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Citation

Leerling, A. T., Ueckert, D. A., Giesen, F. J. van der, Vliet Vlieland, T. P. M., & Winter, E. M. (2024). Role of physical therapy in adult chronic non-bacterial osteitis: patients' and therapists' perspectives. *Scandinavian Journal Of Rheumatology*, 53(5), 335-344.
doi:10.1080/03009742.2024.2352965

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To cite this article: AT Leerling, DA Ueckert, FJ Van der Giesen, TPM Vliet-Vlieland & EM Winter (2024) Role of physical therapy in adult chronic non-bacterial osteitis: patients' and therapists' perspectives, *Scandinavian Journal of Rheumatology*, 53:5, 335-344, DOI: [10.1080/03009742.2024.2352965](https://doi.org/10.1080/03009742.2024.2352965)

To link to this article: <https://doi.org/10.1080/03009742.2024.2352965>



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Role of physical therapy in adult chronic non-bacterial osteitis: patients' and therapists' perspectives

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Objectives: Chronic nonbacterial osteitis (CNO) is a rare bone disease causing pain and functional impairment. We aimed to explore the application of physical therapy (PT) for adult CNO from the patients' and primary care therapists' perspective.

Method: Cross-sectional study among the Dutch adult CNO cohort (1992–present). A survey on PT-use for axial spondylarthritis was adapted for CNO. Patients using PT in the past 2 years (recent PT-users) were asked about modalities, perceived effects, satisfaction, and preferences for provision. Their current physical therapists were invited to complete a self-developed survey covering therapy details and educational preferences.

Results: 80/199 invited patients and 14/16 invited therapists completed the survey respectively. 41 (51%) patients used PT for CNO in the preceding 2 years, 14 (18%) used PT >2 years back, and 25 (31%) never used PT. Recent PT-users (n=41) reported diverse treatment modalities, involving massage (61%), joint mobilizations (44%), breathing exercises (49%), muscle strengthening (32%) and counselling through of home-exercises (46%) and pain education (32%). 64% of all patients desired greater emphasis from physicians regarding the role of PT. Most therapists would appreciate referral letters with CNO-specific information (93%), and consultations with specialized therapists (86%).

Conclusions: In this study, two-third of adult CNO patients had -ever or recently- used PT, which involved variable therapeutic modalities. Most patients were receptive to a larger role of PT in CNO-management and most therapists preferred CNO-specific information to optimize their care. These findings provide a foundation for the development and systematic evaluation of CNO-specific PT.

Chronic non-bacterial osteitis (CNO) is a rare musculoskeletal disease spectrum characterized by sterile bone inflammation (1). Historically, a variety of names has been used for this condition, including synovitis, acne, pustulosis, hyperostosis, osteitis (SAPHO) syndrome, and chronic recurrent multifocal osteomyelitis (CRMO), but a recent consensus initiative opted to use the uniform label of CNO (2). Adults with CNO mainly present with bone pain and swelling, but symptoms may extend to stiffness of the adjacent joints and limited range of

motion, the pattern being dependent on the skeletal distribution of lesions. In adult CNO, the most commonly affected bones include the clavicles, ribs, and sternum, which typically results in pain, reduced mobility, and dysfunction of the shoulder girdle (3). Although isolated osteitis is most common, CNO patients may present with a variety of additional inflammatory features, including pustulosis palmoplantaris, psoriasis, acne, arthritis, sacroiliitis, and enthesitis, suggesting a clinical overlap with axial spondyloarthritis (axSpA) and psoriatic arthritis (4–6). Much like these conditions, CNO usually follows a chronic course characterized by recurring phases of remission and exacerbation. Radiographically, CNO tends to progress over time, with gradual development of structural changes, including sclerosis, hyperostosis, erosions, and ankylosis (7, 8), all of which are associated with a high disease burden.

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Received 8 February 2024; Accepted 6 May 2024

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DOI: <https://doi.org/10.1080/03009742.2024.2352965>

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Owing to a lack of awareness of CNO and the non-specific early symptoms, the diagnostic delay averages 5 years. During this extended period, patients often consult multiple healthcare professionals and are frequently misdiagnosed with conditions such as Tietze's syndrome, shoulder pathology (e.g. subacromial bursitis), or functional pain syndromes (9–11). Upon diagnosis, clinical management of CNO remains challenging because of the lack of evidence-based treatments and clinical guidelines. Current treatments are solely prescribed off-label, and include non-steroidal anti-inflammatory drugs (NSAIDs), intravenous bisphosphonates, and tumour necrosis factor- α inhibitors (12–15).

