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Mindful Self-Compassion to Reduce Pain Interference Among Adults with Osteogenesis Imperfecta

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Abstract

Between 60 and 80% of adults with osteogenesis imperfecta (OI) experience chronic pain and associated interference. Currently available pain therapies often provide marginal efficacy. Mindful self-compassion (MSC) has emerged as a promising intervention for coping with chronic pain. We conducted a single-center 8-week pilot intervention study to assess the feasibility and acceptability of a MSC program among adults with OI and co-occurring chronic pain. Individuals attended the validated MSC course consisting of 8 weekly virtual 2-h sessions. Participants completed a battery of validated questionnaires assessing pain, various aspects of well-being, and physical function at baseline and post-intervention. Participants wore the ActiGraph GT9X Link watch to measure sleep duration and sleep efficiency. Seven adults with OI and co-occurring pain participated in the MSC program. The program was feasible, as indicated by high attendance and high questionnaire completion rates. Participants reported a mean $\pm$ standard deviation (SD) of 3.5 out of 5 ± 0.4 on the Intervention Acceptability Framework. 86% (6/7) of participants found the MSC program to be acceptable. While our pilot study was not powered to show efficacy, we observed a decrease in pain interference on the PROMIS pain interference questionnaire (mean 55.9 ± 5.5 at baseline vs. 50.0 ± 7.3 at 8 weeks). Implementation of the MSC program is feasible as a potential therapeutic option to address chronic pain in OI.

Keywords Osteogenesis imperfecta · Pain · Mindful self-compassion · Chronic disease · Positive psychology

Abbreviations

BEAQ	Brief experiential avoidance questionnaire
CHEO	Children's hospital of eastern Ontario
CPAQ	Chronic pain acceptance questionnaire
DERS	Difficulties in emotion regulation scale
IQR	Interquartile range
MSC	Mindful self-compassion
NSAIDs	Non-steroidal anti-inflammatory drugs
OI	Osteogenesis imperfecta
PCS	Pain catastrophizing scale
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PROMIS	Patient reported outcomes measurement information system
REDcap	Research electronic data capture
SCS	Self-compassion scale
SD	Standard deviation

Introduction

Osteogenesis imperfecta (OI) is a heritable disorder characterized by bone fragility and connective-tissue defects (Rapoport et al., 2023). The severity of OI ranges widely from perinatal mortality to mildly increased fracture risk (Hald et al., 2016; Rauch & Glorieux, 2004). Individuals with moderate to severe OI fall between these extremes, with symptoms such as vertebral compression fractures, long-bone deformities, and short stature. OI has been traditionally classified into four main types: Type I, the mildest form with normal or mild short stature, mild or absent deformities and increased fracture risk; Type II, the perinatal lethal form; Type III, the more severe type in survivors, with severe bone deformities, repeated long-bone fractures and severe short stature; and Type IV, of intermediate severity between OI types I and III (Palomo et al., 2017).

Adults with OI experience chronic pain, defined as pain that persists beyond normal healing time and persists for longer than 3 months (Treede et al., 2015), at levels that critically surpass the general population; 60–80% of individuals with OI are impacted by chronic pain (Muñoz Cortés et al., 2022; Rodriguez Celin et al., 2023) compared to 11–40% of the general population (Cohen et al., 2021). Chronic pain may stem from structural deformities inherent to OI, past vertebral compression fractures (Nghiem et al., 2018), and lifetime fractures (Muñoz Cortés et al., 2022). Individuals with all OI types are impacted by chronic pain (Rodriguez Celin et al., 2023), which significantly undermines emotional and social well-being and impairs their ability to function in daily life (Hill et al., 2022).

Currently available pain therapies for individuals with OI include bisphosphonates, non-steroidal anti-inflammatory drugs (NSAIDs), physiotherapy, opioids, and surgery. However, pain experienced by individuals with OI persists despite these interventions (Nghiem et al., 2018). In addition, most individuals report not wanting to use pharmacological treatments because of fear of stigma or shame from taking opioids or other high-intensity pain relievers, as well as fear of side effects (Shepherd et al., 2024). Despite this, non-pharmacological studies focusing on mitigating the effects of chronic pain with OI have been limited. In addition to pain, almost 60% of individuals with OI report getting less than 7 h of sleep each night, and over 45% indicate that they experience significant sleep problems (Wiese et al., 2024).

