

RESEARCH ARTICLE

Incidence, Time Course and Predictors of Impairments Relating to Caring for the Profoundly Affected arm After Stroke: A Systematic Review

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Abstract

Background and purpose. A significant number of stroke survivors will not recover the use of their affected arm. A proportion will experience pain, stiffness and difficulty with basic care activities. The purpose of the review was to identify predictors of difficulty caring for the profoundly affected arm and establish the incidence and time-course of the related impairments of pain, spasticity and contracture. **Method.** Data sources: Databases (PubMed, MEDLINE, AMED, EMBASE, CINAHL and the Cochrane Controlled Trials Register) were searched from inception to December 2013. Additional studies were identified from citation tracking. Review methods: Independent reviewers used pre-defined criteria to identify eligible studies. Quality assessment and risk of bias were assessed using the McMasters Assessment Tool. A narrative evidence synthesis was performed. **Results.** Thirty-nine articles reporting 34 studies were included. No studies formally measured difficulty caring for the arm, but related impairments were common. Incidence of spasticity in those with weakness ranged from 33% to 78%, shoulder pain affected 22% to 90% and contracture was present in at least 50%. Spasticity and pain appear within 1 week of stroke, and contracture within two weeks. Impairments continued to develop over at least 3–6 months. The most frequent predictors of spasticity and contracture were weakness and reduced motor control, and the risk of pain is most commonly predicted by reduced sensation, shoulder subluxation, weakness and stroke severity. **Discussion.** There is no published evidence on predicting the likelihood of difficulty caring for the arm following stroke. However, the related impairments of spasticity, pain and contracture are common. Given the time-course of development, clinicians may need not only to intervene early but also be prepared to act over a longer time period. Further research is needed to examine difficulty caring for the arm and the relationship with associated impairments to enable researchers and clinicians to develop targeted interventions. © 2015 The Authors Physiotherapy Research International Published by John Wiley & Sons Ltd.

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Keywords

disability; pain; stroke; spasticity

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Introduction

Stroke is the second largest cause of death in adults and the principal cause of long-term severe adult disability

worldwide (Lopez and Mathers, 2006; American Heart Association, 2009; Department of Health, 2009). Seventy per cent of people with stroke will experience arm

weakness, and 62% of these will not recovery dexterity in the arm at 6 months post-stroke (Kwakkel *et al.*, 2003). For the purposes of this review, the term 'profoundly affected arm' is used to describe the situation where a stroke survivor has no movement in the affected arm or when movement is not functionally useful. This term was developed in consultation with a group of stroke survivors.

Current physical therapies in stroke rehabilitation are based predominantly on exercise and task-specific training (Duncan *et al.*, 2005; Intercollegiate Stroke Working Party, 2012). However, most interventions aimed at improving active function require the presence of some movement within the arm initially, and research has shown that additional physiotherapy and practice of motor tasks do not improve active function in those with most significant arm weakness (Parry *et al.*, 1999). For those unlikely to regain active function, a different approach focused on managing disability and avoiding complications in the arm is required. Managing disability involves assessing and reducing impairments in the arm, which can impact negatively on the ability to care for the arm including tasks such as hand washing, nail cutting and dressing (passive function activities) (Sheean, 2001). Impairments, which are commonly associated with the profoundly affected arm and are often targeted for treatment in order to reduce difficulty caring for the arm, include spasticity (Bhakta *et al.*, 2000), contracture (De Jong *et al.*, 2006, 2006) and pain (Shaw *et al.*, 2010). People with arm spasticity and contracture may develop abnormal limb posturing, which can make washing of the axilla, elbow crease and hand difficult, leading to hygiene problems, and potential skin breakdown (Mayer *et al.*, 1997; Fergusson *et al.*, 2007) and increased carer burden (Katalinic *et al.*, 2010). Equally, pain is also often a focus of treatment in improving care of the arm (Ashford and Turner-Stokes, 2009). It is possible that other impairments may impact on passive function of the arm, but there is currently little evidence to support a positive relationship between complications in the arm and impairments such as joint subluxation (Kumar and Swinkels, 2009), and it is difficult to assess the impact of sensory changes in isolation from motor problems (Intercollegiate Stroke Working Party, 2012).

