Standing frames for children with cerebral palsy: a mixed-methods feasibility study

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Scientific summary

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Background

Cerebral palsy (CP) is the most common motor disorder of childhood, affecting 1 in 400 children. CP is associated with abnormalities of muscle tone and posture with secondary musculoskeletal complications. These have an impact on mobility, participation and function for activities of daily living. Various postural management strategies are recommended to reduce symptoms and maintain body structure, including standing frames. A standing frame has a piece of equipment with a rigid frame and a wide base that supports a person in the standing position. There are a variety of proposed structural and functional benefits for standing frame use, including improved bone mineral density, hip stability and ranges of joint movement at the hip, knee and ankle, and those related to enhancing activity and participation. However, the evidence base for their use is limited. Standing frames may also be disadvantageous. Young people report pain and discomfort; families report increased demands on their time, which reduces family and young person participation. Furthermore, standing frames are expensive, require adaptation with the young person's growth, and use therapists' time to prescribe and monitor their use.

The National Institute for Health and Care Excellence (NICE) guideline for spasticity [NICE. *Spasticity in Under 19s: Management*. NICE Guideline (CG 145). NICE. 2012. URL: www.nice.org.uk/guidance/cg145 (accessed 1 November 2017)] highlighted the limited evidence base for postural management programmes for young people with CP. However, little is known about current UK practice with respect to prescribing or actual use of standing frames. An understanding of this, along with stakeholders' perceptions of the acceptability and feasibility of a standing frames trial, is required.

Aims and objectives

1. Aim 1: to determine current standing frame use in UK practice for the postural management of young people aged 1–18 years with CP and severe movement impairment [Gross Motor Function Classification System (GMFCS) levels IV and V].

    i. Objective 1: conduct a survey (survey 1) of parents, health-care providers and education staff to determine current standing frame use for young people with CP.

2. Aim 2: to assess the willingness of parents to have their child randomised in a potential trial, including the acceptability of different treatment regimens, and to assess the preparedness of health-care providers to recruit to a potential randomised controlled trial.

    ii. Objective 2: undertake qualitative research to explore attitudes to standing frame use and the acceptability of evaluating whether or not there is benefit through a trial or trials. This comprised (1) focus groups with parents, health-care providers and education staff and (2) in-depth interviews with young people.
    iii. Objective 3: propose a small number of potential trial designs, structured around a population, intervention, comparison, outcome, timing, setting (PICOTS) framework and informed by the results of survey 1 and the qualitative research.
    iv. Objective 4: conduct a second survey (survey 2) of parents, health-care providers and education staff regarding the acceptability and feasibility of these potential trial designs.

3. Aim 3: to propose a substantive trial design (or designs) that is informed by, and acceptable to, parents and health-care providers.

Methods

We used a sequential mixed-methods design.

Survey 1

The first stage of the study was a survey of current standing frame use for young people with CP with GMFCS levels IV or V (survey 1). The questions encompassed treatment indications, treatment goals, types of frame, duration of intended and actual use, and perceptions and practicalities of standing frame use. Three populations in the UK were sampled:

- professionals, such as physiotherapists, who prescribe standing frames for young people with CP (prescribing clinicians)
- professionals, such as paediatricians, orthopaedic surgeons, physiotherapists and education staff, who do not prescribe standing frames but work with young people with CP who use them (non-prescribing professionals)
- parents of young people (aged < 18 years) with CP who currently use or have used a standing frame (parents).

Qualitative work

The next stage was qualitative to explore attitudes to standing frame use and acceptability of evaluating their benefit through a trial or trials. Five single stakeholder focus groups were conducted, one each for physiotherapists, medical professionals and education professionals, and two for parents. Young people with CP took part in in-depth interviews about using standing frames. After evaluating the results of these, two multistakeholder focus groups were convened to discuss the findings.

Survey 2

A potential trial design, structured around a PICOTS framework was developed, based on the results of survey 1 and the qualitative research. A second survey (survey 2) regarding acceptability and feasibility of research and the potential trial design was conducted. Three populations in the UK were sampled:

- health professionals, such as physiotherapists and paediatricians, who work or have worked with young people with CP who use standing frames
- education professionals, such as teachers, who work or have worked with young people with CP who use standing frames
- parents of young people with CP who currently use or have used a standing frame.