Existing treatments for pain in OI may only be marginally effective in part because they do not specifically target brain regions implicated in chronic pain (Melzack, 2001). In individuals with chronic pain, structural changes often occur in brain regions which are also involved in emotional and attentional modulation of pain (i.e., prefrontal cortex, anterior cingulate cortex, primary and secondary cortices, insula) (Bushnell et al., 2013; Melzack, 2001). These structural changes can lead to impairments in emotional processing and affect (Bushnell et al., 2013). Psychological interventions can target those brain regions, by way of their effect on emotional regulation and affect, making them ideally suited for refractory chronic pain (Day et al., 2012).

Mindful self-compassion (MSC) is a positive psychological construct that involves learning to treat oneself with the kindness and understanding one would show to loved ones. MSC has three core tenants: self-kindness (being kind and understanding toward oneself in instances of pain or failure), common humanity (perceiving one's experiences as part of the larger human experience, rather than seeing them as separate and isolating), and mindfulness (holding painful thoughts and feelings in balance, rather than over identifying with them Neff, 2003; Neff & Germer, 2013a)). It is meant to, in the context of pain management, act on the perception of pain, rather than the pain stimulus and is an indirect way of dealing with the pain. MSC programs are effective at increasing self-compassion, mindfulness, and well-being (Neff & Germer, 2013a). In the realm of chronic pain, MSC has led to increased pain acceptance, increased self-compassion, and a reduction in anxiety among individuals with chronic pain of various etiologies (Torrijos-Zarcero et al., 2021a, 2021b). In the realm of sleep, self-compassion is associated with better self-reported sleep quality (Brown et al., 2021), and self-compassion interventions have been shown to improve sleep quality via decreased rumination (Butz & Stahlberg, 2018).

We propose that a personalized MSC intervention is ideally and specifically suited to decrease pain interference among individuals with OI. First, individuals with OI have painful childhood-onset fractures and deformities, and trauma associated with their pain experiences (Swezey et al., 2019) which may be associated with avoidance behavior (W. C. Rork et al., 2023a, 2023b). Second, their pain negatively influences their relationships (Wiese et al., 2024) and may contribute to dissatisfaction with participation in social roles (Tosi et al., 2019), potentially leading to feelings of isolation. Third, individuals with OI express frequent feelings of self-judgment and shame related to their pain and treatments (i.e., opioids), given the limited understanding of pain experiences in OI among health care providers and the general public (Shepherd et al., 2024). The three core tenants of MSC specifically address malleable deficits observed in OI: mindfulness to counteract avoidance, a sense of common

humanity to counteract isolation, and self-kindness to counteract self-judgment. In turn, the malleable negative effects in OI (avoidance, isolation, and self-judgment), which are targeted by MSC, are associated with increased levels of pain interference (Nghiem et al., 2018; Shepherd et al., 2024) and poor mental health (Wiese et al., 2024).

Previous work demonstrates that MSC can be delivered successfully to many clinical groups (Brooker et al., 2020; Neff & Germer, 2013b; Torrijos-Zarcero et al., 2021a, 2021b). Nevertheless, in the context of OI, the acceptability and feasibility of MSC remain uncertain owing to the disorder's distinctive challenges (W Conor Rork et al., 2023a, 2023b), which may influence willingness to engage and the resonance of program content. Therefore, establishing the acceptability and feasibility of MSC in the OI community is a necessary first step before efficacy testing and broad implementation.

In this study, we evaluated the feasibility and acceptability of an 8-week MSC intervention among adults with OI and co-occurring chronic pain and explored its effect on pain interference, other pain-related outcomes, mental health outcomes, general well-being, and sleep.

Material and Methods

Study Design

We conducted a single-center 8-week pilot intervention study to assess the feasibility and acceptability of the MSC program among seven adults with OI patients and co-occurring chronic pain. A standardized battery of assessments was completed at baseline and 1-week post-MSC intervention, as detailed below. The study was reviewed and approved by the Children's Hospital of Eastern Ontario (CHEO) Research Ethics Board.

Participants & Recruitment

We recruited participants (≥ 18 years) by screening medical records of former patients seen at CHEO and who had transitioned to adult care between September 2019 and December 2023, as well as their family members with a clinical diagnosis of OI confirmed in their own medical charts. We obtained informed consent either virtually via REDcap (Research Electronic Data Capture) platform (Harris et al., 2009, 2019), verbally over the phone, or in-person.

Participants were recruited if they met the following criteria: Age ≥ 18 years, diagnosed with OI (any type), chronic pain (defined as having pain on most days in the previous three months), able to attend at least 6 of the 8 scheduled MSC program sessions (which is aligned with other MSC studies) (Friis et al., 2016), have a regular health care provider so that proper follow-up can be arranged if participants disclosed high psychological distress during the study (i.e., family doctor, endocrinologist, etc.), and able to provide informed consent. Participants were excluded from the study if they met any of the following criteria: do not speak English with enough fluency to complete all study-related tasks, and active participation in another mental health intervention research trial.