Since CNO has a substantial impact on daily functioning and physical activity (16–18), some patients engage in physical therapy (PT) in addition to their pharmacological treatment plan. While no studies exist that support the role of PT in CNO specifically, PT is generally regarded as a valuable part of overall management in other, better characterized musculoskeletal diseases. Consideration of PT is recommended in clinical guidelines for inflammatory arthritis and axSpA in particular, based on the results of clinical trials on active exercise therapy and expert opinion (19–23). Yet, the use of PT in CNO has only been scarcely described in the literature, and there are no data regarding its effects on symptoms, functioning, and daily life (10, 24). We therefore explored the current application of PT in CNO in the primary care setting, evaluated patients' characteristics associated with PT use, and assessed patients' perceived effects and preferences regarding provision of PT. In addition, we surveyed their therapists' perspectives on treatment goals, modalities, and educational and referral preferences.

Method

Study design, participants, and setting

We performed a survey study at the Leiden University Medical Center, which serves as the Dutch national expert and referral centre for CNO. All study reporting complies with the Consensus-Based Checklist for Reporting of Survey Studies (CROSS) guidelines (25). The study does not fall within the remit of the Medical Research Involving Human Subjects (non-WMO), and received approval from the ethics committee of the Leiden University Medical Center (W2022.007). Adult patients with a clinical and radiological diagnosis of CNO, as established by our multidisciplinary expert team, referred between 1993 and 2022, and with available contact information, were eligible for inclusion. In September 2022, all eligible patients were contacted digitally and invited to complete a set of surveys covering the themes of PT and pain profiles, the latter of which is reported elsewhere (26). Patients who actively filled out the surveys (responders) provided written

consent for the use of their corresponding clinical data. Patients who did not complete the surveys (non-responders) were asked if they objected to the use of their medical data for comparison with the responder group. All patients who used PT at the moment of survey completion were asked for consent to invite their therapist to complete a survey. Patients and therapists were reminded twice, with survey closure in January 2023.

With respect to the setting, management at our centre includes initial treatment with NSAIDs at the maximum tolerated and approved dosage, and if insufficiently effective, off-label intravenous pamidronate. Furthermore, CNO patients can receive PT either via referral by their attending physician or through self-referral, as PT is directly accessible in the Netherlands. Importantly, PT is not covered by the mandatory basic insurance, except for certain conditions (which do not include CNO). Consequently, reimbursement is contingent upon whether patients have supplementary insurance and, if so, the extent to which their insurance covers the number of sessions per year.

Surveys

A 21-question survey was developed, containing self-developed questions and questions from an existing survey on PT use in axial spondyloarthritis (axSpA) (27). These were adapted by two of the authors (AL and FG), to align more closely with the problems and management of patients with CNO. A table of question topics, question types, and answer options is given in S1 in the Supplementary material. In brief, questions covered the present and past use of PT, with past use limited to the preceding 2 years, including reasons for non-use if applicable. For patients who had used PT in the past 2 years or were currently using it (recent PT users), further questions addressed the route of referral, duration and frequency of PT, concomitant medication use, and reimbursement. Specific modalities of PT were questioned and categorized as active exercises (muscle strengthening, aerobic, range of motion, functional, breathing and relaxation exercises), manual treatment (massage, joint mobilizations, joint manipulations), physical modalities (dry needling, thermal therapy, kinesiotaping, electrotherapy), and counselling/education (home exercises, pain education, sports education and physical activity promotion, recommendations for stature and working position) (28). Further questions covered the moment of diagnosis of CNO, alternative working diagnosis during PT use, perceived effects, overall satisfaction, and preferences on the location and organization of PT. The final survey was tested with the research team and by three patients, and improved as recommended (see Supplementary material S2 for the full version). The therapist survey was largely self-developed (by AL and FG) and piloted

independently by DU (at the time holding a parallel position as a primary care physical therapist) (see Supplementary material S1 for the table with question topics, types, and answer options, and S2 for the full version). Topics included professional experience, experience with rheumatic musculoskeletal diseases, chronology of CNO diagnosis and referral, duration of PT, use of specific modalities, primary and secondary therapeutic goals, and the perceived usefulness of specific educational initiatives.

