What is the clinical and cost effectiveness of dynamic elastomeric fabric orthoses (DEFOs) for cerebral palsy?

What is a scoping report?
Scoping reports ascertain the quantity and quality of the published clinical and cost-effectiveness evidence on health technologies under consideration by decision makers within NHSScotland. They also serve to clarify definitions related to the research question(s) on that topic. They are intended to provide an overview of the evidence base, including gaps and uncertainties, and inform decisions on the feasibility of producing an evidence review product on the topic. Scoping reports are undertaken in an approximately 1-month period. They are based upon a high-level literature search and selection of the best evidence that Healthcare Improvement Scotland could identify within the time available. The reports are subject to peer review. Scoping reports do not make recommendations for NHSScotland. Further information on scoping reports is available at www.healthcareimprovementscotland.org

Background
Cerebral palsy (CP) consists of a heterogeneous group of non-progressive clinical syndromes that are characterised by motor and postural dysfunction. There is no cure and therapies are aimed at increasing function and reducing long-term disability. Globally, CP affects approximately 1.5–2.5 children in every 1,000 live births1. It is estimated that there are around 15,000 people with CP in Scotland although there are no precise figures1.

Dynamic Elastomeric Fabric Orthoses (or dynamic lycra® garments) consist of sections of lycra® of varying thicknesses stitched together using specific tensions and directions of pull. The garments are made-to-measure and are designed to meet the specific needs of the wearer. They can cover the whole body or a particular area, eg hand and wrist. They are flexible, designed to move with the wearer and hence referred to as dynamic. There are differences in the garments provided from different suppliers. Lycra® garments can be used alongside other types of splints or replace them completely and are likely to be used as adjuncts to other therapies such as physiotherapy. It is suggested that dynamic lycra® splinting may benefit some children suffering from CP and neuromuscular disorders such as muscular dystrophy by improving their balance, muscle control, proximal stability and movement2. This type of splinting is used to a lesser extent in the treatment of adult neurological conditions such as stroke and multiple sclerosis (D McLean, Coordinator in Adult Rehabilitation and Orthotic Manager, NHS Forth Valley. Personal Communication, Dec 2012). Manufacturers suggest, to achieve optimal effect, the DEFO garment should be worn between 6–12 hours per day, 5–6 days per week (Second Skin, Edinburgh. Personal Communication, Dec 2012). The garments are not worn while sleeping. Suppliers of DEFO lycra® in the United Kingdom include DM Orthotics Ltd., Tru-Life, Second Skin, Jobskin. The garments last for around 12 months but may need to be replaced more often in children as they grow.

Colleagues in NHS Lothian have asked whether these garments are clinically and cost effective. NHS Lothian currently funds a maximum of 15 suits per year with a total spend per annum of around £20,000 (J Tomlison, Consultant in Public Health Medicine, NHS Lothian. Personal Communication, Dec 2012). There appears to be inconsistency in funding of lycra® garments across NHS boards.

This scoping report summarises the published evidence on lycra® splinting in the treatment of CP. It is an update of evidence note 11 (EN11) although the scope has been changed to include adults but excludes patients with muscular dystrophy.

The following questions were scoped:
1. What is the clinical effectiveness of dynamic lycra® splinting for cerebral palsy?
2. What is the cost effectiveness of dynamic lycra® splinting for cerebral palsy?
Literature search

A systematic search of the literature was carried out between 15–19 October 2012. Key resources were searched for secondary literature, policy documents and economic studies. Terms used for these searches included:

- compression or pressure suit/garment/orthotic
- dynamic/flexible/lycra®/support/compression AND splint/brace/body/skin suit
- cerebral palsy
- muscle spasticity/hypertonia.

As little evidence was identified, the following databases were searched for primary literature on 22 November 2012:

- Medline
- Medline in process
- Embase
- Cinahl.

Results were limited to studies published from 2005–2012 in English. A search of ongoing trials databases was carried out on 20 January, 2013 but no trials were identified. A full list of resources searched and primary literature search strategies are available on request.

Evidence base

Table 1 Included evidence sources

<table>
<thead>
<tr>
<th>Publication type</th>
<th>Number of publications</th>
<th>References</th>
</tr>
</thead>
<tbody>
<tr>
<td>Systematic review</td>
<td>2</td>
<td>4,5</td>
</tr>
<tr>
<td>Randomised controlled trial (RCT)</td>
<td>2</td>
<td>6,7</td>
</tr>
<tr>
<td>Local policy documents</td>
<td>2</td>
<td>10,11</td>
</tr>
</tbody>
</table>

Findings

1. What is the clinical effectiveness of dynamic lycra® splinting for cerebral palsy?

Secondary evidence

An evidence note by NHS Quality Improvement Scotland (QIS) in 2005 (EN11) examined the effectiveness of dynamic splinting for children with CP. It was based on two, non-systematic reviews. One review (Attard and Rithalia, 2004) included nine primary studies. These were mainly observational, small (less than 40 children) and of short duration. The second review (National Horizon Scanning Centre, 2002) included five studies, two of which were not featured in the Attard and Rithalia review. One of these was from the Royal Hospital for Sick Children, Edinburgh (1999), and included 10 children with body suits and 10 with arm splints, all supplied by Second Skin. The children were monitored for 8 months, and the study reported that while not all the children liked the suits or benefited from them, some showed significant functional improvements. However, as with other studies in this area, methodological shortcomings mean that the results need to be viewed with caution.