MSC Intervention

The MSC program has been validated in several patient populations such as community adults, cancer patients, and patients with chronic pain groups, demonstrating feasibility, acceptability, and psychological benefits (Brooker et al., 2020; Neff & Germer, 2013b; Torrijos-Zarcero et al., 2021a, 2021b). The MSC intervention (Brooker et al., 2020) consists of 8 weekly virtual interactive group-based sessions, each lasting for 2 h (16 h in total). Each session integrates didactic teaching, experiential exercises, guided meditations, and group discussion (Brooker et al., 2020). Core components include formal mindfulness practices such as affectionate breathing, loving-kindness meditation, and the self-compassion break (Brooker et al., 2020). Experiential exercises include writing a compassionate letter to oneself, practicing soothing touch, and engaging in role-play to dialogue with the inner critic (Brooker et al., 2020). Participants are encouraged to engage in daily home practices, including guided meditations and reflective journaling, to reinforce the skills introduced during sessions (Brooker et al., 2020).

Although the course is typically delivered to groups of 12–15 people, in this pilot study, the 7 participants formed a single group for the MSC intervention. The MSC program encourages and teaches the participants to be open to, rather than disconnected from, their own suffering and treating themselves with the same care they would show to a friend. All participants were asked to keep their cameras on for the duration of the sessions to help create a sense of community, facilitate group interaction, and ensure participant safety. Participants were offered optional home-based practices and activities throughout the course, but completion was not mandatory. The trial coordinator provided weekly reminders for the duration of the program, via SMS message or email, depending on participant preference, to maximize participant engagement. Each session of the MSC program focused on a specific topic (Table 1; (Germer & Neff, 2019)). Two trained MSC instructors (one of which was CP) led the program.

Data & Outcome Measures

Participants completed a baseline demographic questionnaire (age, marital status, self-reporting gender identity, race/ethnicity, employment status, medical history).

Table 1 MSC Course Content

Session	MSC Intervention Topic	Example Exercises:
Week 1	Introduction to MSC: what is MSC, misgivings of self-compassion, physiology of self-compassion and self-criticism	Self-compassion break; soothing and supportive touch
Week 2	Practicing mindfulness: wandering mind, resistance	Affectionate breathing meditation; soles of the feet
Week 3	Practicing loving-kindness: kindness and compassion; loving phrases practice	Affectionate breathing, loving-kindness phrases, loving-kindness meditation
Week 4	Discovering your self-compassionatevVoice: stages of progress; self-criticism and safety	Loving kindness for ourselves; motivating ourselves with self-compassion
Week 5	Living deeply: core values, finding hidden value in suffering	Giving and receiving compassion; compassionate listening
Week 6	Meeting difficult emotions: stages of acceptance; approaches to difficult emotions	Loving kindness for ourselves; being with difficult emotions
Week 7	Exploring challenging relationships: challenging relationships, pain of disconnection	Compassionate friend; anger and unmet needs; self-compassion break in relationships; compassion with equanimity
Week 8	Embracing your life: compassion for self and others; maintaining your practice	Compassion for self and others; gratitude for small things; appreciating our qualities

(Germer & Neff, 2019)

Primary Outcomes

Our primary outcomes were *feasibility* and *acceptability* of the MSC program. The intervention was defined as feasible if > 75% of participants completed at least 6 of the 8 classes, in line with other studies using the MSC intervention (Brooker et al., 2020) and < 10% of questions from questionnaires left blank. Acceptability of the intervention was assessed by an Intervention Satisfaction Scale, adapted from other studies using the MSC intervention (Campo et al., 2017), and the Intervention Acceptability Framework (Sekhon et al., 2022).

Secondary Outcomes

Our secondary outcomes included a standardized battery of pain, mental health, and general functioning using validated outcome measures as well as assessment of sleep duration and sleep efficiency.