Ancillary data collection

We retrieved data from the medical records of both responding and non-responding patients who had not objected to the use of their data. These included age (years), sex (male/female), autoinflammatory comorbidity (yes/no), smoking habit (active smoker/not active smoker), body mass index (BMI), educational level (lower, defined as International Standard Classification of Education level ≤ 5 ; higher, defined as level 6–8), work absence due to CNO (any; yes/no), physical activity (none at all, daily walking or cycling for 30 min, sport for > 30 min once or twice per week, sport for > 30 min more than twice per week), number of bones involved, skeletal distribution pattern (anterior chest wall only, anterior chest wall and spine, anterior chest wall and mandible, spine only), and presence of compromised shoulder mobility, as defined by active abduction < 180 degrees (yes/no). For responders specifically, recent patient-reported outcomes were collected, including CNO-related pain scores on the Brief Pain Inventory (BPI) (numerical rating scale 0–10), and shoulder function, measured with the Shoulder Function Assessment (SFA) (total score ranging from 0 to 70) (16–19). To evaluate which patient characteristics were associated with PT use in adult CNO, patients were stratified according to ‘use’ and ‘non-use’.

Statistical analysis

Categorical data are displayed as n values with valid percentages, whereas continuous variables are expressed as mean $\pm$ standard deviation or mean with range. To assess the differences between responders and non-responders and between recent PT users and non-recent users, chi-squared tests or Fisher’s exact test, unpaired t-tests or one-way analysis of variance (ANOVA), and Mann–Whitney U-tests or Kruskal–Wallis tests, for categorical data, parametric numerical data, and non-parametric numerical data, respectively, were used. Statistical analyses were performed using SPSS Statistics version 25 (IBM Corp., Armonk, NY, USA). Open text questions were analysed by two authors (AL and DU), categorizing responses independently (eventual categorization consensus based), and results are shown as n (valid percentage).

Results

Patient characteristics of responders and non-responders

In total, 80 out of the 199 patients who were approached completed the survey (response rate 40%) (Figure 1). Responders and non-responders had similar sociodemographic characteristics, including sex, age, BMI, autoinflammatory comorbidities, smoking habits, work absence, and physical activity patterns, and disease characteristics, including diagnostic delay, duration, comorbidities, skeletal distribution pattern, CNO-related pain scores on the BPI, and shoulder function, as assessed by the SFA (see Supplementary material S3). The proportion of patients with a high educational level was higher in the responder group (45% vs 30%), but not to a statistically significant extent.

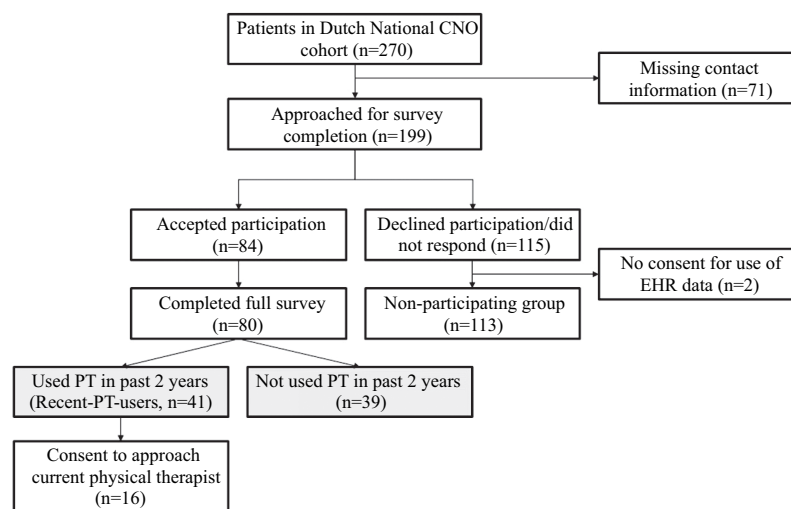


Figure 1. Overview of the study inclusion process. CNO, chronic non-bacterial osteitis; EHR, electronic health record; PT, physical therapy.

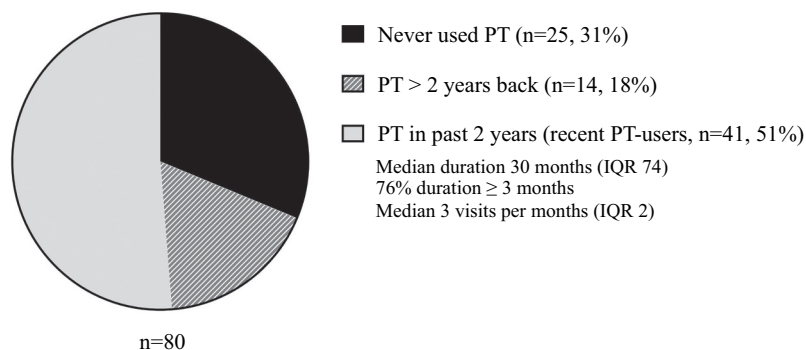


Figure 2. Use of physical therapy (PT) in adult chronic non-bacterial osteitis among responding patients (total n = 80). IQR, interquartile range.