The main conclusions of EN11 were:

- lycra® garments may improve functional abilities in the short term in some children with CP
- suitability needs to be decided on a case-by-case basis
- further research is required to determine the long-term effects of dynamic splinting and which specific patient groups might benefit.

EN11 highlights that the garments can cause a child discomfort and toileting problems. Issues were also noted with putting on and removing the suit and with temperature (particularly in summer). There was limited evidence (in quality and quantity) on carers’ and users’ views on wearing these garments.

A search to update EN11 identified two systematic reviews, an RCT (in two publications) and two local policy documents. A 2010 rapid review by Coghill and Simkiss examined whether lycra® garments (mixed types of garments and manufacturers) improve function and movement in children (aged 0–18 years) with CP.

The authors’ conclusions concurred with those of EN11 and were based on largely the same evidence base. In their commentary, the authors noted that the studies suggest wearing lycra® helped with proximal stability and function in some children and that the suits can cause discomfort and toileting problems. However, there were several methodological shortcomings including: most of the papers were case series with small patient numbers, many of the studies included children with different types of CP and many of the studies did not use objective outcome measures. They also noted that the
evidence suggests that the children in whom the functional benefits of wearing the suits outweigh the disadvantages include those with athetosis (involuntary muscle contractions), ataxia (lack of voluntary coordination) and poor truncal tone. A second systematic review looked at soft splinting on upper limb function (splints made of soft, pliable material such as neoprene and lycra®) in people with CP. No additional studies to those in EN11 or the Coghill and Simkiss review were included. The authors concluded that there was no evidence available to support the use of upper limb soft splinting for people with CP.

The studies within these reviews investigated whether lycra® garments resulted in movement and functional improvement as well as examining attitudes to splinting. A variety of assessment tools were used including questionnaires, motion analysis, three-dimensional (3D) gait analysis, the Melbourne Assessment of Upper Limb Function (an evaluation tool used to objectively measure unilateral upper limb function in children) and the Paediatric Evaluation of Disability Inventory. A variety of design and type of garments were included in the studies. Further, the studies within the systematic reviews were around 6 weeks in length, with one having a splint-wear period of 12 months. There was no literature available on the effect of length of time wearing splint on functional improvement.

No secondary literature on adults with CP was identified.

**Primary evidence**

An Australian RCT with waiting list control conducted in 2011 included 16 children, in two groups of eight (8–15 years) with CP with hypertonic (increased muscle tension) upper limb involvement. Assessments were made at four points in time: at baseline, initial wear of splint, after 3 months of splinting and at immediate removal of the splint.

The results of the RCT are published in two separate publications. The authors describe the study as a ‘randomised parallel group trial with waiting list control’.

The first publication evaluated fluency of movement in children with CP following lycra® splint wear. All participants completed the fluency section of the Melbourne Assessment of Upper Limb Function at each assessment session. In addition, 3D upper limb movement of participants performing four common upper limb tasks was evaluated by a motion analysis system.

There was no change in movement fluency between baseline and 3 months of splint wear for the entire cohort (p=1.00). There were improvements in 5/6 of the movement measures examined. With the exception of one movement measure, these significant differences disappeared on removal of the splint. Improvement in two of the movement measures was greater in children with dystonic CP (n=5), compared with spastic CP (n=10). Based on these results, the authors indicated that children’s movements were faster, more efficient and required less secondary corrections following splint wear.

The second publication looked at the effects of lycra® arm splints in conjunction with goal-directed training (25 minutes of daily active practice of task-specific activities related to the child’s functional goals) on movement in children with CP compared with goal-directed training alone. The authors assessed joint movement and the range of motion (RoM) in three or four joints during four different tasks as well as the Goal Attainment Scale (a tool to detect changes in movement performance of importance to the child and their family) over the splint conditions. Following 3 months of splint wear and goal-directed training, 7/8 children achieved the expected level (for 3 months of training) of goal attainment, while only 1/8 receiving goal-directed training alone achieved this level. When both groups had received training and splint wear, 15/16 children achieved this level (no statistical analysis was carried out).

On immediate splint application, there were significant improvements in only 4/28 measures assessed (RoM or maximum movement). After 3 months of splint wear, there were significant improvements in 20/28 measures assessed. Four out of the 11 improvements (seen in the three tasks that showed significant improvements after 3 months of splint wear) remained on immediate splint removal. Second Skin donated the lycra® garments for use in this trial but had no further involvement in the conduct, analysis and publication of the studies.