Pain-Related Outcomes Whereas MSC does not aim to directly reduce primary suffering (i.e., physical pain), it targets secondary suffering (i.e., response to pain) by reducing critical self-judgments and fostering functional gains in spite of pain (Edwards et al., 2019). For this reason, in addition to pain intensity, we assessed pain interference to capture the functional impact of pain on daily life, pain acceptance to evaluate the capacity to engage in meaningful activities despite pain, and pain catastrophizing to measure maladaptive cognitive-emotional responses to pain, all of which are key targets of the MSC intervention's emphasis on reducing secondary suffering. Pain interference, a measure of the degree of interference that pain has on one's daily activities, was as assessed by the Patient-Reported Outcomes Measurement Information System-Pain Interference (PROMIS-PI) measure (Amtmann et al., 2010). The PROMIS-PI provides a score between 0 and 100, where a score of 50 represents the mean score of a general reference sample, and a score of > 50 represents more pain than that general reference sample. Pain acceptance, a measure of one's willingness to reduce pain control and focus instead on daily activities and personal goals, was assessed by the Chronic Pain Acceptance Questionnaire—Revised (CPAQ) (McCracken & Eccleston, 2006). The CPAQ is scored on a scale of 0–120, with higher scores indicating higher levels of acceptance. Pain catastrophizing, which measures magnification, rumination, and helplessness, was assessed by the Pain Catastrophizing Scale (PCS) (Sullivan et al., 1995). The PCS is scored from 0 to 52, with higher scores representing higher pain catastrophizing. Pain severity was assessed by an 11-point scale numerical rating scale ranging from 0 (no pain) to 10 (worst possible pain) (Ferreira-Valente et al., 2011).

Mental Health Outcomes Self-compassion was assessed by the Self-Compassion Scale (SCS) (Neff, 2003). The SCS is scored on a scale of 1–5, with higher scores representing higher self-compassion. Symptoms of depression were assessed by the PROMIS depression tools (Pilkonis et al., 2011). As with the PROMIS-PI, the PROMIS depression tool measure is scored between 0 and 100, with 50 representing the mean score for a general reference sample, and scores > 50 representing higher depression than that sample. Emotion regulation, defined as the ability to modulate one's own emotion and to act in desired ways, was assessed by the Difficulties in Emotion Regulation Scale (DERS) (Gratz & Roemer, 2004). On the DERS, higher scores are suggestive of greater problems with emotion regulation. Experi-

ential avoidance, defined as the tendency to escape or run away from internal experiences (i.e., sensations, emotions, thoughts, memories) that may cause discomfort or suffering, was assessed by the Brief Experiential Avoidance Questionnaire (BEAQ) (Gámez et al., 2014). Higher scores on the BEAQ indicate more experiential avoidance.

General Functioning Days missed from school/work was determined by asking the participants the number of days they missed in the 8 weeks preceding the MSC intervention at baseline, and the number of days missed during the 8 weeks of the intervention. We chose 8 weeks for the timeline of days missed, as to differentiate between baseline and post-assessment.

Sleep Sleep efficiency and sleep duration were objectively measured using GT9X Link watches (ActiGraph Corp, Pensacola, FL, USA) (Budig et al., 2022). The participants were asked to wear the watch on their non-dominant wrist in the week preceding the intervention and the last week of the intervention. Participants were asked to track their daily activities, and times they removed the watch on a wear log that was provided to them. They were also asked to complete a daily sleep diary, which tracked the time they went to bed, the time they woke up, and a self-assessment of the subjective quality of their sleep.

Exploratory Outcomes

Our exploratory outcome was the feasibility of collecting objective sleep data using the actigraph watches among adults with OI, defined as the proportion of participants from whom we were able to successfully obtain sleep actigraphy data.

Safety and Risk Considerations

We assessed mental health outcomes through participants self-reports directly into the REDCap database. If a score on the PROMIS Depression scale suggested severe depression (score $\geq$ 70 (Kroenke et al., 2020)), the principal investigator (PI) and study coordinator were alerted. The PI had to contact the participant's primary care provider within 5 days to ensure appropriate follow-up.

Statistical Analysis

Given that this is a pilot study, we chose a convenience sample of 8 adults to allow for an assessment of the program's feasibility.

Descriptive statistics (means and standard deviation [SD] or median, interquartile range [IQR] and range, as appropriate) were used to describe our cohort, the feasibility data, and the patient-reported outcome measures, including intervention satisfaction and acceptability.

Results

A total of 17 participants were contacted for the study. Of the 17 participants contacted, 5 (29%) did not have chronic pain and were deemed ineligible. Among the remaining 12 eligible individuals, 8 participants (67%) were enrolled. One participant (18-year old female with OI type III) withdrew from the study before the end of the intervention due to feeling that she could not relate to the older participants and therefore did not engage with the program material. This participant is not included in the main analysis, but is included in Appendix A. Seven participants completed the MSC intervention, assessments at baseline and post-intervention (5 individuals with OI type I and 2 with OI type IV). Figure 1 depicts the study flowchart. A summary of participants' characteristics and self-reported demographic information is presented in Table 2. No safety events occurred (i.e., no individual had a PROMIS Depression score suggesting severe depression).