PT use among adult CNO patients

Of the 80 responding patients, 41 (51%) had used PT in the preceding 2 years or were currently using it (henceforth referred to as recent PT users) (Figure 2). Among patients who had not used PT in the past 2 years, 14/39 (36%) reported that PT treatment prior to this period under study had not been effective, 12/39 (31%) stated that PT had never been recommended by their clinician, and 8/39 (21%) indicated that they did not need PT. Another 6/39 (15%) answered that PT was too expensive. Of the recent PT users, 33/41 (80%) had already been diagnosed with CNO upon the initiation of therapy, whereas in eight patients (20%) PT was initiated during the period of diagnostic delay. For seven of these eight patients, the therapist referred them back to the general practitioner because of a suspicion of inflammatory pathology. Over half (22/41, 54%) of recent PT users started PT through self-referral. The median treatment duration was 30 months [interquartile range (IQR) 74], with a median of 3 (IQR 2) visits per month. Moreover, 31/41 patients (76%) reported a treatment duration of 3 months or longer, indicating the chronic nature of the complaint. Of recent PT users, 15/41 (37%) had already quit therapy owing to perceived inefficacy (n = 8), high costs (n = 2), time and logistics (n = 1), or remission of CNO (n = 3). With respect to the costs, a minority of 18/41 (44%) of recent PT users had their expenses partly or fully reimbursed by additional healthcare insurance. Comparing recent PT users to non-recent PT users (Table 1), recent PT users were more likely to be female, and tended to be younger [mean difference 5 (95% confidence interval -0.6 to 10.3) years, $p = 0.084$], and to have a higher educational level (58% vs 30%, $p = 0.054$). Other sociodemographic and disease characteristics were not statistically significantly different between groups. The proportions of recent PT users who were concurrently treated primarily with NSAIDs and bisphosphonates were 28/41 (68%) and 17/41 (42%), respectively.

Recent PT users reported a variety of modalities (Figure 3), of which massage and breathing exercises were most frequently applied (reported by 25/41, 61%, and 20/41, 49%, respectively). They also received exercises to carry out at home (19/41, 46%), joint

mobilization (18/41, 44%), and muscle strength training (17/41, 42%). Aerobic exercises, relaxation exercises, joint manipulation, thermotherapy and electrotherapy, sports education and physical activity promotion, and stature or positional advice were rarely applied.

Patient-reported effects of PT

Patient-reported effects of PT are visualized in Figure 4. The largest proportion of patients reporting an improvement was seen in the domain of range of motion and pain (19/41, 46%, and 11/41, 28%, vs 19/41, 46%, and 10/41, 27%, reporting a little and major improvement, respectively). For the domains of sport and sleep, the latter often being disturbed by inflammatory and mechanical skeletal pain, the majority of patients reported neither a negative nor a positive effect of PT (23/41, 56%, and 26/41, 63%, neutral effect, respectively). In total, 34/41 (83%) of patients experienced a small or major improvement in at least one domain. In the free-text space, 35/41 patients specified other perceived positive or negative effects of PT (see Supplementary material S4). Here, the notion that 'PT is necessary to prevent muscle stiffness and the feeling of joints becoming "blocked"' was expressed by 17/41 (49%). Also, 5/41 (14%) mentioned that muscle strengthening exercises caused aggravation of pain, and were therefore discontinued or replaced with manual treatment. Overall satisfaction with PT was good, with median score of 8 out of 10. However, satisfaction ranged from 1 to 10, with 4/41 (10%) scoring 4 or lower.

Preferences and needs regarding PT

Of all responders, 36/80 (45%) wished to start or continue PT in the future, 12/80 (15%) did not, and 32/80 (40%) were unsure. For 35/80 (44%) patients, future PT was only an option if it was reimbursed by their healthcare insurance. Also, a majority of 51/80 patients (64%) wished to receive more information on the role of PT for CNO, preferably from their CNO specialist at the expert centre (28/51, 55%), via an information leaflet

Table 1. Patient characteristics of recent physical therapy (PT) users and non-recent PT users.

