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PERSPECTIVES IN REHABILITATION

Development of clinical practice guidelines for rehabilitation after diagnosis for primary bone and soft tissue tumours

Abby McCarthy[a], Lucy Dean[b], Debbie Artis[c], Lynsey Green[d], Maali Khouri[e], Craig Gerrand[a] and Sherron Furtado[a,f]

[a]The London Sarcoma Service, Royal National Orthopaedic Hospital NHS Trust, Stanmore, UK; [b]The Royal Marsden NHS Foundation Trust, London, UK; [c]Leeds Sarcoma Service, Leeds Institute of Oncology, St James' Hospital, Leeds, UK; [d]East Midlands Sarcoma Service, Nottingham University Hospitals NHS Trust (City Campus), Nottingham, UK; [e]UCL Institute of Orthopaedics Library, UCL Library Services, Royal National Orthopaedic Hospital, London, UK; [f]Department of Orthopaedics and Musculoskeletal Science, University College London, London, UK

ABSTRACT

Background and aims: Clinical practice guidelines for rehabilitation in musculoskeletal oncology are lacking. Guidelines should include recommendation statements aimed at optimising care to enhance recovery and quality of life. The project aim was to provide a foundation of best practice based upon expert consensus and evidence.

Design: Consensus-based guidelines supported by systematic literature search.

Methods: This was a consensus-based guideline developed with the support of the British Sarcoma Group (BSG). A group of national rehabilitation experts working within bone or soft tissue sarcoma centres across the UK met from December 2019. Evidence was gathered from a narrative literature review. Recommendations were developed with a variety of stakeholders to achieve consensus.

Results: During the face to face consultation event and literature review seven themes were identified which should guide rehabilitation: (1) access to specialist Allied Health Professionals (AHPs) for treatment; (2) documented referral pathway for specialist AHP care; (3) assessment of individual issues, personal and multi-dimensional (holistic) needs; (4) patient centred care; (5) evidence-based rehabilitation treatment; (6) effective communication and provision of information; (7) patient support through access to other services.

Conclusions: Development of consensus-based, evidence-informed rehabilitation guidelines for those treated for primary malignant musculoskeletal tumours, provides rationale and evidence-based recommendations.

KEYWORDS
Sarcoma; rehabilitation; guidelines; surgery; physiotherapy

IMPLICATIONS FOR REHABILITATION

- Those undergoing limb surgery following a diagnosis of sarcoma should have access to specialised Allied Health Professionals (AHPs) from the point of diagnosis until the long-term to optimise functional outcomes and patient experience.
- Specialist centres and local rehabilitation professionals need to work collaboratively to ensure optimal outcomes following treatment for the management of sarcoma.
- Allied Health Professionals need to embrace holistic individualised assessment and person centred approaches to rehabilitation.
- Although rehabilitation is essential in the early post-treatment phase, it remains important through the pathway to address late effects of treatment and to end of life.

Introduction

Sarcomas are a rare group of malignancies, arising from bone and connective tissues, which can occur anywhere in the body. Their heterogeneity means that treatment and outcomes are highly variable [1,2]. Whilst there are over 100 subtypes of sarcoma, they are broadly separated into those arising from bone or soft tissue [3]. With approximately 5300 new diagnoses of sarcoma in the UK per year [4], sarcomas account for just 1.4% of all cancer diagnoses [4]. Treatment typically involves surgery with or without radiotherapy and/or chemotherapy [1].

Sarcoma survival rates are slowly rising [5], furthermore, medical and surgical advances have expanded the possibility of limb salvage surgery, and increasingly amongst those with comorbidities and frailty. As more people live with the consequences of sarcoma and its treatment, timely access to rehabilitation is critical.

Despite its important role managing treatment consequences and optimising health-related quality of life (HRQoL), neither the National Institute for Health and Care Excellence quality standards nor the European and UK guidelines for the management of bone and soft tissue sarcomas include recommendations for rehabilitation [1,6,7].

Sarcoma survivors are faced with wide ranging physical disabilities including abnormal gait, proprioceptive loss, muscle weakness, altered biomechanics and postural balance, diminished physical activity (PA) and falls. In severe cases, there is significant

CONTACT Abby McCarthy abigail.mccarthy@nhs.net The London Sarcoma Service, Royal National Orthopaedic Hospital NHS Trust, Stanmore, UK

disability, and individuals may be wheelchair or bed bound [8–11] leading to poor quality of life (QoL) [12,13]. This affects psychosocial functioning, and is associated with problems returning to work, education, and employment, with consequent financial burdens [9,11] as well as influencing survival, and risk of comorbidities including obesity, diabetes, heart disease, and depression [14].

In the UK, delivering rehabilitation is challenging, due to geographical variation in facilities [15] and different funding/healthcare models [16]. In the UK, the medical management of sarcoma is co-ordinated and delivered at specialist sarcoma centres, where follow-up may continue for years. Whilst early rehabilitation may be provided at specialist centres, in the longer term, this is delivered closer to home to mitigate travel costs, time commitment, family disruption, and missing work [17,18].

Non-specialist rehabilitation professionals may be unfamiliar with the specific and unique needs of those with sarcoma. With a lack of best practice guidelines, evidence-based assessments and targeted rehabilitation [9] little high-quality information about "living with and beyond cancer" is available and may prevent clinicians from providing high quality rehabilitation [15].

Whilst research into interventions for very rare diseases has become a UK priority in recent years, high quality, evidence-based and holistic, literature regarding rehabilitation in those with sarcoma is scarce but required in order to achieve optimal outcomes, especially given the improved survival rates [19].

In circumstances where evidence is lacking, consensus-based methods offer a valid alternative for producing clinical guidelines. Consensus-based methods can be useful drawing conclusions for developing recommendations. A National Sarcoma UK Survey in 2020 demonstrated that 41% of respondents did not get referred to any rehabilitation services, with only 36% referred to physiotherapy, 17% to emotional support, and 12% to occupational therapy, with major gaps in care [4].

NICE Guidelines [20] emphasise the importance of evidence-based physiotherapy and rehabilitation. Personalised care packages are a priority in the NHS Long Term Plan for Cancer Care [21] and the NHS specialised commissioning specifications [22]. There is a pressing need therefore in the national agenda, to develop novel evidence-based rehabilitation guidelines and expert high-quality care recommendations [23,24].

This is a first guidance document for rehabilitation following a diagnosis of limb/trunk/pelvic sarcoma. It provides a platform to guide service delivery and re-design to improve rehabilitation at a national level and helps providers deliver the National Cancer Survivorship Initiative's five elements: focus on survivorship outcomes; holistic assessments; multiple options for rehabilitation tailored to patients; self-management; and collection of measures of outcome and improving patient experience [25].