Primary Outcomes

The delivery of the MSC program was found to be both feasible and acceptable for adults with OI. Eighty-six percent (6/7) of participants met the definition for good attendance of the MSC classes. Less than 2% of the questions on various assessments were left unanswered by the participants. From our intervention satisfaction scale, 86% (6/7) of participants found the MSC program to be acceptable, with the other participant indicating a neutral option.

Participants generally provided positive feedback on the structure and delivery of the MSC course. Regarding session duration, 4 out of 7 participants (57%) agreed that two hours per session was appropriate, while 1 participant (14%) strongly agreed, 1 (14%) disagreed, and 1 (14%) remained neutral. Similarly, for session frequency, 4 participants (57%) agreed that one session per week was the right amount, 1 (14%) strongly agreed, 1 (14%) disagreed, and 1 (14%) neither agreed nor disagreed. The online/virtual format was well received, with 4 participants (57%) agreeing that they liked this format, 2 (29%) remaining neutral, and 1 (14%) disagreeing. In terms of course difficulty, 5 participants (71%) rated the material as "just right," while 1 (14%) found it slightly difficult and 1 (14%) found it slightly easy. Our participants reported a mean $\pm$ SD of 3.5 out of 5 $\pm$ 0.4 on the Intervention Acceptability Framework.

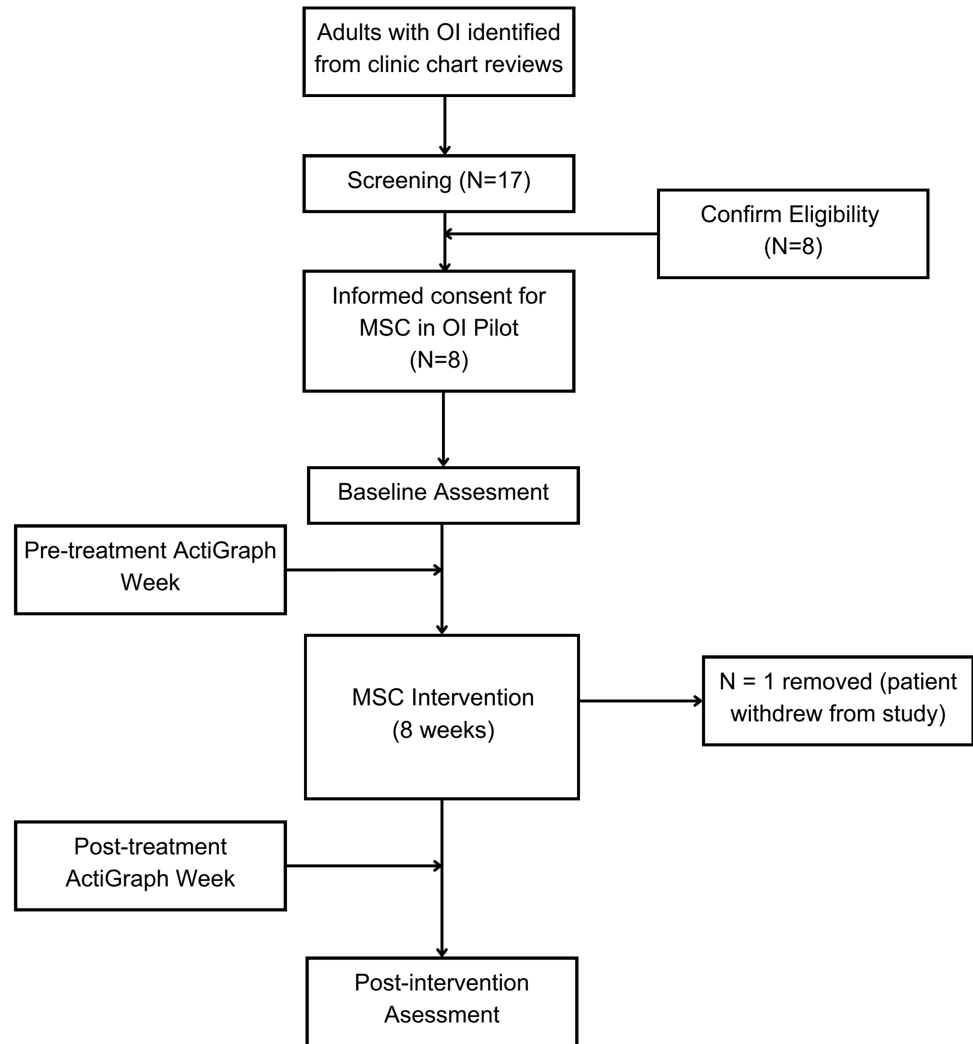

Fig. 1 Study design. A total of 8 adults were enrolled in the eight-week MSC intervention. Baseline and post-intervention assessments were conducted one week prior to and post-intervention, respectively

Secondary Outcomes

Table 3 shows a summary of the outcome measures at baseline and post-intervention. Figure 2 shows bar charts of the baseline and post-intervention results for all questionnaires. We observed a mean improvement in pain interference scores (Table 3).