	Recent PT users (n = 41)	Non-recent PT users (n = 39)	Mean difference (95% CI)	p
Sociodemographic characteristics				
Sex, female	40 (100)	35 (90)	–	0.038
Age (years)	49 (28–79)	53 (27–74)	4.8 (–0.6 to 10.3)	0.084
BMI (kg/m ²)	27 (19–40)	26 (18–34)	1.2 (–5.1 to 2.6)	0.526
Auto-inflammatory comorbidity (any)	9 (23)	14 (36)	–	0.214
Active smoking	6 (17)	9 (24)	–	0.453
Current work absence due to CNO	14 (47)	14 (47)	–	1.000
Higher education	14 (58)	7 (30)		0.054
Physical activity				0.948
None at all	5 (23)	6 (26)		
Daily walking/cycling (30 min)	5 (23)	6 (26)		
Sport > 30 min, 1–2 times/week	9 (41)	9 (39)		
Sport > 30 min, > 2 times/week	3 (14)	2 (9)		
Disease characteristics				
Diagnostic delay (years)	6 (0–29)	6 (0–21)	0.3 (–2.4 to 3.0)	0.826
Disease duration (years)	13 (1–39)	14 (2–40)	0.4 (–3.5 to 4.3)	0.835
Number of bones involved	3 (1–5)	3 (1–6)	0.02 (–0.8 to 0.9)	0.966
Compromised shoulder mobility	18 (58)	15 (52)		0.622
Skeletal distribution				
Anterior chest wall only	26 (90)	27 (87)	–	0.582
Anterior chest wall + spine	1 (3)	1 (3)		
Anterior chest wall + mandible	1 (3)	3 (10)		
Spine only	1 (3)	–		
CNO-related pain scores, assessed by BPI (10 indicates worst pain)				
Maximal pain (0–10)	7.2 ± 2.1	6.6 ± 2.1	0.6 (–1.7 to 0.5)	0.296
Minimal pain (0–10)	3.1 ± 2.0	3.5 ± 2.4	0.4 (–0.8 to 1.6)	0.487
Average pain (0–10)	5.4 ± 1.9	5.2 ± 2.2	0.2 (–1.3 to 0.9)	0.701
Sleep disturbance (0–10)	4.7 ± 3.2	4.9 ± 2.8	0.2 (–1.4 to 1.8)	0.784
Shoulder function, assessed by SFA (70 indicates best function)				
Shoulder function (0–70)	47.3 ± 13.8	51.0 ± 12.7	3.8 (–3.2 to 10.8)	0.285
Pharmacological treatment during PT				
NSAIDs	28 (68)			
Bisphosphonates	17 (42)			
TNF-α inhibitors	5 (12)			
Intra-articular corticosteroids	3 (7)			
Other	9 (22)			

Data are shown as n (%), mean (range), or mean ± sd.

BMI, body mass index; CNO, chronic non-bacterial osteitis; BPI, Brief Pain Inventory; SFA, Shoulder Function Assessment; NSAID, non-steroidal anti-inflammatory drug; TNF-α, tumour necrosis factor-α; CI, confidence interval.

(15/51, 29%), or from a public website (5/51, 10%). Regarding the setting of PT, 55/80 patients (69%) would prefer individual therapy over group therapy, even when group therapy offers contact with fellow patients. As for location, a majority of patients indicated that they preferred PT at a primary care practice close to home (71/80, 89%), rather than visiting a specialized centre with a longer travel distance. A considerable proportion of patients felt that it is very important for their therapist to have expertise in rheumatic diseases in general (67/80, 84%) or CNO specifically (63/80, 79%). A final question leaving room for free-text remarks about PT in general was answered by 35/80 responders (see Supplementary material S4), expressing the need for more awareness of CNO among therapists and the

educational responsibility of the expert centre (6/35, 17%). One participant suggested a combination of short-term PT at a specialized centre followed by an informed referral to primary care as a potential approach to organize PT for people with CNO.