Study selection (inclusion criteria)

Eligibility of participants:

- Professionals (e.g. of health, education) who work or have worked with young people with CP who use standing frames could take part in the surveys and/or focus groups.
- Parent/carers of young people with CP who currently use or have used a standing frame could take part in the surveys and/or focus groups.
- Young people with CP aged 8–18 years who use or have previously used a standing frame could take part in the interviews.

People could take part in all suitable stages, with the exception of single stakeholder focus group participants, who could not participate in the multistakeholder focus groups.

Data synthesis

The results from survey 1 (current UK standing frame practice), focus groups, interviews and survey 2 (acceptability and feasibility of a trial) were synthesised to provide recommendations for research.

Results

Survey 1

Survey 1 participants included:

- prescribing clinicians ($n = 305$)
- non-prescribing professionals ($n = 155$)
- parents ($n = 91$).

The survey provided insight into current standing frame use. Prescribing practice was consistent across the UK, but achieving the prescribed use was not always possible due to resource, environmental, child and family factors. Professionals and parents of young people with CP were invested in using standing frames. They reported a variety of benefits; however, they also recognised many challenges associated with standing frame use.

Qualitative work

There were three to nine participants in each single stakeholder focus group ($n = 33$ participants). The single stakeholder focus groups added greater understanding to survey 1 results. Orthopaedic surgeons and physiotherapists had a strong belief that without standing frames there may be progressive deterioration in body structure and body function for young people with CP. The views of parents were entrenched in the idea that standing frames are good, and many were surprised to hear about the lack of robust evidence. A number of the proposed outcomes, particularly regarding body structure and body function, would require a longitudinal study in order to answer the question about the impact of standing frames. Other outcomes would be feasible to measure. There was no consensus regarding duration of intervention or comparators.

The interview participants were 12 young people with CP who were currently using or had used standing frames. The young people had clear opinions about standing frame use, but reported that they did not often get the chance to express them. Feelings about standing frames were unique to the individual; however, participation and activity engagement were particularly important to young people.

Two multistakeholder focus groups were convened, both in England: one in the North and one in the South. In the Northern group, participants were two education professionals (mainstream – classroom support assistants), one parent, one orthopaedic surgeon, one neurodisability paediatrician, one paediatric neurologist, one research occupational therapist and one physiotherapist. In the Southern group, participants were five physiotherapists, one community occupational therapist, one paediatrician, and one education professional (early years key worker). The multistakeholder focus groups added more in-depth clinical insight into potential trial designs for the different stakeholder groups. There were education barriers to overcome for all stakeholders as each person brought their unique experiences and biases when sharing their perceptions of the value of standing frames. Professionals, parents and young people were not in emotional equipoise despite understanding the evidential equipoise.

Survey 2

Survey 2 explored the acceptability and feasibility of research trials. Participants included:

- health professionals, $n = 467$
- education professionals, $n = 44$
- parents, $n = 74$.

Most respondents believed that standing frames research is necessary and they were willing to engage in a trial. The maximum amount of time most health professionals and parents would agree to suspend standing frame use was 12 weeks. There were factors that would stop professionals and parents participating in a standing frames study, such as fear that suspending use would cause irreversible damage. Factors such as these are important when considering trial recruitment.

The collated study results (survey 1, single stakeholder focus groups, interviews, multistakeholder focus groups, survey 2) were presented in two multistakeholder design workshops. These design workshops discussed the (1) study's findings, (2) priorities for research studies, (3) potential trial designs and (4) conclusions and recommendations. Attendees at the design workshops included co-applicants, steering group members and various stakeholders such as physiotherapists, orthopaedic surgeons, paediatricians, parents and a young person with CP.

Conclusions

Our findings suggest that a trial could examine standing frame use in young people with CP GMFCS III–V. The primary outcome of the trial should be selected from patient-reported outcomes (e.g. participation), with quality of life, subjective well-being, body function and body structure as secondary outcomes. There would be multiple factors to consider in a trial design including the heterogeneity of the population, significant challenges to recruitment and retention, and adherence to protocol. These challenges could be overcome by clinical understanding of the population and careful trial design, including an internal pilot.

