Orthotic management of instability of the knee related to neuromuscular and central nervous system disorders: systematic review, qualitative study, survey and costing analysis

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Scientific summary

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Background

There are several mechanisms that may lead to knee instability in neuromuscular disease (NMD) and central nervous system (CNS) conditions, which can cause several problems for the individual, including pain, falls and a range of mobility issues. Knee instability may be treated using orthoses. A knee–ankle–foot orthosis (KAFO) is usually prescribed when other forms of bracing, such as an ankle–foot orthosis (AFO) or knee orthosis (KO), are insufficient to adequately control knee instability due to weakness or joint laxity. Modern KAFOs made from thermoplastics or carbon fibre composites are lighter and fit more closely, potentially affording better control of the limb compared with ‘conventional’ KAFOs made of metal and leather. Historically, KAFOs were either entirely locked or entirely unlocked at the knee; the locked-knee type requires the individual to alter his/her gait to allow their foot to clear the ground in the swing phase of walking. Recent years have seen the introduction of stance control KAFOs (SCKAFOs), whereby mechanical or microprocessor-controlled knee joints allow the knee to flex during the swing phase of walking but lock when the knee is extended during the stance phase of walking and when weight is borne through the leg to provide stability to the knee in order to allow a more normal walking pattern. Hip KAFOs (HKAFOs) extend across the hip joint connecting to a pelvic band or lumbar or thoracic spinal support. Hip guidance orthoses (HGOs) and reciprocating gait orthoses (RGOs) are examples of HKAFOs with different locking/unlocking mechanisms. There is uncertainty about the acceptability of these devices to patients, the extent to which prescribed devices are used, and factors that determine their usage.

Objectives

To:

- assess the evidence base for the effectiveness of orthotic devices for management of instability of the knee in adults who have NMD or a CNS disorder
- identify the most important outcomes for patients
- identify the types of orthotic devices currently being provided by the NHS for these conditions, the frequency of their use and their cost
- identify any implications for clinical practice, any gaps in the evidence and future research needs.

Methods

We undertook (1) a systematic review of the effectiveness of orthotic devices for management of instability of the knee in adults with NMD or CNS disorders; (2) a qualitative study of the perspective of users of orthotic devices; and (3) a survey of health-care professionals (HCPs) and a costing analysis of KAFOs.

Systematic review

We searched MEDLINE, MEDLINE In-Process & Other Non-Indexed Citations, Cumulative Index to Nursing and Allied Health, EMBASE, PASCAL, Scopus, Science Citation Index, BIOSIS Previews, Physiotherapy Evidence Database, Recal Legacy, Cochrane Database of Systematic Reviews, Database of Abstracts of Reviews of Effects, Health Technology Assessment database and Cochrane Central Register of Controlled Trials in May 2014, and updated in November 2014. Studies in progress, unpublished research or research reported in the grey literature were identified by searching multiple other electronic databases and websites. There were no language restrictions. Studies of adults using an orthosis to manage impaired walking ability due to instability of the knee related to NMD or a CNS disorder were included, provided...
that the orthosis had been used in a real-life setting. Randomised controlled trials (RCTs) and other study designs, with and without a comparator group, were eligible. Key study characteristics and results were extracted and quality assessed. The findings were discussed in a narrative synthesis, grouped by clinical condition, type of orthosis evaluated and outcome.

**Qualitative study of views of orthoses users**

A qualitative in-depth interview study of people with a NMD or CNS condition who had been offered an orthotic device for knee instability was undertaken. The interviews elicited views and experiences of using an orthosis, their perceptions of the treatment they have received, and views of treatment goals and outcomes. A topic guide was used and interviews were audio-recorded, fully transcribed and a sample checked for accuracy. Data were analysed for thematic content following sequential steps of familiarisation with the data; development of a coding scheme and attribution of data to individual codes; collating codes into themes; and interpretation through seeking meaning, salience and connections. Coding procedures were cross-checked in a sample of early transcripts. Data handling and retrieval was assisted by the use of the computer software package NVivo (version 10, QSR International, Warrington, UK). The analytic approach used was both systematic and iterative. An inductive approach was undertaken in order to identify themes in a ‘bottom-up’ way.