Therapists' perspective

Overall, 16/26 patients using PT at the time of survey completion consented to invite their current therapist for the therapist survey, of whom 14 completed it (10 therapists at bachelor's level, four at master's level). Eight of these 14 therapists had some or much experience with rheumatic musculoskeletal diseases, of whom three had

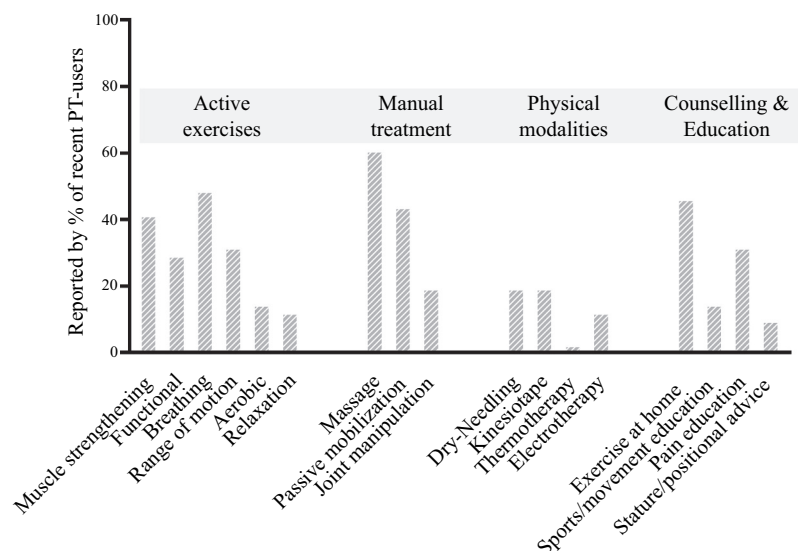


Figure 3. Modalities of physical therapy (PT) in adult chronic non-bacterial osteitis, as reported by recent PT users (total n = 41).

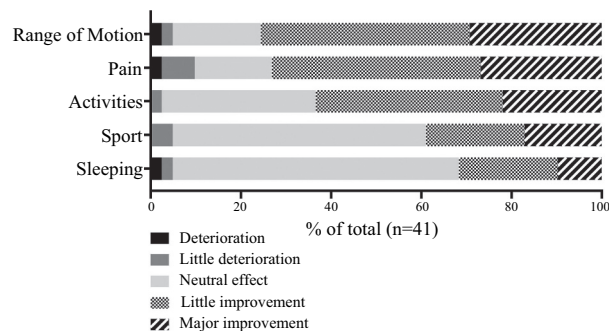


Figure 4. Patient-reported effects of physical therapy in adult chronic non-bacterial osteitis (total n = 41).

specific prior experience with CNO. Primary treatment goals according to the therapist varied, but the most reported were improving joint mobility (n = 5) and muscle function (n = 4). Secondary goals of treatment included joint mobility (n = 9), but also pain education (n = 7), relaxation (n = 7), and work participation (n = 5). Among the array of modalities employed (Figure 5), active exercises aimed at muscle strengthening and range of motion, relaxation, and breathing were common (sometimes or often used by n = 13/14, 13/14, 13/14, and 10/14, respectively). Manual treatment was also used to a considerable extent, especially passive mobilization and massage (sometimes or often used by n = 12/14 and 10/14). In the free-text responses, therapists mentioned additional modalities, such as addressing sleep quality, providing advice on appropriate footwear, and fascial therapy (all n = 1/14). Considering educational needs, 13/14 would value a referral letter with specific information on disease manifestations and prognosis of CNO, and 11/14 would appreciate specific treatment recommendations for the condition. CNO-related information on a public website for healthcare professionals would also be useful, according to most therapists (13/14), as would tailored e-learning

for CNO (12/14) and remote consultation options with a physical therapist experienced in working with patients with CNO (12/14).

Discussion

This study explored the current application of PT in adults with CNO in the primary care setting, patient factors associated with PT use, and preferences and needs regarding PT, from both the patients' and therapists' perspective. We thus aimed to lay the groundwork to further develop the role of this non-pharmacological treatment for this rare disease.

We found that 51% of patients had received PT for CNO in the preceding 2 years, another 18% had used PT > 2 years ago, and 31% had never used PT. This rate of use is only slightly lower than that seen in other inflammatory musculoskeletal diseases such as axSpA, where 83% of Dutch patients were identified as either current or past PT users and only 17% had never opted for this treatment (27). However, for axSpA, PT is recommended as standard in leading clinical guidelines, whereas PT has no formal role in the management of CNO (19). This suggests that CNO patients seek PT despite not receiving standard clinical counselling on its potential value, which may be facilitated by the accessibility of PT via self-referral in the Netherlands.