Aims and objectives

The primary aim of the rehabilitation guidelines developed in this project is to provide a foundation of best practice based upon expert consensus and available evidence, with the intention they will be reviewed and updated as the evidence base grows.

The secondary objective is to provide a framework for the rehabilitation of those with sarcoma from diagnosis through to end-of-life. Its purpose is not to be prescriptive but to provide evidence-informed guidance to facilitate best practice.

Scope of guidelines

These recommendations apply to adults who have received a diagnosis of pelvis or extremity bone or soft tissue sarcoma. It has been divided into the four key rehabilitation phases as defined by Macmillan Cancer Support [26].

- Diagnosis and care planning
- Treatment
- Post treatment
- Palliative and end of life care

For those undergoing limb amputation, these guidelines should be considered alongside the British Association of Chartered Physiotherapists in limb Absence Rehabilitation (BACPAR) [27,28].

Methods

This national guidance was developed utilising consensus techniques from relevant stakeholders and subject experts in the field and a narrative literature review. The findings from these were synthesised into recommendations using an iterative process, Figure 1 demonstrates a summary of this.

Consensus techniques from national stakeholders

The initial development of these guidelines was informed by a sarcoma rehabilitation consensus meeting held on 5 December 2019. This was chaired and funded by the British Sarcoma Group (BSG), a national body whose membership includes specialist clinicians, nurses, and supporting professionals working within the field of sarcoma in England, Wales, Scotland, and Northern Ireland.

Participants from all UK sarcoma centres were invited to the meeting in Birmingham, which is readily accessible from most areas of the UK. The event was publicised on the BSG website, and an email invitation sent to lead therapists at each UK sarcoma centre. Twenty people from 12 centres attended representing professional groups including Surgeons, Clinical Nurse Specialists, Occupational Therapists, and Physiotherapists.

Attendees were split into small groups, each tasked with pooling guidance documents, studies and clinical practice experience relevant to the four phases of the patient pathway as follows: diagnosis, treatment, after treatment, palliative care, and end of life.

At the end of the meeting, volunteers were asked to join a working group to take forward the information gathered during the day, complete a systematic review and compile the guidance. Four people came forward and formed the core group.

The information collected at the December meeting was collated and organised into themes by the group comprising of four specialised physiotherapists. They compiled and revised this information into a guidance document. The group met online approximately every 8 weeks and each member took a point of the pathway and conducted a narrative literature search to see if there was any further evidence missing. The evidence in musculoskeletal oncology and the wider cancer population was also reviewed and where relevant related to sarcoma. The working group then met to review and confirm the information for each part of the pathway.

In October 2021, the draft guidelines were sent to specialist sarcoma Allied Health Professionals (AHPs) across the UK, providing an opportunity for feedback. They were given the context of

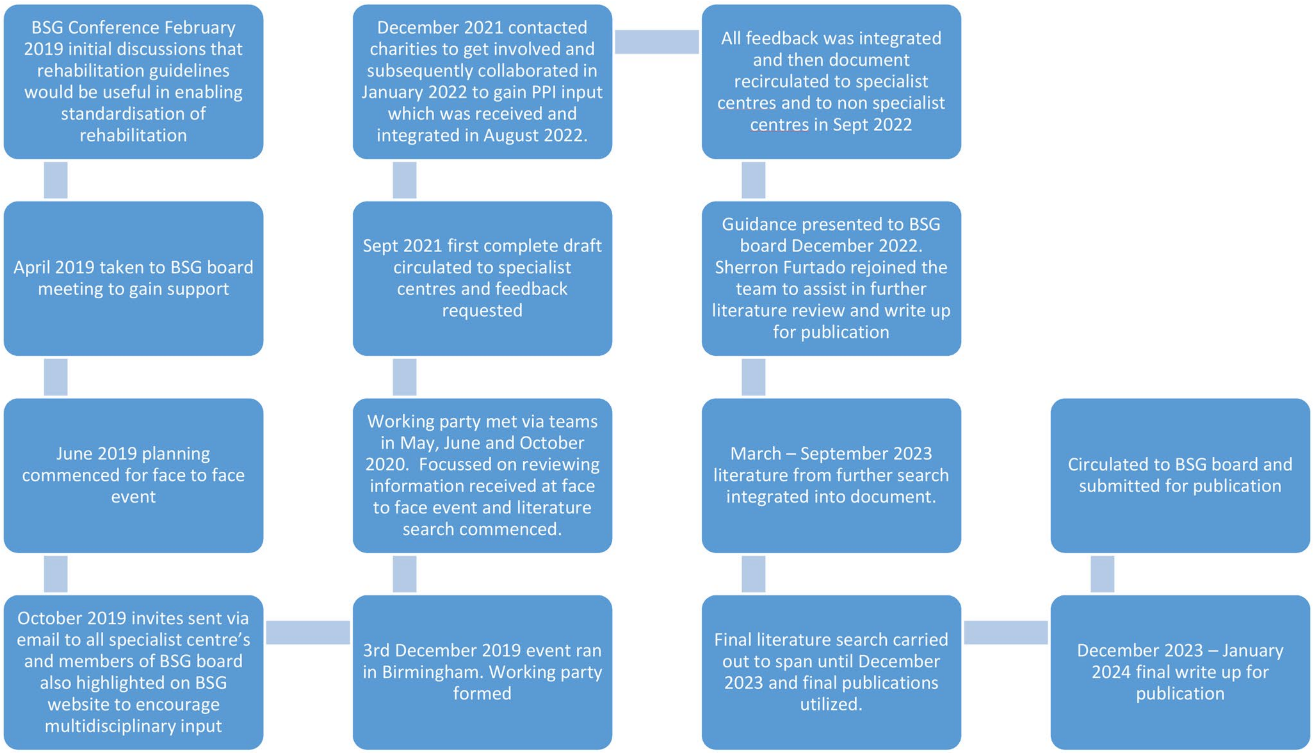

Figure 1. Flowchart of consensus methodology used for BSG national rehabilitation guidelines development.

the original national meeting and then returned comments via email for input.

The next round of consultation was with national sarcoma charities; Sarcoma UK and Bone Cancer Research Trust (BCRT) who met via Microsoft Teams to discuss the document's key results and guidance.

In January 2022, the working group met with BCRT and Sarcoma UK to present the development of the guidelines thus far and to seek review via Patient and Public Involvement (PPI). The national charities, reviewed and provided feedback into the document in February 2022. Sarcoma UK then circulated the guidance to non-specialist therapists nationally in September 2022.

The Sarcoma UK PPI group, comprising of approximately 120 individuals, was invited to participate in the review. Interested members were telephoned by a member of the project team to outline the project aims, answer questions, and request any response was returned via email or post by October 2022.