Participants exhibited improvements above the minimal important change (MIC) across the three pain-related measures with validated MIC: PROMIS Pain Interference, Pain Catastrophizing Scale, and Pain Numeric Rating Scale. For the PROMIS Pain Interference, 71% (5 out of 7) of participants achieved a reduction above the MIC (≥ 3 points) (Beaumont et al., 2021; Terwee et al., 2021). For the Pain Catastrophizing Scale, 40% (2 out of 5, among those with a baseline score above 0) demonstrated an improvement above the MIC (≥ 6 points) (Suzuki et al., 2020). Lastly, when evaluating self-reported pain intensity using the Pain Numeric Rating Scale, 43% (3 out of 7) of participants reported reduction above the MIC (≥ 2 points) when considering the highest scores for worse pain at baseline (Farrar et al., 2001).

Exploratory Outcomes

Objective collection of sleep data using the actigraph watches was considered feasible, with all participants (100%) wearing the watches during sleep periods for the week before and the last week of the MSC intervention. Sixteen sleep periods were adjusted manually, out of the total 112 sleep periods for all participants (14%). It is possible for a participant to have > 1 recorded sleep period per night, as an awakening of ≥ 15 min in the middle of the night would result in that night counting as 2 sleep periods. Manual adjustments were done in cases where the watch data and sleep diaries were not matching and adjusted on the actigraph database to the subjective report by participants.

Table 2 Description of the Cohort (N = 7)

Age (years), median (IQR, range)	41 (6.5, 25–53)
OI Type, n (%)	
Type I	5 (71)
Type IV	2 (29)
Gender, n (%)	
Male	2 (29)
Female	3 (43)
Gender diverse	1 (14)
Prefer not to answer*	1 (14)
Marital Status, n (%)	
Married	5 (71)
Common law	2 (29)
Years of education, mean (SD, range)	17 (2, 15–20)
Employment Status, n (%)	
Employed full-time	5 (71)
Self-employed	1 (14)
Prefer not to answer*	1 (14)
Race, n (%)	
White	6 (86)
Prefer not to answer	1 (14)
Pain Characteristics	
Numeric pain rating, mean (SD, range)[a]	5 (2.3, 0–6.2)
Pain duration (years), median (IQR, range)	10 (15, 4–30)
Pain Regions (number of participants), n (%)[b]	
Back	4 (57)
Neck	2 (29)
Hand	2 (29)
Legs	3 (43)
Feet	1 (14)
Bones and joints	6 (86)
Days missed from work in previous 8 weeks, median (IQR, range)	0 (3, 0–14)

* "Prefer not to answer" indicates that the participant chose not to disclose their response to a question, typically due to its sensitive nature

[a] Presented as a mean of the highest pain ranges

[b] Participants were able to select multiple pain regions

Discussion

In this study of an MSC program for adults with OI and co-occurring chronic pain, we found the MSC intervention to be both feasible and acceptable. We observed a decrease in pain interference, experiential avoidance, pain catastrophizing, absolute pain levels, and days missed from work, as well as an improvement in pain acceptance, self-compassion, and emotion regulation. Using actigraphy to measure sleep in adults with OI was feasible. These findings suggest that the MSC program holds promise as a potential intervention for addressing pain interference in adults living with OI.

We have shown that the MSC program is feasible among adults with OI, in alignment with findings from other studies using MSC in conditions such as adult cancer survivors and individuals with subsyndromal depression (Bluth et al., 2024; Campo et al., 2017). Recognizing that we had a small sample size, we nonetheless saw 86% of participants meet our definition of good attendance (attending at least 6/8 sessions), which is consistent with others who have shown average attendance rates between 70 and 80% (Bluth et al., 2024; Campo et al., 2017). When accounting for the participant who withdrew, our effective feasibility and acceptability rates were both 75%, instead of 86%. The MSC intervention was previously defined as feasible if > 75% of participants completed at least 6 of the 8 classes, in line with other studies using the MSC intervention (Brooker et al., 2020). Previous research indicates that acceptance rates of 75% reflect good acceptability of mindfulness-based interventions in various patient populations (Ljungvall et al., 2022; Ruskin et al., 2017; Winger et al., 2022). Therefore, even the 75% feasibility and acceptability rate observed in this study when considering the participant exclusion support the feasibility and positive reception of the MSC intervention among patients with OI.