Management of the profoundly affected arm is a complex intervention, which has traditionally included techniques such as splinting (Lannin *et al.*, 2007),

positioning (De Jong *et al.*, 2006), stretching (Bovend'Eerd *et al.*, 2008) and the use of medications such as botulinum toxin (Bhakta *et al.*, 2000). However, evidence to support these interventions is mixed, and frequently, trials have been designed without considering the natural course of impairments and disability in this condition. For example, in a study of splinting to prevent contracture (Lannin *et al.*, 2007), the intervention was provided within the first 8 weeks of stroke for a period of 4 weeks, but there was no rationale to suggest if these timings reflect the period where risk of contracture is greatest (Manigandan and Charles, 2007). Currently, little is known about which people with a profoundly affected arm are most at risk of developing associated impairments or difficulty with passive function. This systematic review has two aims. Firstly, to identify the incidence and natural course of pain, spasticity, contracture and difficulty with passive care in the profoundly affected arm. Secondly, to identify potential predictors that could be used in routine clinical settings in the early stages of post-stroke care to identify those most at risk of difficulty caring for the arm or these related impairments. This is important, as more knowledge of how the profoundly affected arm changes over time will assist researchers and clinicians in designing and evaluating appropriately timed and targeted interventions to ultimately benefit the stroke survivor and their carers.

Methods

Search strategy

The following databases were systematically searched: PubMed, MEDLINE, EMBASE, CINAHL, AMED and the Cochrane Library, from the inception date of each database up to December 2013. The search terms are given in Appendix 1. In addition, citation tracking of journals was undertaken.

Criteria for inclusion of studies

The review included published research articles that fulfilled the following PICOS (Liberati *et al.*, 2009) criteria:

Participants: adults (over 18 years of age) with arm weakness post-stroke

Interventions: the review was not designed to evaluate a specific intervention but did not exclude reports of data

from intervention studies that provided data to answer the review questions (for example, data from control groups identifying changes over time)

Comparators: not applicable

Outcomes: ease or difficulty of passive function of the arm, pain, spasticity or contracture.

Study design: (1) Observational studies of the natural course of events post-stroke and (2) studies evaluating the ability of identified factors (either demographic factors or impairments related to post-stroke presentation) that were assessed within the first 8 weeks of stroke to predict pain, impairment and capacity to care for the arm after stroke.

Studies were excluded if they were not available in English, targeted children or if purely laboratory-based tests such as medical imaging used as predictors. Case series and case reports were excluded owing to the high potential for bias in these study designs. Studies that considered recovery of active function in the arm only were also omitted.

Study selection

Initially, titles, then abstracts, were screened by two members of the review team, working independently. Full studies that met the inclusion criteria were obtained for more detailed evaluation.

Data extraction, management and assessment of potential risk of bias

Two reviewers, working independently, undertook the data extraction and identification of risk of bias, using structured formats. Key data extraction included the following items: general study information (title, author and country of study); study design and characteristics (participant characteristics, potential predictors and outcomes); and findings including length of follow-up. Agreement between reviewers was calculated using kappa scores, and any differences in data extraction were resolved by mutual agreement, and where necessary, referred to a third person. Quality assessment and risk of bias in the selected studies were appraised using a tool adapted from the Quality Assessment Tool for quantitative studies developed by the Effective Public Health Practice Project at McMaster's University in Canada (Effective Public Health Practice Project, 2008).

Summary measures and synthesis of results

The principle summary measures were incidence of each impairment and risk ratio for predictors of either impairment or difficulty with passive care (when this was reported). Data were narratively synthesized via a series of summary tables and reported incidence, change over time and results of any evaluation of predictors. Meta-analysis was not indicated because of inherent heterogeneity of the studies.

Results

Study selection

A total of 539 references were initially identified. There were 219 duplicate references. Figure 1 summarizes the search and reasons for exclusion. Fifty-eight full articles were retrieved, but a further 19 were excluded because they focused on only active rather than passive function, did not include the arm, evaluated laboratory-based tests or imaging or included people with arm weakness for other reasons than stroke. In total, 39 publications were suitable for quality assessment. Five pairs of articles (Table 2) presented differing data from the same studies, but to prevent double reporting, this review includes 39 publications, describing 34 different studies.