There are some reporting and methodology issues with these two publications so results should be interpreted with caution.
The literature search identified a phase one exploratory study and an audit undertaken by NHS England.

The exploratory study was undertaken to establish proof-of-concept of the effects of lycra® orthoses on the gait of children with spastic CP (n=8). The study indicated that lycra® leggings had a beneficial effect on the gait of some children. Power calculations supported the feasibility of a larger, controlled study.

The English audit explored and compared views of parents, physiotherapists and teachers on lycra® splinting of 54 children. Results showed parents and professionals perceived lycra® splints as a useful component of postural management for children with CP and used them regularly. This audit was only available as a conference abstract and the limited detail available means that these findings should be treated with caution. No subsequent publications were identified.

Local criteria policy documents and tools

A brief additional search to identify examples of policy documents on the use of lycra® garments within the NHS was carried out.

A commissioning policy document from Wakefield District NHS board on lycra® garments for children with CP and movement disorders stated that referrals should be considered on a case-by-case basis and included a protocol for criteria required for a child to be offered a lycra® garment.

A commissioning policy statement from Western Cheshire NHS (from December 2009 and due for update December 2012) designated lycra® suits for CP as a treatment with limited clinical value and advised that lycra® suits should not be normally commissioned for management of CP.

Further information

A lycra® clinic runs on a monthly basis in the Royal Hospital for Sick Children, Glasgow. There are approximately 15–20 new referrals per year, with about 50–60 existing children who visit the clinic each month, with large waiting lists. A senior physiotherapist stated that both the physiotherapy and occupational therapy teams and parents rate the garments highly (G Marshall, Senior Physiotherapist, NHS Greater Glasgow & Clyde. Personal Communication, Jan 2013). As far as she is aware, it is the only clinic of its kind in Scotland. She reports that feelings towards the garments differ with each child and on the whole, the children find no problems with heat and only a few with toileting. The clinic use two companies; DM Orthotics and Jobskin (G Marshall, Senior Physiotherapist, NHS Greater Glasgow & Clyde. Personal Communication, Jan 2013).

Queen Margaret University are establishing a pilot study on lycra® leggings for ambulant children with CP and a group of individuals who have experience in lycra® meet regularly (J Tomlison, Consultant in Public Health Medicine, NHS Lothian. Personal Communication, Dec 2012).

2. What is the cost effectiveness of dynamic lycra® splinting for cerebral palsy?

EN11 identified no published evidence on the cost effectiveness of dynamic lycra® splinting. The updated literature search highlighted no published cost-effectiveness evidence.


Summary

This scoping report identified limited clinical and no cost-effectiveness evidence on this topic. Although two systematic reviews published subsequently to NHS QIS EN11 were identified, neither included additional studies. The reviews found that splinting may improve functional abilities in some children with CP. Results from an RCT (with some shortcomings) on upper limb splints in CP suggested that lycra® is a beneficial intervention which is most effective when worn for a period of time (3 months), with small carry-over effects on removal of the splint. However this was a small study (n=16) and so the results should be treated with caution. Synthesis of the literature is difficult due to the differences in the types of orthoses worn (eg glove/body suit), manufacturers’ designs, types of CP in clinical samples and outcomes measured. Further research, with larger numbers, longer follow ups and homogeneity in terms of type of garment...
and manufacturers’ design, is required to determine the effects of lycra® splinting in CP.

No evidence was identified in relation to adults with CP.

The evidence base is limited in amount and in quality with little development since the publication of EN 11.

**Further work for Healthcare Improvement Scotland**

The evidence base was limited therefore no further work on this topic is anticipated.

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**Equality and diversity**

Healthcare Improvement Scotland is committed to equality and diversity in respect of the nine equality groups defined by age, disability, gender reassignment, marriage and civil partnership, pregnancy and maternity, race, religion, sex, and sexual orientation. As a scoping report summarises information and does not provide recommendations a full EQIA assessment is not deemed necessary.

The scoping report process has been assessed and no adverse impact across any of these groups is expected. The completed equality and diversity checklist is available on www.healthcareimprovementscotland.org

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- Dr Kavi C Jagadamma, Lecturer in Physiotherapy, Queen Margaret University, Edinburgh, independent clinical expert
- Gillian Marshall, Senior physiotherapist, NHS Greater Glasgow & Clyde, independent clinical expert
- Donald McLean, Coordinator in Adult Rehabilitation and Orthotic Manager, NHS Forth Valley, independent clinical expert
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- Second Skin and D.M.Orthotics Ltd, manufacturer of technology

Declarations of interest were sought from the clinical advisor and all peer reviewers. All contributions from peer reviewers were considered by the group. However the peer reviewers had no role in authorship or editorial control and the views expressed are those of Healthcare Improvement Scotland.

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- Members of the SHTG evidence review committee

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NICE has accredited the process used by Healthcare Improvement Scotland to produce its evidence review products. Accreditation is valid for 5 years from January 2013. More information on accreditation can be viewed at www.nice.org.uk/accreditation
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