**Survey of health-care professionals**

We undertook a web survey of orthotists, physiotherapists and rehabilitation medicine physicians. The questionnaire was distributed using Qualtric® (Qualtrics LLC, Provo, UT, USA) software as an open survey, via an e-mail from the relevant professional bodies, which included an invitation letter and a link to the questionnaire. Reminder e-mails were sent 2 weeks and 4 weeks later. The results were downloaded into Microsoft Excel® (2010, Microsoft Corporation, Redmond, WA, USA). Responses collected for each question were analysed and the response rate for each question calculated.

The cost of KAFOs was estimated using information collected from four telephone interviews undertaken with orthotists, the overall survey, and unit costs estimates collected from NHS sources and expert opinion.

**Engagement and dissemination**

We planned and implemented a combined engagement and dissemination strategy from the early stages of the project. We used a project blog, Twitter (Twitter, Inc., San Francisco, CA, USA; www.twitter.com) feed and articles in newsletters to raise awareness of the project, explain the research methods being used and promote involvement by HCPs in the survey.

**Results**

**Qualitative study**

We interviewed 19 users of orthotic devices across three geographically dispersed NHS sites and five from outside the NHS across different areas of England (n = 24). Half of the sample had been diagnosed as having poliomyelitis; other participants had multiple sclerosis, Charcot–Marie–Tooth (CMT) disease, spinal injury or spina bifida, or had experienced stroke. The median age was 64.5 years (range 36–80 years). Half were engaged in either full- or part-time paid employment; half were retired.

Participants relied on orthotic devices to enable them to engage, as far as possible, in ‘normal’ daily activities, such as working, driving, using public transport, outdoor activities and taking part in social events and gatherings. They used a range of orthoses (KAFO, AFOs and KOs) and mobility aids (sticks, crutches, wheelchairs, mobility scooters), ‘mixing and matching’ these according to differing circumstances and contexts, in order to achieve maximum comfort and independence. Of major importance was whether or not they had a ‘spare’ device in case the currently used device required adjustment or repair or failed unexpectedly. Participants also spoke at length about the footwear associated with their orthosis, and expressed a range of views relating to desirable and undesirable characteristics.
The prime desired outcomes were a reduction in pain, falls or trips, with improved balance and stability. Effectiveness, reliability, comfort and durability were the most valued features of orthoses, and were related to reported use of orthotic devices. Goals for mobility were defined in terms of what they wished to achieve in their daily lives, according to their individual circumstances. They did not discuss treatment outcomes by how far or how fast they could walk. Rather, they focused on different activities that they wished to pursue and judged the success of treatment in terms of how far it enabled them to participate in these activities. The extent to which their orthosis enabled participants to engage in paid employment, outdoor activities (such as gardening), family visits and social events was the yardstick used to assess the effectiveness of treatment. Being able to take part in these activities was regarded as important by participants for both their physical and mental well-being.

Participants expressed frustration with referral routes into orthotic services, channelled through general practitioners and orthopaedic services, which resulted in delays in obtaining effective treatment. People under the care of a consultant in rehabilitation medicine appreciated the consultant’s role in coordinating their care and monitoring their condition, while making proactive and timely decisions to refer them to orthotics and other specialist services, such as to neurophysiotherapy. Many of those interviewed expressed a degree of frustration with deficiencies in the appointment systems in operation in orthotic services. They reported delays in receiving treatment, as well as inconvenience and, sometimes, financial consequences when they had to take time off from work to attend appointments. A particular aspect of orthotic service provision that generated a great deal of commentary among participants related to provision of ‘in-house’ workshops within orthotics departments for the manufacture, adjustment and repair of orthoses. Availability of this facility was associated with delivery of timely, good-quality orthotic care, particularly for minor or emergency repairs to devices.

**Systematic review**

Twenty-one studies (n = 478 patients) were included: two RCTs (one with a crossover design); two non-randomised studies with a control group (one with a crossover design); one cohort study; and 16 case series. Sample sizes were small (range 5–67 participants). Eight studies reported knee instability as a result of NMD (153 patients), seven patients with post-polio syndrome and one patient with inclusion body myositis; 13 studies reported knee instability resulting from CNS causes (325 patients), either post stroke or spinal cord injury. The devices evaluated were KAFOs (mainly carbon fibre), SCKAFO and HKAFOs (RGO and HGO designs).

