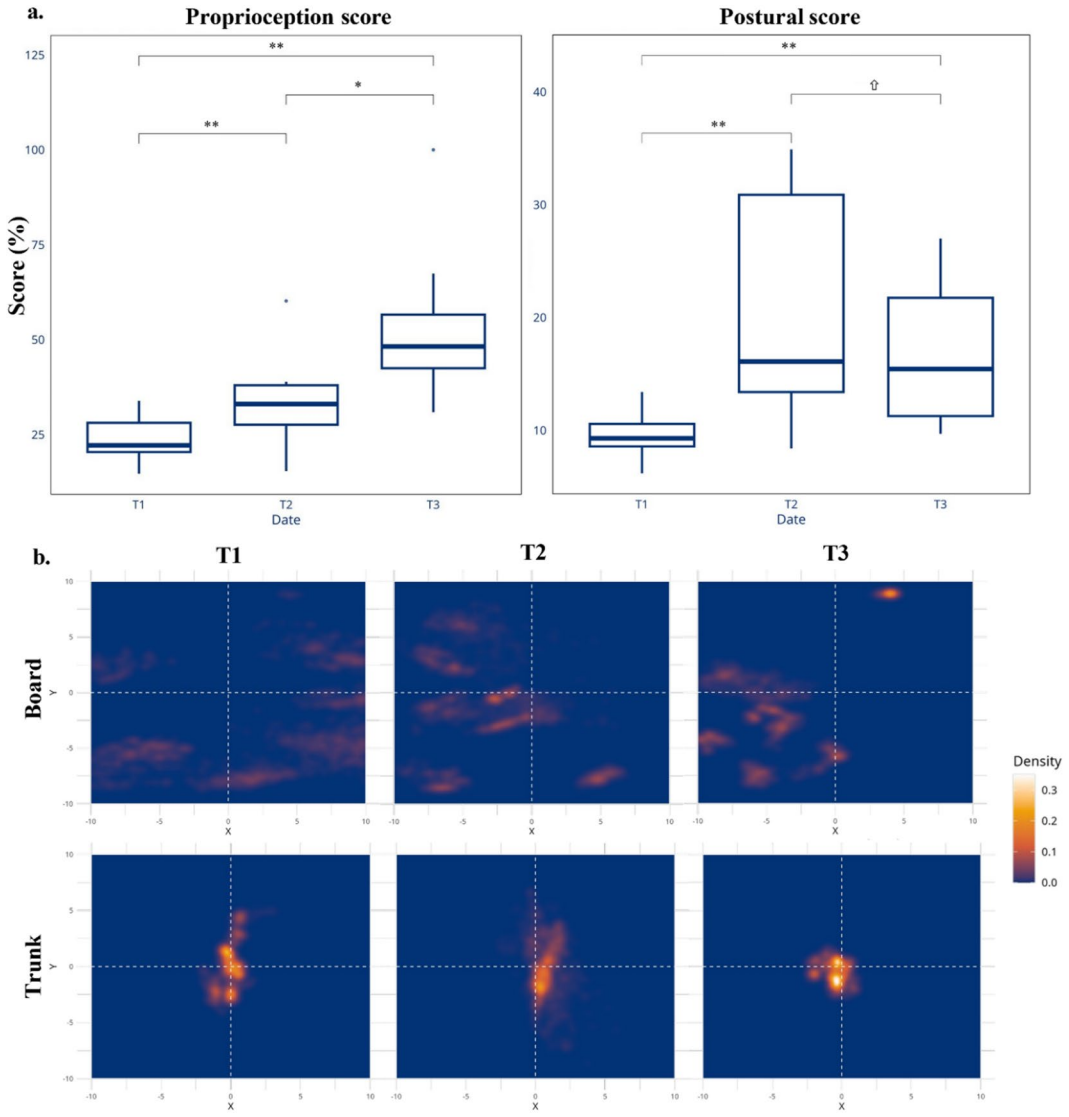

Fig. 5. Evaluation of proprioception, posture, and balance over time (eyes open) ($N = 8$). (**a**) Box plots of proprioception (left) and posture (right) scores measured at three time points: T1 (pre-intervention), T2 (post- intervention), and T3 (three-month follow-up). (**b**) Heatmaps represent the balance metrics in the anteroposterior (y-axis) and mediolateral (x-axis) directions for all eight participants: COP (top) and trunk sensor (bottom) coordinates during a 30-second static balance test at T1, T2, and T3.