There are practically no data on the benefits, safety, or outcomes of PT in CNO at present. Only one study documented negative effects of PT in a limited number of CNO patients (10), and one case study discussed a successful and effective rehabilitation (24). However, CNO resembles diseases for which PT is already well established and integrated in clinical guidelines, such as inflammatory arthritis, and specifically axSpA and psoriatic arthritis (23, 29, 30). As in these conditions, CNO patients may present with osteoarticular swelling and stiffness, as well as radiographic signs of erosion and

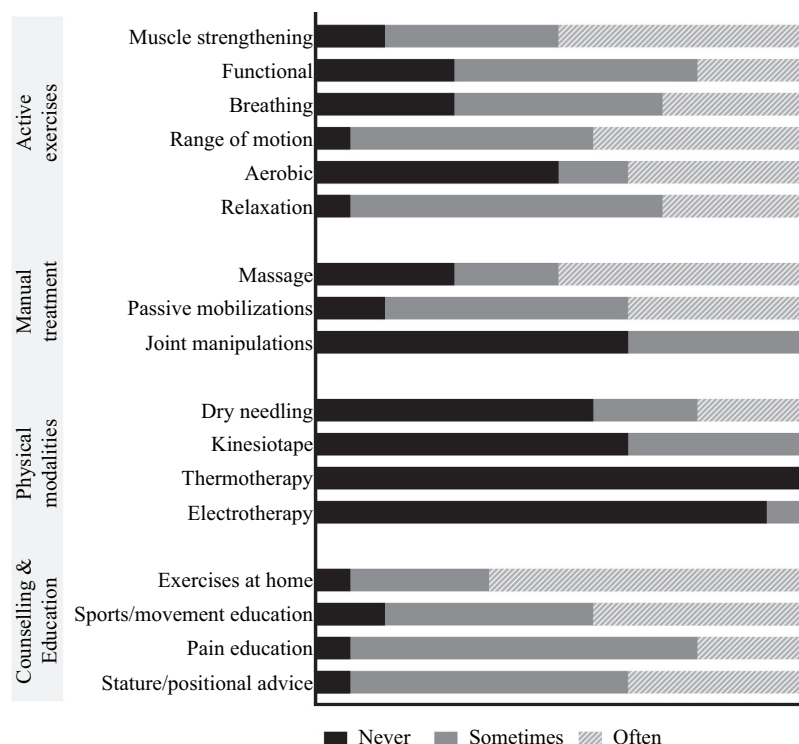


Figure 5. Therapist-reported modalities applied in adult chronic non-bacterial osteitis (total $n = 14$).

new bone formation (5, 9, 16, 31, 32). Considering these similarities, it is reasonable to assume that there is some clinical utility in PT for CNO as well, even when proper effectiveness studies are yet to be conducted. We believe that this should encourage healthcare providers to emphasize PT as part of the comprehensive management of CNO. This viewpoint was reflected in the present study, where 64% of patients expressed a desire for their physicians to pay more attention to this treatment option. While PT may be offered to all CNO patients experiencing functional complaints, it is worth considering that in this study, recent PT users tended to be more often female, younger, and better educated. As a result, male, older, and less well-educated patients should be specifically informed about the potential role of PT.

Regarding the contents of PT for CNO, we found a wide range of treatment goals, as specified by therapists. These included improving the range of motion and muscular strengthening, but also pain education and enhanced relaxation. Regarding range of motion, this may be indeed be a relevant domain in CNO since the disease is characterized by swelling, stiffness, hyperostosis, and soft tissue ossification, all of which may limit mobility (3, 7, 8). Regarding muscle strengthening, we speculate that CNO patients may have lower levels of physical activity owing to their high disease burden, which may result in poor muscle strength and general fitness. A similar tendency towards lower activity levels has been seen in other musculoskeletal diseases as well (33, 34). The fact that pain education emerged as a treatment goal is noteworthy. This may

be due to the fact that CNO patients, and those with musculoskeletal diseases in general, are susceptible to developing chronic pain, for which pain education is a relatively novel intervention (16, 35–37). To achieve the aforementioned goals, PT-using patients reported being treated with a wide variety of therapeutic modalities. These included massage (61%) and joint mobilization (44%) to address range of motion, active muscle strengthening exercises (42%) to improve muscle function, and breathing exercises (49%) to enhance relaxation and/or exercise capacity. This last modality may particularly be applicable in CNO owing to the frequent involvement of the anterior chest wall in the disease, which may result in breathing difficulties due to pain and increased local muscle tension. The PT content found in this study roughly aligns with current PT guidelines for other musculoskeletal diseases, which promote mostly active exercise therapy, possibly combined with manual treatments if applied in a timely manner (27, 38, 39).