In another round of consultation, Sarcoma UK sent the guidance to shared care centres, for example, Southampton University Hospital, to gain comment from more general therapists working in non-specialist centres.

Drafts were sent to BSG board in December 2022 and February 2024 for reaction.

The comments from all channels were implemented and relevant changes were made. This resulted in several critical rounds of response and consultation in an iterative process.

Literature review to provide an evidence-base to the guidance

A first comprehensive narrative literature review was performed in December 2019 in PubMed (utilising search terms agreed during a brainstorming session in the initial meeting with national specialists) to identify literature published from 2014 to 2024. This period was selected to ensure articles relevant to recent/modern techniques of treatments (including chemotherapy, radiotherapy, and surgery) in musculoskeletal oncology and applicable rehabilitation approaches were utilised.

The initial search combined four main search terms using the Boolean "AND" operator: (1) bone neoplasms OR soft tissue neoplasms; (2) rehabilitation; (3) functional outcomes (Appendix 1). The titles and abstracts of identified references were screened/reviewed within the national working group and a selection of studies was undertaken. Differences in opinion were resolved by consensus. Additional hand searching of reference lists was also performed. Studies were selected using the eligibility criteria outlined, that the article should contribute to the rehabilitation literature in musculoskeletal oncology. Unpublished grey literature available from national teams was also compiled during this process.

The search was updated in February 2023 and December 2023 in Medline and Embase. This was also repeated in the original PubMed database to update the search and include all relevant articles from the three databases until December 2023. Articles identified were imported into EndNote using the bibliographic management software (Thomson Reuters, Endnote version 20, Toronto, Canada). In summary, the relevant studies were identified using a robust process of comprehensively searching three scientific databases: Medline, PubMed, and Embase.

Synthesis of recommendations: Consensus on the National Guidelines, informed by evidence

After embedding the relevant references into the document at the three phases of the literature searches, this was subsequently circulated amongst stakeholders on multiple instances, to review the document and provide feedback on its content, clinical relevance, accessibility and to ensure that nothing important was omitted. Stakeholders included patients, representatives from Sarcoma UK and BCRT, and AHP's employed by specialist sarcoma centres. Feedback was incorporated into the final document.

The synthesis of clinical treatments and management was conducted using themes generated, which indicated that this varied based on the different phases of the pathway and corresponding categories for which a detailed rationale and each guideline is provided.

The four major clinical management stages identified were:

1. Diagnosis and care planning for people with a diagnosis of bone or soft tissue sarcoma.
2. Rehabilitation during treatment for people with a diagnosis of bone or soft tissue sarcoma.
3. Rehabilitation following end of treatment for people with a diagnosis of bone or soft tissue sarcoma.
4. Rehabilitation during palliative and end of life care for people with a diagnosis of bone or soft tissue sarcoma.

Results

During the initial literature search carried out in 2019 and the update in February and December 2023 to identify relevant papers, three databases were searched: Medline, PubMed, and EMBASE. One thousand four hundred and eighteen papers were identified through the database search; 197 duplicates were removed using the software EndNote x20. One thousand two hundred and twenty-one papers were screened using the selection criteria outlined in the methods section and 882 were excluded. Three hundred and thirty-nine papers were retrieved and 282 were excluded based on title and abstract, English language and irrelevant to the aims of the guideline. The remaining 57 papers were eligible for full text, 55 were excluded, and two papers were included in the study (Figure 2).

The articles identified covered topics such as functional outcomes and risk factors to consider during treatment of these tumours and the rehabilitation approaches available.

During the initial face to face consultation event and then the literature review, seven themes were identified to be important in the delivery of rehabilitation at each point in the pathway:

1. Access to specialist AHPs for treatment.
2. Documented referral pathway for specialist AHP care.
3. Assessment of individual issues, personal and multi-dimensional (holistic) needs.
4. Patient centred care.
5. Evidence-based rehabilitation treatment.

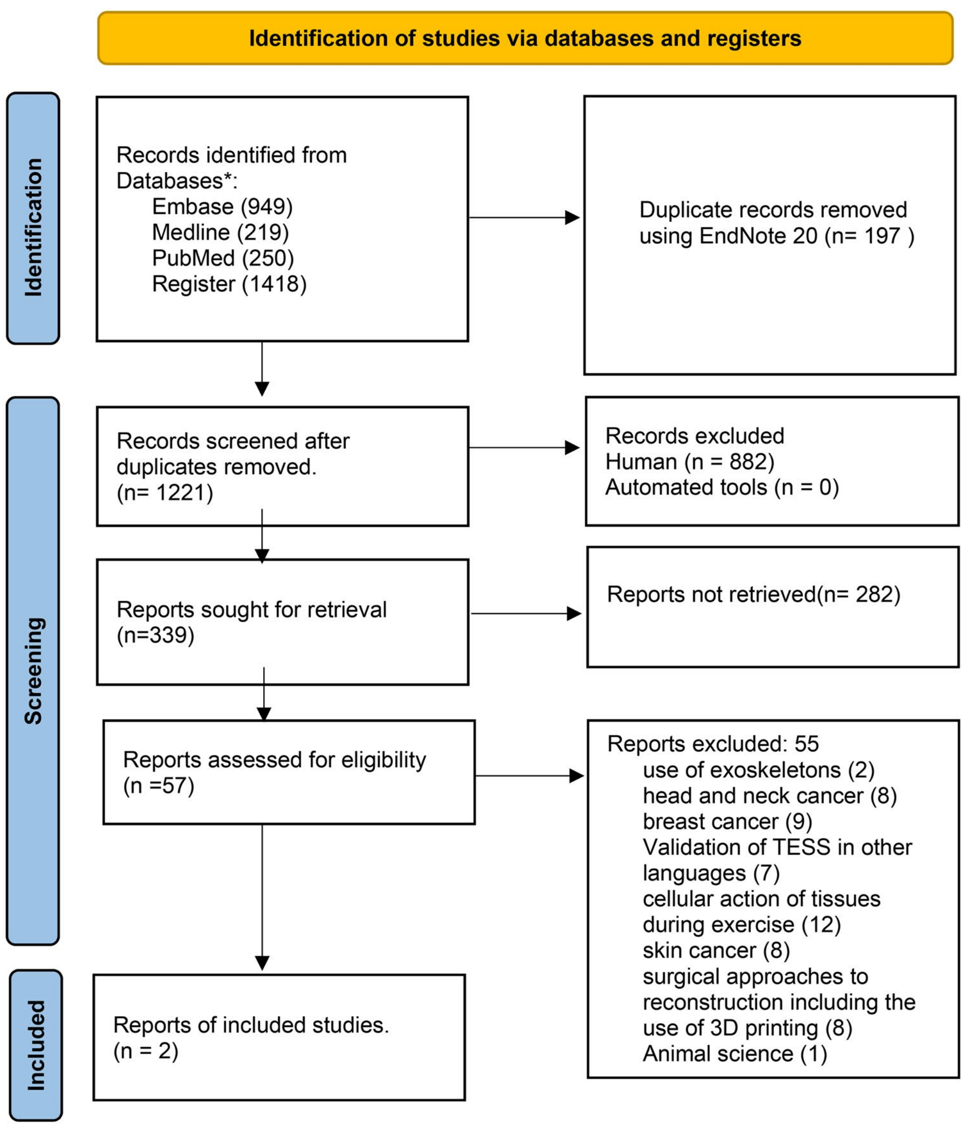

Figure 2. PRISMA flowchart.