therapeutic approaches, effective rehabilitation strategies for these conditions remain scarce, intensifying the burden of disease for patients and their families[1,2]. This study addresses these unmet needs by evaluating the feasibility and safety of a sailing-based rehabilitative intervention while exploring its potential to improve both physical and psychological outcomes through an innovative, multidisciplinary approach.

Our findings demonstrate that a sailing-based intervention is feasible and safe for adolescents with RSDs. High recruitment, retention, compliance, and adherence rates indicate strong participant engagement and acceptability. Moreover, mild and transient adverse effects underline the safety of this intervention. Notably, participants experienced positive benefits in physical functioning, psychological well-being, as well as HRQoL. These results align with prior evidence suggesting that sailing-based interventions could effectively address the multifaceted needs of individuals with chronic conditions by combining physical activity with social interaction in an engaging environment[8–13].

The observed benefits may be attributed to several mechanisms. The intense and repetitive physical activities of sailing likely contributed to improved proprioception and postural stability, as evidenced by positive trends in sensory-motor integration and adaptive balance mechanisms. Additionally, mastering sailing skills may

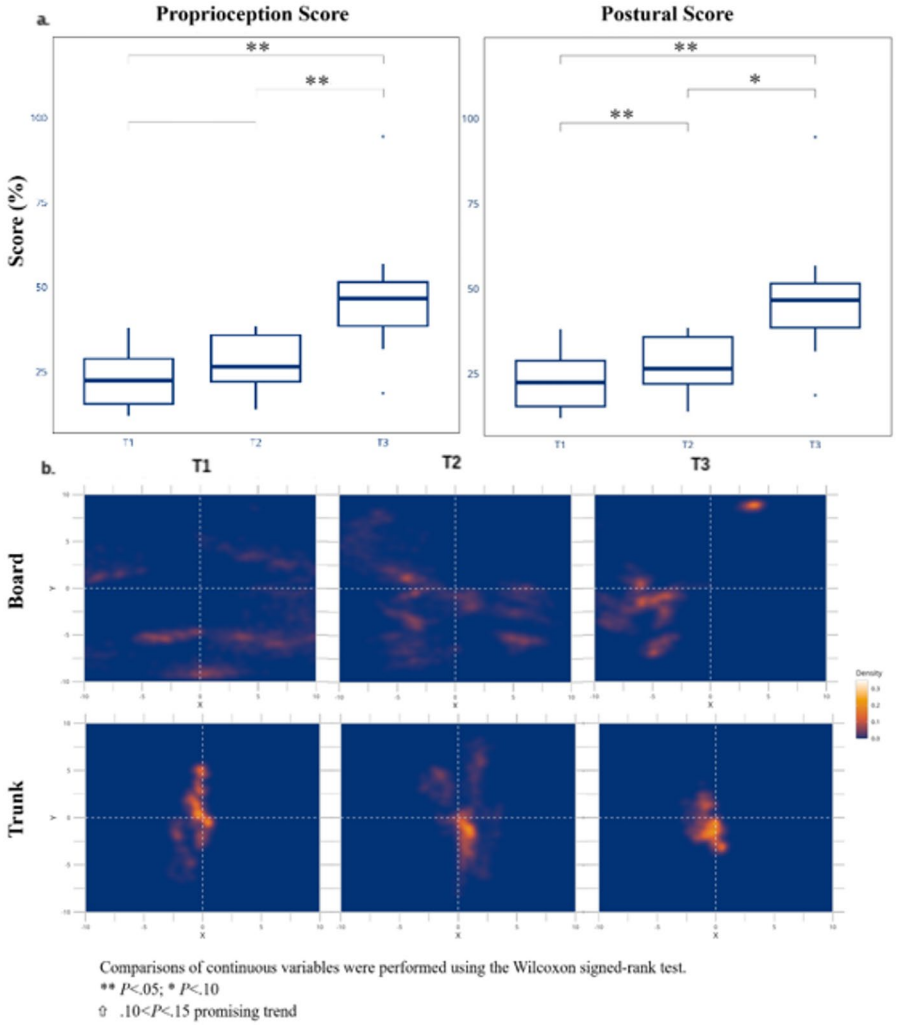

Fig. 6. Evaluation of proprioception, posture, and balance over time (eyes closed) ($N = 8$). (**a**) Box plots of proprioception (left) and posture (right) scores measured at three time points: T1 (pre-intervention), T2 (post- intervention), and T3 (three-month follow-up). (**b**) Heatmaps represent the balance metrics in the anteroposterior (y-axis) and mediolateral (x-axis) directions for all eight participants: COP (top) and trunk sensor (bottom) coordinates during a 30-second static balance test at T1, T2, and T3.