There were three key findings from the review. First, all of the studies were at risk of bias, in particular in how participants were selected for inclusion or allocated to treatments (in controlled studies) and in how outcomes were assessed. In general reporting was poor. Second, there was a mismatch between the outcomes that were assessed in the studies and the outcomes that were identified in the qualitative study as important to users. The most systematically assessed outcomes were mechanical outcomes, such as gait analysis and energy consumption. In contrast, participants in the qualitative study wanted their orthosis to reduce pain, falls or trips; improve balance and stability; and allow participation in work and a range of other family and social activities. Third, the focus of the effectiveness studies tended to be on the device in isolation. Few studies reported the orthosis ‘dose’ given to the patient, that is, the amount of time per day/week that they were advised to use their orthosis; reporting of fitting and training in use of the device and ongoing review was limited. A strong theme emerging from the qualitative study was that users did not see the device itself in isolation from how they were assessed for provision of the device, measured and fitted, how it functions with footwear, ongoing adjustment of the device and review. Provision of an orthosis is essentially a complex intervention, and this was generally not reflected in the effectiveness literature.
Health-care professional survey
Two hundred and thirty-eight HCPs responded to the survey. Of the 229 who responded to the question on their occupation, there were 80 orthotists, 94 physiotherapists and 50 rehabilitation medicine physicians (five were from other professions). No single group dominated provision among NMD conditions: between 65% and 76% of respondents who managed patients with NMD reported managing patients with poliomyelitis, post-polio syndrome, muscular dystrophy, CMT disease, motor neurone disease and Guillain–Barré Syndrome. Similarly, no single group dominated provision among CNS conditions, although HCPs most commonly reported managing patients who had experienced a stroke (100%) and multiple sclerosis (83%). A range of orthoses are prescribed for knee instability that is related to NMD or CNS conditions: KAFOs (75%), AFOs (94%) and knee brace (89%). A substantial proportion also prescribed shoe adaptations (66%) and insoles (70%). Approximately half of the devices prescribed or fitted were reported to be custom-made (range 0–100%). At least 50% of HCPs thought that comfort and confidence in mobility were extremely important outcomes from treatment. Just over one-quarter of respondents reported that no formal outcome measure was used to assess the effectiveness of the devices provided. No single outcome measure was used by the remaining respondents: the most commonly used measures were the visual analogue scale, the 10-m Walk Test and the Timed Up and Go Test. One-third formally assessed patient satisfaction.

The cost of individual KAFOs was highly variable, ranging from £73 to £3553.

Conclusions
Several different orthoses, both custom-made and prefabricated, are provided in the NHS to manage knee instability that is related to NMD and CNS disorders. Orthoses can play a crucial role in maintaining, promoting and enhancing physical and psychological health and well-being by enabling patients to pursue paid employment, thereby financially supporting their family and facilitating involvement in social and community activities. There is a large gap in the evidence on the effectiveness of KAFOs, AFOs and other orthotic devices for managing knee instability that is related to NMD and CNS conditions. In particular, the outcomes that are important to orthoses users are not being systematically assessed in studies of effectiveness.

Implications for health care
Given the paucity of evidence, it is not appropriate to make conclusions about the effectiveness of specific orthotic devices for knee instability that is related to NMD or CNS disorders.

Better understanding of models of delivery that ensure maximum benefit for patients and best value for money is required.

Use of a core set of patient-reported outcome measures in the clinical setting would facilitate assessment of the impact of any change in device or management strategy on individual patients and would also facilitate audit.

Recommendations for research
Research is required on the effectiveness of orthoses in managing knee instability that is related to NMD and CNS conditions and using outcome measures relevant to patients’ everyday lives. Owing to challenges identified during this research, it is suggested that any future trial be informed by a feasibility study. Given the relative rarity of some of the populations and the personalised nature of the intervention, particularly for custom-made devices, a national registry may be an appropriate way forward.
Development of a core set of outcome measures would be beneficial. Reduction in pain, falls and trips, improved balance and stability, as well as participation in paid employment, outdoor activities (such as gardening), family visits and social events, were all identified as important to patients.

To date there is scant evidence about the views and experiences of people who are given orthoses for knee instability, and further studies are required to investigate further some of the issues raised in our exploratory study.

It is suggested that future research should explore different models of delivery of orthotic service for people with NMD and CNS conditions to identify best practice in terms of greatest benefit to patients and value for money.

**Study registration**

This study is registered as PROSPERO CRD42014010180. The qualitative study is registered as ISRCTN65240228.

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