6. Effective communication and provision of information.
7. Patient support through access to other services.

The overview of the rationale and primary recommendations about the four phases of the pathway are synthesised in Table 1.

Discussion

Principal finding

This project is the first of its kind focusing on developing national guidance in the field of rehabilitation in musculoskeletal oncology with a view to guide NHS health systems and policy. The evaluation analysed the evidence available in the field and consensus development approaches were utilised. The review identified the wide range of physical limitations people treated for musculoskeletal tumours face and gaps in the literature regarding rehabilitation approaches. The consensus methods used in developing this guidance exercised various rounds of consultations with a mixture of stakeholders to ensure a wide perspective was gained.

Clinical interpretation of key results

The key results derived from this guideline development evaluate and reflect on important elements of rehabilitation practice in the field of musculoskeletal oncology and implementing these could significantly improve the standardisation of care delivery nationally.

Access to specialist AHPs for treatment

As musculoskeletal tumours are rare and complex, AHPs with sarcoma-specific knowledge and experience are key in the pathway. To provide relevant, accurate and timely information, specialist AHPs, with sarcoma-specific knowledge and experience should work closely with the wider sarcoma multidisciplinary team (MDT) [37,38] and general therapists at a national level [39].

AHPs can support many of the strategic aims of the NHS to improve QoL and transform care by improving the health and wellbeing of a person across the whole pathway; from diagnosis, through to palliative and end of life care [40].

Documented referral pathway for specialist AHP care

Surgery following a diagnosis of bone or soft tissue sarcoma is carried out at specialist centres in the UK and rehabilitation following discharge is usually undertaken by providers closer to home [38]. Access to specialist referral pathways within the specialised centres in the UK assists in streamlining services [2]. As sarcoma management comprises of complex multimodality assessment and therapy including surgery, chemotherapy, or radiotherapy; unique rehabilitation; and palliative care needs may arise due to impact of treatments [38]. Early postoperative gain in activity levels after treatment for lower extremity sarcoma positively affects long-term PA and performance [41]. Delays in starting rehabilitation can increase the risk of poor post-surgical outcomes. The first two years following surgery are important, when the patient will optimise their outcome [42].

Assessment of individualised issues, personal and multi-dimensional (holistic) needs

Treatment for sarcoma depends on various factors including location, size, depth, and subtype of the tumour. Treatment can include surgery, radiotherapy, and chemotherapy, all with their own range of potential side effects.

Surgery may include the removal of muscle, bone, or nerves through limb-sparing surgery, or may necessitate amputation, all of which can impact physical, psychological, and social functioning [9]. Physical impairments can be wide-ranging depending on the structures involved and the type and extent of treatment. Pain, fatigue, and psychological distress [43] are also common in the sarcoma population and should be assessed for and monitored at regular intervals [4,9,44]. Both limb-sparing surgery and amputation can increase the risk of falls, particularly after specific procedure such as femoral nerve resection [36,45].

Lymphoedema can result from disturbance of the lymphatics because of surgery and/or radiotherapy and is associated with poorer outcomes [8]. Radiotherapy can also result in radiation-induced fibrosis, which may manifest clinically months or years after treatment. This can lead to tissue tightness, weakness, and dysfunction [46].

Timely, holistic and personalised care can improve QoL [47,48]. Personalised care also benefits the NHS and wider society by redirecting the use of health and social care resources effectively and helping people return to activities that benefit the economy, such as work, education, or spending money on holidays and socialising [21], emphasising the importance of the timely assessment and management of individualised issues, personal and multi-dimensional (holistic) needs. A structured treatment summary provides valuable information and prompts for further support, so that health and wellbeing can be optimised in the years after treatment.

Patient-centred care

People with sarcoma face many challenges, including the psychological difficulties of diagnosis with a life-threatening disease and the debilitating side effects of treatments [1,2]. A patient centred, integrated approach to providing services is fundamental to delivering high quality care [10]. Patient-centred communication and care is therefore gaining importance [49]. Stakeholders widely advocate the adoption of patient-centred care [50]. A growing body of evidence supports relationships between patient-centred care and improvements in adherence to treatment recommendations, management of health outcomes [51].

As individuals cope with their illness, treatment of their condition, and attempt to create a healthy lifestyle during their recovery from surgery, they may face further challenges with communicating their feelings with health care providers.

A holistic needs assessment (HNA), with appropriately selected outcome measurement to guide evidence-based care, personalised care planning and review should be an ongoing and proactive process that is both planned and responsive to changing needs. HNA should continue at regular intervals and key milestones, during and after treatment. This ensures that individual needs can be identified and addressed in a timely manner [47]. Personalised rehabilitation means people have choice and control over the way their care is planned and delivered, based on what matters to them. Personalised needs-based planning involves shared decision making between the individual and the professionals supporting them, putting the person at the centre of decisions about their rehabilitation. Vindrola-Padros et al. [52] evidenced that people's personal strengths, preferences, aspirations, and needs help inform goal setting.

Evidence-based rehabilitation treatment

A key enabler or barrier to implementation of evidence-based rehabilitation is the experience and knowledge of the healthcare provider and AHPs delivering care. Staying up to date with evidence is challenging for busy health professionals, however some helpful frameworks exist.

Table 1. Rationale and recommendations about the four phases of the pathway.