have enhanced participants' confidence in managing their condition and daily activities, thereby fostering self-efficacy. The collaborative nature of sailing promoted social relationships among participants, potentially reducing feelings of isolation that are common among RSD patients.

Furthermore, overcoming challenges in an unconventional environment with others may have contributed to the participants' promotion of psychological well-being, such as happiness and better perception of overall health status, consistent with research on nature-based therapies[49–51]. These psychological aspects are particularly relevant for patients with RSDs, who often navigate fragmented healthcare systems and limited opportunities for social engagement[4,5].

Objective assessments using IMUs integrated with a pressure board revealed positive improvements in proprioception, postural stability, and neuromotor coordination. These findings are consistent with evidence supporting balance-challenging interventions aimed at improving sensory-motor integration[14,52]. In addition, improvements in gait quality and upper limb function suggest that sailing-based activities may act as a comprehensive rehabilitative tool targeting multiple domains affected by RSDs. However, the observed decline in outcomes at the three-month follow-up highlights the need for sustained interventions to maintain these benefits over time. This aligns with studies advocating longitudinal strategies to promote lasting functional benefits in chronic musculoskeletal populations[53,54].

The integration of advanced technologies such as IMUs represents methodological advancement in rehabilitation research. These tools provide precise, objective measures of movement patterns that complement PROs, offering a multidimensional approach to evaluating intervention[15,19]. For instance, combining IMU data with validated PROs such as EQ-5D or PODCI enables a comprehensive assessment of both physical and