Study characteristics

Participants

The characteristics of study participants are summarized in Table 1. Overall a total of 20,590 patients participated in the studies. None of the studies specifically targeted people with a profoundly affected arm. Broadly, they focused on either general populations of people recovering from stroke (including those with a weak arm) or targeted specific populations including people with stroke and hemiplegia, weakness or those who needed rehabilitation. Five studies limited recruitment to people who had sustained ischaemic stroke only, but the others did not differentiate between people with sub-types of stroke. One study explicitly included people with more severe stroke moving to care homes (Sackley *et al.*, 2008). Six studies were from the UK, 11 from Europe, 3 from North America and 14 from other countries. One study involved participants in 35 different countries (O'Donnell *et al.*,

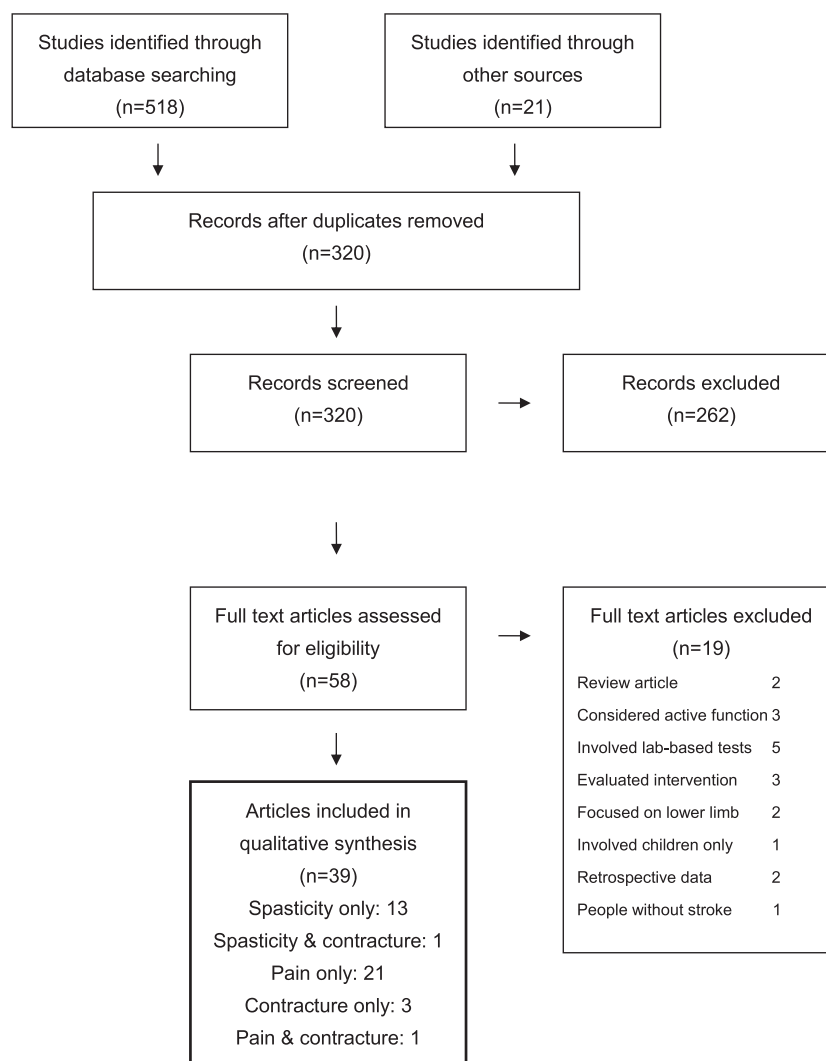


Figure 1. Search results

2013). The average age of study participants was 65.5 years, and they were recruited at any point between the onset of stroke and 1 year after.