Phase of journey	Benefits of having a recommendation	Summary of recommendations
Diagnosis and care planning for people with a diagnosis of bone or soft tissue sarcoma		
Individual rehabilitation needs should be assessed at or shortly after the point of diagnosis, to facilitate timely access to rehabilitation services.	People with sarcoma may present with physical, emotional, social, and financial support needs from the point of diagnosis. Treatment can have a significant impact on physical function, which can further affect physical and psychosocial wellbeing in the short and long term. All patients, and where appropriate those close to the person diagnosed with a sarcoma, should be informed of the anticipated impact on physical function such as long-term reduction in strength and limb movement which may impact upon future independence to allow them to prepare for how this may affect them and their families. Supporting the management of expectations through shared decision making is an important part of gaining informed consent for treatment [29]. Early identification of patient needs and the anticipated impact of treatment through a proactive approach may help to improve post treatment outcomes. Early referrals to other rehabilitation services, e.g., orthotics or occupational therapy, as indicated, can help prepare patients for treatment, whilst also avoiding discharge delays during inpatient stays.	Patients should have access to specialist AHPs, who are extended members of the Sarcoma multidisciplinary team (MDT), to ensure timely intervention and information provision, in coordination with the wider MDT. Sarcoma MDTs should have an agreed documented referral pathway to specialist AHPs for all patients with a new diagnosis of sarcoma to ensure all patients have access regardless of age, diagnosis, or geographical area. Patients' holistic needs should be assessed as close to the point of diagnosis as appropriate to support proactive rehabilitation referral. This should include consideration of psychological wellbeing, fatigue, nutritional status, frailty, cognition, and home situation. Specialist AHP's should work closely with the wide sarcoma MDT to ensure all patients are educated regarding the anticipated functional outcome of their treatment and the effect it may have on their activities of daily living, work, and hobbies. The sarcoma specialist AHP should co-ordinate early referral to other services, at a geographically convenient location, such as amputee and prosthetic services, wheelchair services and local community occupational therapy teams. Where appropriate, patients should be signposted to appropriate local support services as appropriate such as Maggies, Macmillan Cancer Support, Sarcoma UK, support groups, or psychological services
Patients should have access to tailored prehabilitation after diagnosis and prior to surgical treatment	Prehabilitation (prehab) empowers patients to prepare for treatment by promoting healthy behaviours and optimise physical fitness, nutrition status, and psychological wellbeing. Benefits include improved cardiovascular fitness and nutritional status, enhancing recovery following treatment and reducing complications, reducing length of stay, and improving QoL [30]. Due to the complexities of sarcoma and its treatment, prehab interventions should be guided by specialist AHPs, taking into account patients' individual needs including weightbearing status where there is a risk of pathological fracture	A clear prehabilitation pathway should be established for all specialist centres. Prehabilitation should be included in all documented rehabilitation plans. This should encompass nutritional, physical and psychological optimisation dependent on the identified needs at assessment. All patients should be educated on the benefits of physical activity and encouraged to be as active as possible. Barriers to physical activity should be explored and a personalised plan developed to help overcome these where possible. Patients should be provided with an individualised intervention prescription targeted to their specific prehabilitation needs with ongoing support and evaluation to aid adherence. The level of intervention should be determined by the individually assessed needs and complexity of the patient, as per the prehabilitation guidelines [30]. Those requiring targeted or specialist intervention should be managed by a sarcoma specialist AHP, working closely with the MDT to provide rehab that considers the needs specific to their sarcoma treatment. Patient preferred access to prehabilitation should be considered to minimise exclusion. Delivery methods may include face-to-face sessions (1:1 or groups), virtual appointments, local services, or technology. Information needs should be assessed and information provided in the most suitable format for the individual. This should include: Advice on possible treatment side effects and complications, including prophylactic/preventative management Symptoms to be aware of and when the patient needs to seek advice Sources of information and support for patient's diagnosed with sarcoma Anticipated rehabilitation pathway Patients assessed as high risk of developing lymphoedema should be provided with prophylactic advice and baseline measurements recorded.

(*Continued*)

Table 1. Continued.

Phase of journey	Benefits of having a recommendation	Summary of recommendations
Rehabilitation during treatment for people with a diagnosis of bone or soft tissue sarcoma		
People should have timely access to the rehabilitation services that they require during and after treatment for sarcoma. Where clinically appropriate, a personalised rehabilitation plan will be devised and modified according to ongoing outcome measure evaluation.	Timely access to rehabilitation services and development of an individualised rehabilitation plan can support the patients' return to function by enabling targeted treatment. Tailored strategies to prevent and manage pain, fatigue, poor function, and falls based on assessment, including patient education and the provision of orthoses, are important to support return to normal living [15,31]	Specialist Allied Health Professionals form part of the sarcoma multi-disciplinary team to support timely referral and to inform decision-making regarding intervention. The rehabilitation needs are considered in all patients undergoing treatment for sarcoma as soon as clinically appropriate, ideally prior to initial treatment. A holistic needs assessment (such as the Macmillan HNA or eHNA) should form the basis of a multi-disciplinary assessment alongside a specific assessment by appropriate identified specialist Allied Health Professionals to address the physical, psychological, and social needs identified. Other carefully selected screening tools, such as the clinical frailty scale, may also help to inform personalised rehab plans. On identification of rehabilitation needs, appropriate referrals are made. Signposting to support services, information, and charities are initiated. Initial assessment by rehabilitation professionals, the development of personalised rehabilitation plans and initiation of interventions takes place as soon as clinically appropriate. This is regularly reviewed and documented. Appropriate outcome measures are selected and used to contribute to assessment and treatment planning and to monitor for change. Specific risk-factors associated with sarcoma and its treatment should be assessed for and considered when planning interventions and providing tailored advice. This includes fatigue, mobility and function, risk of falls, and lymphoedema. Rehabilitation planning, goal setting, and outcome measurement is personalised and where possible, involves shared decision making based on what matters to the individual and their individual strengths, needs and preferences. Information regarding return to physical activity and exercise should be discussed with advice around how to do this safely. Patients should have baseline, post rehabilitation intervention, and post treatment outcome measures recorded.
Rehabilitation should commence on an immediate basis based on personalised needs captured through evidence-based outcome measures and individualised rehabilitation plans should be delivered	Where possible "rehabilitation" should be integrated into daily care and support the principles of pacing and energy management. This in turn will empower people to regain physical functioning and independence as soon as possible. The implementation of early rehabilitation after surgery [32] is associated with improved functional outcome [33].	Decision on when to start rehabilitation is undertaken in discussion with the multidisciplinary team including the patient Rehabilitation is holistic, based on the personalised needs assessment, outcome measurement, and rehabilitation plan During rehabilitation the person's clinical presentation will require monitoring via a combination of professionals. Timing, intensity, and frequency of rehabilitation is personalised and flexible to clinical and individual needs Principles of pacing and energy management, including recognition of signs of cancer related fatigue, are incorporated into rehabilitation interventions, as well as supported self-monitoring and/or management The physiotherapy workforce share knowledge, skills, and expertise with the multidisciplinary team so that rehabilitation and symptom management is integrated within daily care and activity. Behaviour change interventions are embedded to best facilitate patient adherence, symptom management, and sustainable lifestyle changes. There is no work specific to behaviour change and modification for sarcoma patients however it seems logical to include interventions within their treatment and support.