Most participants in the study reported a reduction in pain interference. Our findings align with previous studies showing that MSC interventions have led to decreased pain interference in adults with chronic pain, compared to cognitive-behavioral therapy (Torrijos-Zarcero et al., 2021a, 2021b) or wait-list control (Buhrman et al., 2023). While the mechanisms by which MSC may reduce pain interference have not been studied in any chronic pain population, our findings suggest three mechanisms (experiential avoidance, emotional regulation, and pain acceptance) that could be explored in future studies of MSC in individuals with OI.

Our pilot study has several strengths. To our knowledge, this study was the first to use a psychological intervention to address pain-related outcomes in a population with OI and chronic pain. We incorporated a wearable device, the GT9X Link watches, to collect outcome measures. This objective method to collect sleep data and measure activity levels provides another layer of analysis and metrics while also offering a comprehensive means to assess sleep quality and efficiency in individuals with OI. The successful collection of pain-related outcomes and sleep data emphasizes the feasibility of this methodological approach, as well as the application of MSC to individuals with OI.

Limitations of our study include the small convenience sample of seven participants. While our results suggest high acceptability and positive engagement, these outcomes should be interpreted cautiously. Our limited sample size restricts our ability to assess the broader variability in response and to draw robust conclusions about efficacy. Future studies with larger sample sizes are needed. Another limitation of this study is the

Table 3 Summary of outcome measures at baseline and post-intervention

Outcome	Baseline	8 Weeks
PROMIS Pain Interference, mean (SD)	55.9 (5.5)	50.0 (7.3)
Pain Catastrophizing Scale, median (IQR, range)	9 (10, 0–22)	5 (11, 0–24)
Pain Numeric Rating Scale, mean (SD)	5 (2.3)	3.7 (2.1)
PROMIS Depressive Symptoms, mean (SD)	49.3 (10.1)	48.8 (6.8)
Difficulties in Emotion Regulation Scale (DERS), median (IQR, range)	50 (30.5, 36–96)	44 (6, 32–61)
Brief Experiential Avoidance Questionnaire (BEAQ), mean (SD)	41.6 (15.7)	33.7 (9.4)
Days Missed, median (IQR, range)	0 (3, 0–14)	0 (0, 0–2)
Chronic Pain Acceptance Questionnaire – Revised, median, (IQR, range)	67 (6.5, 51–72)	69 (5.5, 63–74)
Self-Compassion Scale Short Form, median (IQR, range)	3.0 (0.7, 2.6–3.9)	3.5 (0.3, 3.0–4.3)
Sleep efficiency (%), mean (SD)	87.4 (6.1)	85.2 (6.3)
Sleep duration, minutes, mean (SD)	384 (48)	390 (45)