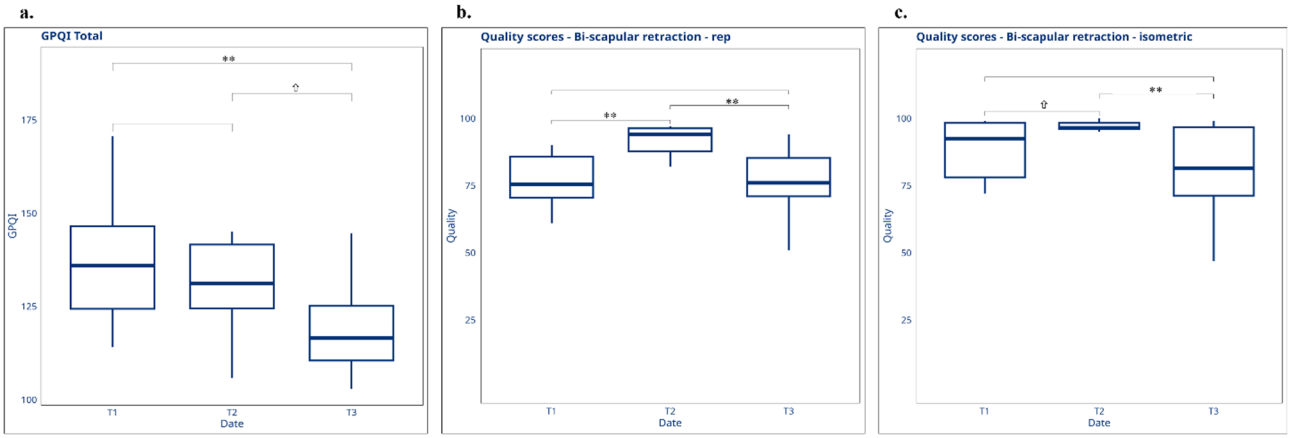

Fig. 7. Evaluation of Gait Phase Quality Index (GPQI), and Arm Quality Index (AQI) over time ($N = 8$). Box plots of GPQI total scores (**a**), AQI in repeated bi-scapular retraction (**b**), and AQI in isometric bi-scapular retraction (**c**) measured at three time points: T1 (pre-intervention), T2 (post- intervention), and T3 (three-month follow-up).

psychological domains. This approach aligns with current trends in evidence-based medicine that emphasize integrating objective data with patient-centered measures to inform clinical practice[25,30].

While these findings are promising, several limitations must be acknowledged. The small sample size limits statistical significance and generalizability. Additionally, the absence of a control group precludes definitive conclusions regarding the efficacy of this intervention compared to standard treatments. The brief follow-up period limits our ability to assess the long-term sustainability of benefits. Future studies should address these limitations by adopting randomized controlled designs with larger sample sizes and extended follow-up periods, while also exploring complementary approaches such as virtual reality-based sailing simulations to enhance accessibility for patients unable to participate in physical activities[55]. Such studies would provide more robust evidence on the effectiveness of sailing-based rehabilitation while allowing for a deeper understanding of its mechanisms of action.

Conclusion

In conclusion, this study contributes valuable evidence supporting the feasibility and safety of sailing-based rehabilitation for individuals with RSDs. By addressing both physical impairments and psychosocial challenges within a holistic framework, this intervention aligns with the principles of evidence-based medicine by offering an innovative solution to a pressing public health issue. Future research should build on these findings by leveraging advanced technologies and multidisciplinary approaches to develop scalable, patient-centered rehabilitation strategies adapted to the unique needs of individuals with RSDs and beyond.

Data availability

The data supporting the conclusions of this manuscript will be made available by the corresponding author on a reasonable request. The data is 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. FB: Formal analysis; Methodology; Validation; Writing – review & editing. DS: Data curation; Formal analysis; Methodology; Validation; Visualization; Writing – original draft; Writing – review & editing. GMF: Conceptualization; Data curation; Investigation; Methodology; Validation; Writing – original draft; Writing – review & editing. GR: Data curation; Investigation; Methodology; Validation; Visualization; Writing – original draft; Writing – review & editing. EDS, and GV: Investigation; Methodology; Software; Validation; Writing – review & editing. DP, LB, AL, and SS: Conceptualization; Investigation; Methodology; Writing – review & editing. SF, AS: Conceptualization; Methodology; Writing – review & editing. ML: Investigation; Writing - review & editing. LS: Conceptualization; Funding acquisition; Investigation; Methodology; Resources; Supervision; Validation; Visualization; Writing – original draft; Writing – review & editing. All authors participated in the reading, critical review, and approval of the final manuscript. Multiple authors directly accessed and verified the underlying data reported in the study. All authors had full access to the study data and assumed responsibility for the decision to submit the manuscript for publication. Written informed consent for the publication of identifiable images and/or video recordings of participants was obtained from all adult participants involved in the study. For participants under the age of 18, written informed consent for publication was obtained from their parents/guardians.

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Declarations

Competing interests
The authors declare no competing interests.

Ethical statement and informed consent
The study was approved by the Local Ethical Committee, Comitato Etico Area Vasta Emilia Centro (CE-AVEC), under protocol number: 115/2024/Sper/IOR, and conducted in accordance with the 1964 Declaration of Helsinki. Written informed consent was obtained from all participants aged ≥ 18 years and from parents/guardians of those participants aged < 18 years.

Additional information
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