(*Continued*)

Table 1. Continued.

Phase of journey	Benefits of having a recommendation	Summary of recommendations
Ensure communication with people diagnosed with sarcoma and their families and carers is effective, with empathetic listening and information shared in an accessible way, personalised to their needs and preferences.	The clear and timely communication of assessment findings, interventions, and discharge plans ensures that rehabilitation is coordinated and personalised to the individual's needs and goals [34]. Honest conversations, for example, regarding realistic expectations for recovery of function, need to be held with sensitivity and it is important that there is a coordinated multidisciplinary approach to communication. Provision of information tailored to a person's preferences, which they can understand and act on, ensures that they are actively involved in shared decision making. Timely and responsive communication and information provision recognises that communication is a two-way process and that the person's circumstances and needs are likely to change over time.	*Recommendations* Additional support such as an interpreter, translator, or advocate may be required. Extra time may be necessary for effective communication. It is important to use language and terminology that is understood and acceptable to people. Communicate information in a personalised, accessible and timely way in order to manage expectations, facilitate decision making and support rehabilitation, discharge and self-management People have their preferences for sharing information with their family members and carers established, respected, and reviewed throughout their care The rehabilitation workforce communicate personalised information using consistent and appropriate language/ terminology and ensure that the person can demonstrate understanding of all information – whilst considering that treatment such as chemotherapy can affect cognitive function. Utilise technology where appropriate taking into account access, digital literacy, needs, and preferences. Appropriate information regarding current and future rehabilitation programmes should be given in the patients preferred format. This includes contact details of any services people are referred on to.
People who undergo treatment following a diagnosis of sarcoma should receive equitable, individualised rehabilitation that is coordinated within multidisciplinary teams and across all relevant settings and services.	Coordinated transfer of care between services is required to enable continuity of care and improves the person's experience of transfer along their care pathway. Effective sharing of information and the rehabilitation plan is essential to delivering integrated rehabilitation and involves close collaboration between health and social care practitioners within and across different services and organisations. Ongoing rehabilitation supports physical, psychological, and emotional recovery. Regular assessment ensures that progress towards personalised goals and outcomes are evaluated. Clearly defined, equitable community rehabilitation pathways and referral processes are required to ensure people in community settings, including care homes, can access the right support from the right service and/or member of the MDT at the right time.	*Recommendations* Rehabilitation is delivered by multidisciplinary teams and is based on regular personalised needs assessment The individualised rehabilitation plan is communicated effectively with the person and with teams responsible for their ongoing care at every transfer point along their care pathway Liaise with discharge coordination teams and hospital discharge teams to ensure that discharge arrangements, including equipment provision and appropriate referrals for the necessary ongoing care, are in place before completing the discharge Consider the psychological wellbeing of the patient The physiotherapy workforce is aware of, and contributes to the development of, equitable rehabilitation models, referral criteria and follow-up arrangements.
Rehabilitation services undertake evaluation, audit, research and share good practice to understand the needs of people diagnosed with sarcoma, improve the quality of services, optimise outcome and experience, and address inequalities.	Co-production is vital in developing and improving outcomes working in equal partnership with people who use rehabilitation services, carers and communities to develop, deliver, monitor, and evaluate rehabilitation services and models. Co-production with seldom heard groups gives voice to people who may have previously been considered hard to reach. This inclusive participation is an important factor in enabling people to feel more involved with the services they use [35]. The rehabilitation workforce should actively participate in the collection of specific, agreed data at a local, regional and national level. Data collected along with emerging research findings can be used to facilitate service evaluation and quality improvement initiatives. National and regional comparison supports the reduction in variation in access to, and quality of, rehabilitation services for those with a diagnosis of sarcoma. That said this is challenging there is a deficit of studies quantifying balance, gait and PA in patients with lower extremity sarcoma [36]. There is an urgent need to develop novel objective measures of physical functioning in this patient group to encourage evidence-based clinical care. Developing the evidence base, standards and guidance and their subsequent impact on rehabilitation is essential to provide a foundation for future improvements.	*Recommendations* Consider the evidence and data requirements for understanding the needs of people with sarcoma, assessing quality of rehabilitation service delivery, measuring patient and carer outcomes and experience, and monitoring for inequalities throughout rehabilitation models. Rehabilitation services collaborate with people diagnosed with sarcoma, their families and their carers to evaluate, improve and redesign services and rehabilitation models. Rehabilitation services have robust systems of measurement and monitoring that, where available, are standardised to enable local evaluation and regional and national interpretation. Good practice, knowledge, and lessons learnt are shared locally, regionally, nationally, and internationally.

(*Continued*)

Table 1. Continued.