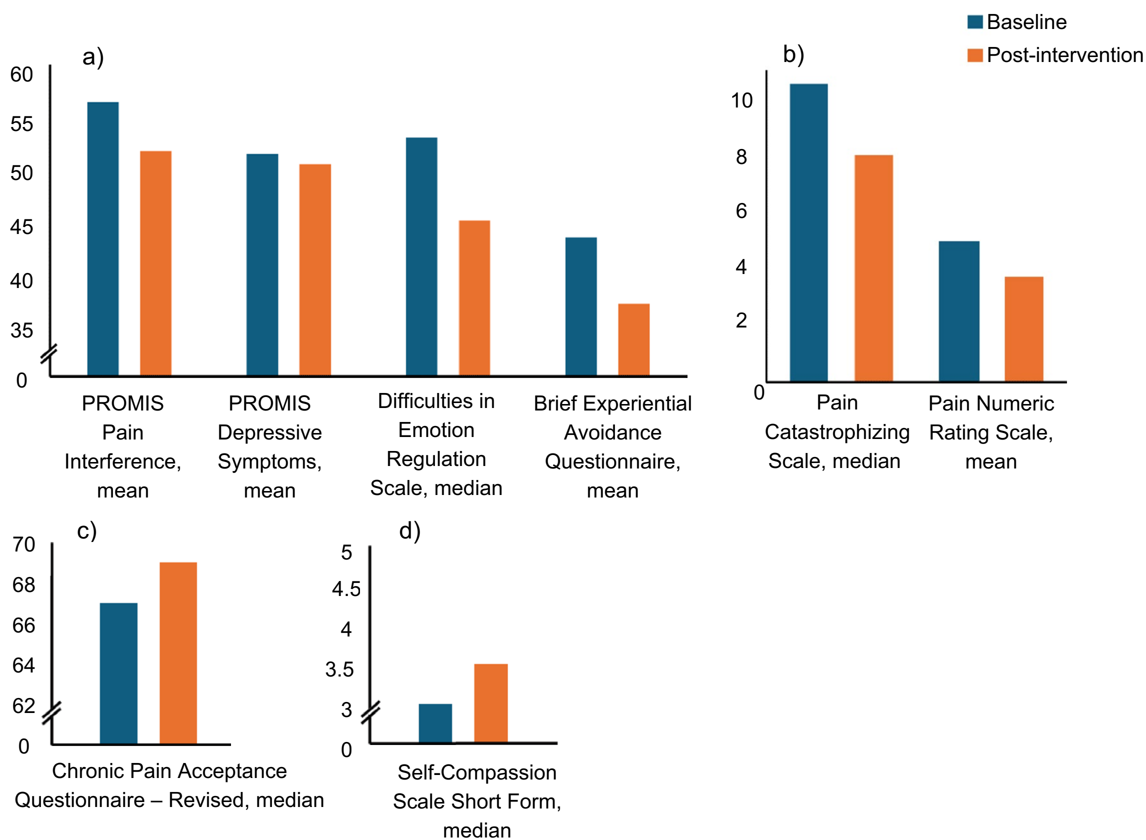

Fig. 2 Outcome measures at baseline and post-intervention. **a** and **b** represent outcome measures at baseline and post-intervention where a decrease represents an improvement. **c** and **d** represent outcome measures at baseline and post-intervention, where an increase represents an improvement

lack of a comparison group, which prevents predictions about the efficacy of MSC in reducing pain interference and improving well-being among adults with OI. The study only included individuals with OI types I and IV; however, pain interference and pain are independent of OI types, and we do not expect that exclusion of other OI types would alter the results. The short duration of the pilot study also prevents a comprehensive understanding of the long-term effects of the MSC program. Finally, one participant withdrew before the end of the study. We performed a sensitivity analysis including the available data from this participant (Appendix A) which shows that the direction of our conclusions do not change. These limitations

underscore the necessity for future larger research on such treatment to address chronic pain in OI.

Conclusion

Our preliminary study suggests that the MSC intervention is both feasible and acceptable among adults with OI and co-occurring pain. Larger definitive efficacy trials should be conducted to confirm and further delineate our findings.

Supplementary Information The online version contains supplementary material available at https://doi.org/10.1007/s10880-025-10092-2.

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Author Contributions Amena Sediqi contributed toward writing—original draft and data collection. Roya Al-Khalili contributed toward writing—original draft and data collection. Saunya Dover contributed toward writing—review and editing, methodology, data collection and curation, analysis, and project administration. Corien Peeters contributed toward intervention delivery. Adam Khalif contributed toward data collection and project administration. V. Reid Sutton contributed toward critical review of the manuscript. Frank Rauch contributed toward critical review of the manuscript. Brendan Lee contributed toward critical review of the manuscript. Eric A. Storch contributed toward critical review of the manuscript. Marie-Eve Robinson contributed toward conceptualization, writing—review and editing, methodology, data curation, and funding acquisition. All authors have approved the final version of the manuscript.

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Data Availability No datasets were generated or analyzed during the current study.

Declarations

Competing Interests Saunya Dover reports full-time employment at Canada's Drug Agency – Agence des Médicaments du Canada outside the submitted work. Frank Rauch reports study grants to institution from Mesentech, Ultragenyx, and Kirin. Brendan Lee reports consultancy fees from Biomarin and study grants to institution from Sanofi. Eric Storch reports receiving research funding to his institution from the Ream Foundation, International OCD Foundation, and NIH. He was a consultant for Brainsway and Biohaven Pharmaceuticals in the past 12 months. He owns stock less than $5000 in NView (for distribution of the Y-BOCS and CY-BOCS). He receives book royalties from Elsevier, Wiley, Oxford, American Psychological Association, Guildford, Springer, Routledge, and Jessica Kingsley. Marie-Eve Robinson reports study grants to institution from Ascendis Biopharma, Ipsen Biopharmaceuticals, and QED therapeutics; consultancy fees to institution from Ultragenyx and Ipsen Biopharmaceuticals.

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