We found that 20% of PT-using CNO patients received PT during the period of diagnostic delay, a concept that continues to be a significant challenge in CNO. Adults are diagnosed after an average symptomatic period of 5 years, and longer delays are associated with reduced quality of life and poorer patient-reported outcomes (9, 10). Hence, early recognition is a clinical priority. Our findings indicate that physical therapists may often be among the initial healthcare providers to interact with patients who will later be diagnosed with CNO, particularly since many patients seek PT on their own initiative without having consulted a general

practitioner first (54% in this study). Furthermore, seven out of eight patients receiving PT, yet undiagnosed, reported that they were referred back to their general practitioner because their therapist suspected an inflammatory disease. Although these numbers are small and need confirmation in larger patient groups, they suggest that physical therapists could play a significant role in the early diagnosis of CNO. Disseminating information about this rare condition from expert centres may further enhance this role.

Regarding perceived effects, patients were generally satisfied with PT. Most positive patient-reported outcomes were seen in the domain of range of motion (76% reporting a little or major improvement), implying that stiffness and limited mobility resulting from CNO may be responsive to PT. For the domain of pain, a majority of 73% reported experiencing either a little or major improvement, which is remarkable since pain management in CNO traditionally revolves around pharmacological disease control. These findings suggest that pain in CNO may be further reduced through ancillary PT. Nevertheless, a small group of patients reported negative outcomes after PT, and a considerable proportion of patients expressed neutral effects, implying the preservation of baseline function. Because the natural disease course of CNO is unknown, it remains elusive whether the preservation of function might be considered a positive outcome of PT, postulating that the patient's condition could have worsened without it. However, the present study design cannot evaluate the efficacy of PT in adult CNO, and prospective studies are warranted to comprehensively assess the feasibility and efficacy of PT and exercise programmes.

In terms of awareness of CNO, 79% of patients expressed a desire for their therapists to have a better understanding of their condition, ideally through information provided by the expert centres. In this context, short on-demand educational materials could be developed, as has also been proposed for other rare diseases such as systemic sclerosis (40). These materials can enhance the quality of care and potentially aid in the early detection of CNO if patients use PT during the diagnostic delay period. Likewise, therapists indicated that they would appreciate referral letters from clinicians containing patient-specific information, and to have the opportunity to consult with an CNO-experienced physical therapist. These suggestions could be implemented in current clinical care, aiming to improve communication between CNO healthcare providers and thus overall CNO management.

This study acknowledges potential limitations inherent to cross-sectional survey research, such as the risk of self-selection bias, where those who respond may have different experiences from non-responders. However, these groups were similar in key characteristics, suggesting that our data represent the adult CNO population and their experiences with PT. In long-running

cohorts such as ours, attrition bias may arise from deaths or relocations. However, deaths are uncommon given the relatively young age of CNO onset, and relocations do not typically lead to loss to follow-up since our centre participates in shared-care arrangements for patients living farther away. Another limitation arises from the risk of recall bias, which we minimized by focusing on PT experiences in the past 2 years. This excluded patients who had used PT more than 2 years prior, but had quit owing to a lack of efficacy, from further questions on contents and effects, which may have introduced an overestimation of the overall level of satisfaction. Also, therapists were included by the researchers approaching the patient first, with only 16 of the 26 patients providing consent, inducing some selection bias. Finally, the surveys used in this study were not validated. However, some of the questions were derived from a previously used survey (27), and the final survey underwent thorough detailing and pilot testing to enhance its reliability and relevance.

Conclusion

This study provides the first clinically relevant overview of current PT use in adult CNO. A significant proportion of patients in this study expressed a desire for increased focus on PT in clinical care, while most therapists sought more disease-specific information to enhance their treatment of this rare condition. Future research should explore the efficacy of exercise programmes specifically designed for CNO, to determine the precise role of PT in managing the condition.

Disclosure statement

The authors declare no competing interests pertaining to the current study.

Funding

This research did not receive any specific grant from any funding agency in the public, commercial, or not-for-profit sector.

Data availability statement

The data sets generated during and/or analysed during the current study are available from the corresponding author upon reasonable request.

Ethical approval

This study was approved by the medical ethical review committee associated with the Leiden University Medical Center.

Declaration of generative artificial intelligence (AI) and AI-assisted technologies in the writing process: During the preparation of this work the authors used ChatGPT to improve readability.

After using this tool, the authors reviewed and edited the content as needed and take full responsibility for the content of the publication.

Supplementary material

Supplemental data for this article can be accessed online at <https://doi.org/10.1080/03009742.2024.2352965>

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