Phase of journey	Benefits of having a recommendation	Summary of recommendations
Rehabilitation following end of treatment for people with bone or soft tissue sarcoma		
People who have completed treatment for a bone or soft tissue sarcoma will have their rehabilitation needs assessed and a holistic rehabilitation plan will be developed if clinically appropriate, signposting to relevant services.	A patient centred, integrated approach to providing services is fundamental to delivering high quality care for people with sarcoma. It is important to sign post to physical activity programmes for support and advice. Better understanding of physical functioning may help target interventions aimed at supporting long-term sarcoma survivors. To signpost to appropriate service providers, e.g., Support groups, orthotics, psychology, lymphoedema services, pain clinic, sexual health, and fertility support	*Recommendations* To identify physical needs/rehabilitation potential post treatment completion, identifying patient centred goals. Assessment is holistic and includes consideration of comorbidities (presence of two or more diseases) and prognosis, considering multidisciplinary team discussions. Identify psychological needs post treatment completion. To signpost as required to appropriate service providers when needs are identified. To use patient reported outcome measures to address a change in need and/or outcome. To consider screening tools to inform personalised rehab planning. To recognise signs of a deteriorating patient. To ensure patients are aware of the long-term consequences of treatment they may experience and how these may manifest. Information given on how to access support in the future should any long-term consequences present, e.g., lymphoedema or neuropathy.
Patients highlighted as having either physical and/or rehabilitation needs should be seen in a timely manner, which is coordinated within the multi-disciplinary team and across all relevant settings and services.	As sarcoma is such a rare form of cancer, specialist knowledge of the condition and its rehabilitation is not well known. Therefore, specialist physiotherapists leading the rehabilitation and physical assessments are needed to highlight individual needs and provide specialist guidance/support in all stages of their care post treatment. Examples of moving towards a sarcoma-specialised mainstay of treatment include: Move towards nurse and AHP led clinics – holistic assessment, improved patient experience Functional assessment as part of follow up Management of late effects Benign follow up Access to rehabilitation, potentially via patient initiated follow up for life. Review of sarcoma service specification to include AHP's	*Recommendations* A flexible approach is taken within the rehabilitation sessions, considering the intensity, timing, and frequency of these sessions, allowing for adaptations to clinical and patient's individual needs. Rehabilitation and physical needs assessments are integrated within the patient's daily care and activities, with physiotherapists sharing their knowledge, skills, and expertise within the multidisciplinary team. To continuously identify the ever changing clinical and physical presentation of a patient throughout their rehabilitation, giving reassurance, education, and support when clinically identified. To treat patients within a variety of clinical settings, using appropriate functional assessments within a physiotherapist's scope of practice. To continuously communicate information with the multidisciplinary team in a timely, accessible manner to aid decision making and support rehabilitation and a patient's physical needs. Ensure that rehabilitation is embedded as an ongoing process which people might dip in and out of as their lifestyle and function alter across their lifetime.
Rehabilitation in palliative and end of life care for people with bone or soft tissue sarcoma		
All patients diagnosed with an incurable or terminal sarcoma diagnosis should have access to rehabilitation to optimise their QoL by enabling them to remain as active and productive as possible with minimal dependence on others regardless of life expectancy.	Rehabilitative palliative care aims to optimise people's function and wellbeing and to enable them to live as independently and fully as possible, with choice and independence, within the limitations of advancing illness. It is an approach that empowers people to adapt to their new state of being with dignity and provides an active support system to help them anticipate and cope constructively with losses resulting from deteriorating health. Rehabilitative palliative care supports people to live fully until they die.	Assessment is holistic and includes consideration of comorbidities (presence of two or more diseases) and prognosis, considering multidisciplinary team discussions. Identify psychological needs To recognise signs of a deteriorating patient. A flexible approach is taken within the rehabilitation sessions, considering the intensity, timing, and frequency of these sessions, allowing for adaptations to clinical and patient's individual needs. Rehabilitation and physical needs assessments are integrated within the patient's daily care and activities, with physiotherapists sharing their knowledge, skills, and expertise within the multidisciplinary team. To continuously identify the ever changing clinical and physical presentation of the patient, giving reassurance, education, and support when clinically identified. To treat patients within a variety of clinical settings, using appropriate functional assessments within a physiotherapist's scope of practice. To continuously communicate information with the multidisciplinary team in a timely, accessible manner to aid decision making and support rehabilitation and a patient's physical needs.

Novak et al. [53–55] uses a traffic light colour coded evidence system to provide useful and comparative information to inform rapid decision-making using evidence. The traffic light approach provides a platform for approaching, measuring, and choosing information. The colour systems are green = "do it" (proven effective), yellow = "probably do it", and red = "don't do it" (proven ineffective or harmful) [55]. However, as there is limited evidence in the field of musculoskeletal oncology, evidence should be assessed with caution.

The rehabilitation needs of people diagnosed with sarcoma are not fully evidenced. Analysis has shown that female sex, increasing age, body mass index (BMI), smoking and having a bone lesion have an adverse effect on physical function following limb salvage, as indicated by the mean TESS score; however, further detail and specifics are very limited [56].

Patients treated surgically for extremity sarcomas report long-term reduction in functional outcomes. Factors associated with poor outcomes included large resections of muscles and amputation. Better understanding of physical functioning may help target interventions aimed at supporting long-term sarcoma survivors [57,58] and sign-posting to appropriate service providers, e.g., support groups, orthotics, psychology, lymphoedema services, pain clinic, sexual health and fertility support health and wellbeing events can address unmet need.

It is important to sign post to PA programmes for support and advice. Vindrola-Padros et al. [52] noted that despite participants wanting to be physically active they all report they have experienced significant barriers including: "Fear of damaging the prosthesis"; "Pain from the joint"; "Loss of confidence" following surgery; "stigma"; and a lack of understanding in which exercises they are capable to participate in.

Further studies have enrolled participants in a pilot project utilising Musculoskeletal Tumour Society (MSTS) scoring system, Toronto Extremity Salvage Score (TESS) and accelerometers [59,60,61] embedded in a multi-dimensional biopsychosocial model, International Classification of Functioning Disability and Health (ICF) [59]. The ICF was used to identify functional deficits and then develop personalised rehabilitation programmes to create personalised and objective assessments and standardise clinical care [59]. This proved feasible in single centre and has plans to roll out in multiple centres.

Furtado et al. [36] carried out a systematic review as they noted that physical impairments and activity restrictions cause significant morbidity after surgery for sarcoma, there is a deficit of studies and they do not use consistent, valid, and reliable instruments [62]. There is an urgent need to develop novel objective measures of physical functioning to encourage evidence-based clinical care. Gait kinematics, postural control, and deviations captured in laboratory-based systems and using portable systems can give an accurate picture of balance and gait as an important outcome to consider [63,59].

Where there is not access to gait laboratories in hospitals, portable devices such as markerless sensors or accelerometers can be utilised for outcome assessment, as they can be more sensitive to functional change than PROMS such as TESS and MSTS [59,60]. Prospective multi-centred clinical trials assessing quantitative physical function and QoL are required to identify ideal approaches for individuals [35].

The postoperative functional status of people with primary malignant bone tumours of the lower extremities greatly influences QoL with lower levels of physical function, activities of daily living (ADL), and social participation indicative of poorer QoL [64].

Services for survivors of extremity sarcoma should include rehabilitation and psychological support, sexual health services, expert pain management, and support to return to work [11,65] and sport [66].

Romantic relationships are a key part of a person's life and can have a major impact on their QoL [67]. People diagnosed with sarcoma felt that their condition had negatively affected their relationships as well as making it more difficult to start relationships as it hurt their confidence. This is a common problem in survivors of childhood cancer; however, this has not been well researched in lower limb sarcoma survivors.

Effective communication and provision of information

Emerging evidence supports positive associations between communication behaviours of healthcare professionals and positive health outcomes, including individuals understanding, recall, and adherence to therapy [68]. A consensus about what constitutes "best practice" for effective communication included: (1) fostering the relationship, (2) gathering information, (3) providing information (NHS England and C4PC formerly Coalition for Collaborative Care) making decisions, (5) responding to emotions, and (6) enabling disease- and treatment-related behaviour [50,69–76]. Although this study was not specifically conducted in AHPs, it provides a guide to effective communication and information provision in clinical practice.

Support through access to other services

As the number of people living with and beyond cancer is increasing, so is the importance of gaining access to support services [77]. Care is best delivered by multi-modal therapies from rehabilitation services, MDTs, and support services, which includes access to local professionals, charities, community services, and more to meet the individualised needs of people undergoing treatment for sarcoma [77].