Interventions/comparators

The search did not specifically target studies that had evaluated a specific intervention. However, six of the articles included reported data that had been collected as part of larger studies designed to evaluate interventions. Two studies presented data from control groups of intervention trials (Pandyan *et al.*, 2003; Malhotra *et al.*, 2011), one study presented data from both arms of a trial comparing day hospital and community-based therapies (Wanklyn *et al.*, 1996) and one study presented data from all cohorts in a study of antiplatelet

therapy (O'Donnell *et al.*, 2013). Two publications presented data from studies of the predictive value of motor evoked potentials (van Kuijk *et al.*, 2007) and transcranial magnetic stimulation (De Jong *et al.*, 2011).

Outcomes

Table 2 summarizes outcomes measures and predictor variables used in the studies. Three of the studies briefly referred to the ease or difficulty with passive care of the arm (Lundstrom *et al.*, 2008; Kong *et al.*, 2010; Lundström *et al.*, 2010). However, none of these studies measured this as an outcome in a systematic way, although increasingly, measures of difficulty with passive care of the arm are being developed (Bhakta *et al.*, 2000). Fourteen of the publications examined spasticity

Table 1. Characteristics of participants and studies

	Setting	Sample size	Targeted population	Time since stroke at recruitment (days)	Average age at recruitment (years)	Impairment studied	Design
Appelros, 2006	Sweden	253	People with first-ever stroke	Fixed: onset	74 (33–95)	Pain-general	Longitudinal
Ada et al., 2006	Australia	18	People with stroke, with hemiplegia	Variable: 17 (14–28)	63 (36–82)	Contracture-elbow	Longitudinal
Aras et al., 2004	Turkey	85	People with hemiplegia and receiving rehabilitation	Variable: not stated	59.5 (47–70)	Pain-shoulder	Cross-sectional
Bohannon, 1988	United States	30	People with stroke, with hemiplegia and receiving rehabilitation	Variable: 31 (SD 15)	68 (SD 10.6)	Pain-shoulder	Longitudinal
Cheng et al., 1995	Taiwan	50	People with stroke, receiving inpatient rehabilitation	Variable: 21–180	62 (40–79)	Pain-shoulder	Cross-sectional
De Jong et al., 2011	The Netherlands	50	People with first ischaemic stroke with arm weakness, receiving TMS	Variable: within 48 hours	70.3 (58–82)	Spasticity-elbow	Longitudinal
Gamble et al. 2002	UK	123	People with stroke	Fixed: 14	70.6 (29–93)	Pain-shoulder	Longitudinal
Gamble et al., 2000	Iran	152	People with stroke	Variable: 0–60	61 (40–75)	Pain-shoulder	Longitudinal
Hadianfard and Hadianfard, 2008	Singapore	148	People with stroke, with weakness and receiving rehabilitation	Variable: not reported	63 (53–76)	Spasticity-arm	Longitudinal
Kong et al., 2012	Singapore	140	People with stroke, with weakness and receiving rehabilitation	Variable: 15 (SD 14.6)	61 (SD 13.3)	Spasticity-arm	Longitudinal
Kong et al., 2010	Holland	40	People with ischaemic stroke, with complete arm paralysis	Fixed: onset	68 (59–77)	Spasticity-arm	Longitudinal
Kuptniratsaikul et al., 2013	Thailand	214	People with stroke	Variable: (median 24)	62 (59–75)	Spasticity-general	Longitudinal
Kwah et al., 2012	Australia	165	People with stroke	Variable: up to 28 days	78 (IQR 65–84)	Contracture-general	Longitudinal
Leathley et al., 2004	UK	106	People with stroke	Fixed: onset	70 (SD 11.3)	Spasticity-general	Longitudinal
Watkins et al., 2002	Sweden	327	People with first ever stroke	Fixed: onset	73 (17–102)	Pain-shoulder	Longitudinal
Lindgren et al., 2007		58 (subset)	People with first ever stroke, with motor or sensory deficit and pain		71		
Lindgren et al., 2012	Sweden	47	People with first ever stroke and initial weakness	Variable: 2–10 days	74 (34–84)	Spasticity-arm or leg	Longitudinal
Lundstrom et al., 2010	Sweden	140	People with first stroke	Fixed: 12 months	71 (SD 13)	Pain-general	Cross sectional
Lundstrom et al., 2009	UK	30	People with first stroke and no function of arm	Variable: 21 (range 7–35)	70 (52–90)	Spasticity-arm	Longitudinal
Lundstrom et al., 2008						Contracture-wrist	
Malhotra et al., 2011	Brazil	146	People with ischaemic stroke	Fixed: onset	64 (25–88)	Spasticity-general	Longitudinal
Moura et al. 2009	35 countries	15754		Variable: (median 15)	65 (not reported)	Pain-general	Longitudinal