A mixed-methods approach that captures quantitative and qualitative data about users' experience would be necessary. We have shown that it is possible to obtain young people's views, which are highly valuable with respect to their engagement in clinical intervention and research.

Despite the publication of the *International Classification of Functioning, Disability and Health* in 2001 [World Health Organization (WHO). *International Classification of Functioning, Disability, and Health: ICF*. Geneva, Switzerland: WHO; 2001] [and the *Children and Youth Version* in 2007 (WHO. *International Classification of Functioning, Disability, and Health: Children & Youth Version: ICF-CY*. Geneva, Switzerland: WHO; 2007)], research and clinical focus still tends to be directed only at body structure and body function. To engage stakeholders in a trial, there would need to be explanation of why measures of patient-reported outcomes (e.g. quality of life, participation and subjective well-being) were important clinical outcomes.

Recommendations for research

We present here our recommendations for a research trial, using the PICOTS framework.

Population: young people with cerebral palsy (Gross Motor Function Classification System III–V)

A study in those of infant and primary school age (4–11 years) is likely to be the most practical, with respect to ease of facilitation of standing frame use in school, size of standing frames and the typical age range in which most young people with CP start using standing frames.

The commissioned call suggested research into young people with CP GMFCS IV and V; however, our survey showed that standing frame use in younger people with GMFCS III was widespread, in keeping with our clinical experience, and we therefore recommend that inclusion criteria should include young people with CP GMFCS III.

Intervention: standing frame use (3 days per week)

We found that the recommended standing frame use was for 30–60 minutes a day for ≥ 5 days each week but that this was not usually achieved. Pragmatically, standing frame intervention for a duration tolerated by the young person for 3 days per week would be an appropriate dosage based on the results from survey 1 (UK standing frame practice).

Comparator: no standing frame use

Standing frame use versus no use, or versus alternative therapy or equipment (e.g. hydrotherapy or disability exercise bike) was discussed in detail throughout the study. Consensus suggests that standing frame use versus no use would be feasible and most likely to detect change. All young people would be likely to have other therapy, orthotics and activities regardless of whether they were in the intervention or non-intervention group ('treatment as usual') but randomisation should lead to a balance with respect to these factors across trial groups.

Outcomes: selected from patient-reported outcomes (e.g. participation), body function and structure

- Primary outcome:
  - a selected patient-reported outcome (participation).
- Secondary outcomes:
  - patient-reported outcomes not included as a primary outcome (e.g. quality of life, subjective well-being)
  - body function (including bowel function, speech, breath control and feeding)
  - body structure (including loss of range of movement).

Measurement tools should address the primary and secondary outcomes of study. Patient-reported outcome measures could assess quality of life, participation and subjective well-being. All measures should ideally be adaptable to the young person's communication level and cognitive ability. There may be a need for parent- or education staff-proxy reports of the child's patient-reported outcomes (quality of life, participation and subjective well-being), although the ideal would be a young person's self-report. It will also be important to assess impact on parents and family life. Secondary outcome measures of body function may include respiratory function, bowel function and pain; and of body structure may include clinical measures of joint range of movement and growth.

Timing: 6–12 weeks

Through survey 1 and the qualitative work, we found that young people often had a break from using standing frames during school holidays. Survey 2 demonstrated that suspending or delaying standing frame use would be acceptable and ethical for a period of 6–12 weeks. However, qualitative data from parents reflecting on past experiences revealed that delayed use (i.e. a waiting list control design) would not be an acceptable trial design. Therefore, we recommend suspended use for 6–12 weeks.

Setting: specialist school environment

Standing frame use in the specialist school environment is recommended because this is where most young people with CP GMFCS III–V are educated. Specialist schools would be better equipped to support standing frame use for the purposes of a trial as they tend to be used in this environment anyway. However, there may be challenges with education, training and support of education professionals in conducting a trial in that setting. For adequate statistical power, a trial would need multicentre recruitment.

Implications for health care

It is important to note that lack of evidence to support standing frame use in young people with CP does not necessarily imply lack of benefit. Many stakeholders (including young people with CP) perceive positive outcomes associated with standing frame use despite the paucity of evidence. As such, standing frames may continue to be prescribed and used even if a future trial demonstrates that they are not effective. Participants suggested that there would need to be evidence of standing frames causing harm in order for people to stop using them.

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