Any new physical, psychological, or emotional problems identified require referral to appropriate services. On discharge from rehabilitation, people need to be informed of contact points available if further rehabilitation becomes necessary [78] demonstrated rehabilitation needs are still apparent 10 years post-surgery and beyond as the long-term consequences combine with the effects of ageing to demonstrate functional decline.

Input from community rehabilitation services needs to be flexible and responsive to changing demands, for example, neutropenia during chemotherapy treatment. Some people may present with long-term conditions at a later stage and need to be able to re-access rehabilitation services as those demands arise for example following recurrence, late effects of treatments, revision surgery, or deterioration of function secondary to other illness.

Implications and interpretation of process of guideline development

Guidelines are developed based on the best available evidence, and across health agencies such as the National Institute of Clinical Excellence [28,79] and the World Health Organization (WHO) [80], these clinical guidelines are developed by conducting systematic reviews, considering that sufficient evidence is present to collate data. These methodologies are considered appropriate as health care services acknowledge the importance and relevance of evidence-based care. The problem arises when limited evidence is available during guideline development, as recommendations still need to be derived and inform clinical care to ascertain optimal use of the available evidence [81].

When evidence is lacking, clinical care recommendations are collated through informal consensus. Informal consensus methods use a deliberative process to describe the efforts amongst a body of experts to discuss the interpretation and use of heterogeneous evidence, with the view to informally reach consensus. Although this deliberative process allows an informal approach to consensus, it is an active participatory platform for experts and stakeholders, to engage face-to-face to assess all available evidence, so it possesses strengths.

Strengths, limitations, and future work

A previous literature review about consensus methodology stated that there is a "danger with informal group consensus when there is no rigour in the methods used" [82]. Despite this being an informal consensus methodology supported by evidence for a rare disease, this methodology has several strengths. The process was conducted rigorously, with systematic, transparent, and reproducible methods throughout. Two reviewers from the authorship conducted the literature searches during the full process and assisted in selection of articles. This can significantly minimise bias. Generated statements were checked by a core group and circulated to relevant stakeholders, including a final approval from the UK's national body of sarcomas, "The BSG" to ensure accurate clinical context. Additionally, although relevant published literature and grey literature were used to support statements, these were assessed prior to use for applicability of context.

Although formal consensus exercises were not used, for example in the form of voting through consensus methods, we recognise this as a limitation and will utilise these formal methods in future updates to these national guidelines.

As part of the consensus development was performed during Covid-19, our team would like to highlight the limitations of using remote consensus and the impact of not receiving responses consistently with remote methods. The bias of an informal group decision-making on the final statement recommendations cannot be excluded entirely. It is also accepted widely that guideline development cannot be based on evidence alone, therefore using a combined approach of consensus development and evidence support, in presence of meagre literature holds merit and facilitates crucial discussions from important stakeholders [83]. Using and documenting these consensus methodologies with a structured literature review enables the process to become more rigorous and effective to generating relevant and clinically useful statements. Additionally, as some of the processes were held remotely, this provides an opportunity for members providing consensus to provide comments anonymously, than those less dominant characters who do not often say their viewpoint face-to-face. This helps to limit bias and ensure all view viewpoints are included.

This project was limited due to it being unfunded and reliant on four clinical staff, it is recognised that more time and resource would be required to involve a methodologist and develop a stronger project. Future iterations of these recommendations should aim to follow a methodology like NICE guidelines through a stakeholder consultation process, which are externally validated. This procedure safeguards against possible bias brought by individual committee members. It is important to acknowledge that in guideline development, using formal consensus with a standard evidence review can strengthen the process further and will be conducted when the next update of these guidelines is taken.

This work was carried out in the UK within the setting of the NHS, much of the literature was however international. The concepts and principles could potentially be utilised internationally; however, the different healthcare settings around the world have implications for the delivery of rehabilitation services and their structure.

Conclusions

The aim of this project was to provide a foundation of best practice based upon expert consensus and best available evidence to develop a framework for the rehabilitation of those with sarcoma from diagnosis through to end-of-life.

Although the current consensus methods and evidence collated created a strong base for this first guideline development; in future, with appropriate planning and selection of topics, a formal consensus approach should be considered for use within future guideline updates when agreed criteria are met. The latter provides transparent methodology, ensuring stakeholders can see how recommendations were developed, despite limited evidence on a topic being available. This work also signified a need to prioritise research into intervention studies relating to sarcoma rehabilitation that could inform practice and guidelines.

Ethical approval

Each author certifies that the British Sarcoma Group endorses and approves the guideline development and that all investigations were conducted in conformity with ethical principles of guideline development.

Disclosure statement

No potential conflict of interest was reported by the author(s).

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ORCID

Craig Gerrand http://orcid.org/0000-0003-3043-4507

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Appendix 1

Database: Ovid MEDLINE(R) ALL <1946 to February 09, 2023>

Search Strategy:

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1. exp Treatment Outcome/ or functional outcome.mp. (1245622)
2. limit 1 to yr="2014 - 2023" (565123)
3. exp "Activities of Daily Living"/ or physical function.mp. (134537)
4. limit 3 to yr="2014 -Current" (57316)
5. activity restrictions.mp. (403)
6. limit 5 to yr="2014 -Current" (229)
7. physical disability.mp. (5350)
8. limit 7 to yr="2014 -Current" (2678)
9. exp Sarcoma/rh, su, th (NHS England and C4PC formerly Coalition for Collaborative Care, 2020) (37981)
10. limit 9 to yr="2014 -Current" (10064)
11. exp Sarcoma/ or exp Bone Neoplasms/ or exp Muscle Neoplasms/ or musculoskeletal tumour.mp. or exp Soft Tissue Neoplasms/ (265756)
12. limit 11 to yr="2014 -Current" (56336)
13. 2 or 4 or 6 or 8 (618112)
14. 10 or 12 (56336)
15. 13 and 14 (9000)
16. extremity.mp. or exp Extremities/ (428104)
17. limit 16 to yr="2014 -Current" (125001)
18. 15 and 17 (850)
19. functional outcome.mp. (30582)
20. limit 19 to yr="2014 -Current" (17278)
21. 4 or 6 or 8 or 20 (76520)
22. 10 or 12 (56336)
23. 21 and 22 (502)
24. 17 and 23 (146)
25. exp "Physical and Rehabilitation Medicine"/ or exp Rehabilitation Research/ or exp Rehabilitation/ or rehabilitation.mp. (597658)
26. 21 or 25 (632617)
27. 24 and 26 (146)
28. 22 and 26 (933)
29. 17 and 28 (189)