(Continues)

Table 1. (Continued)

	Setting	Sample size	Targeted population	Time since stroke at recruitment (days)	Average age at recruitment (years)	Impairment studied	Design
Paci et al., 2007	Italy	107	People with non-severe ischaemic stroke, over 50 years	Variable: 7–27 days	72 (62–82)	Pain-shoulder	Longitudinal
Pandyan et al., 2003	UK	22	People with first stroke and hemiplegia receiving rehabilitation	Variable: 14–28	65 (40–93)	Contracture-wrist Spasticity- wrist	Longitudinal
Picelli et al., 2013	Italy	72	People with stroke, with weakness	Variable: within 7 days	71 (SD 10)	Spasticity-arm	Longitudinal
Pong et al., 2012	Taiwan	76	People with first ischaemic stroke, with hemiplegia and receiving rehabilitation but not receiving medications for spasticity	Variable: not reported	59 (SD 13)	Pain-shoulder	Longitudinal
Poulin de Courval et al., 1990	Canada	94	People with first stroke, with hemiplegia and receiving rehabilitation	Variable: 21–35	Not reported	Pain-shoulder	Cross-sectional
Rajaratnam et al., 2007	Singapore	135	People with unilateral stroke	Variable: 2–14	64 (SD 10.8)	Pain-shoulder	Cross sectional
Ratnasabapathy et al., 2003	New Zealand	1201	People with first ever stroke	Variable: 0–14	Not reported	Pain-shoulder	Longitudinal
Roosink et al., 2011	The Netherlands	31	People with first ever stroke, with sensory or motor signs	Fixed: 2 weeks	67 (52–82)	Pain-shoulder	Longitudinal
Sackley et al. 2008	UK	73	People with Barthel score of <10 at 3 months post-stroke	Fixed: 3 months	76 (31–98)	Pain-shoulder	Longitudinal
Sommerfeld et al., 2004	Sweden	95	People with first ever stroke	Fixed: onset	78 (SD 9.5)	Contracture-general Pain-general	Longitudinal
Sommerfeld and Welmer, 2012		66 (subset)				Spasticity-general	
Suehanapornkul and Kuptniratsaikul, 2008	Thailand	327	People with stroke who could sit out of bed for 30 minutes	Variable: 84	62 (50–74)	Pain-shoulder	Longitudinal
Urban et al., 2010	Germany	211	People with first stroke, with weakness	Variable: not reported	68 (59–78)	Spasticity-general Spasticity-arm	Longitudinal
Wanklyn et al., 1996	UK	108	People with stroke, with ongoing disability returning home	Variable: not reported	Not reported	Pain- shoulder	Longitudinal
Zorowitz et al. 1996	United States	20	People with stroke presenting with shoulder subluxation	Variable: 13–40	63 (42–83)	Pain-shoulder	Cross sectional

SD = Standard deviation; TMD = Transcranial magnetic stimulation.

Table 2. Outcomes and predictor measures used in the studies

	Outcome measures	Predictors of impairment which were assessed		
Pain				
De Jong <i>et al.</i> , 2011	MAS (elbow flexors)	Motor control (FMMA)		
Kong <i>et al.</i> , 2012	AS (shoulder, elbow, wrist and fingers)	Stroke severity (NIHSS)	Global function (mod BI)	Weakness (UEMI)
		Sensation (MAND)		
Kong <i>et al.</i> , 2010	AS (shoulder, elbow, wrist and fingers)	NA		
Kuptniratsaikul <i>et al.</i> , 2013	MAS (elbow and knee)	NA		
Leathley <i>et al.</i> , 2004	Tone assessment scale	Higher cortical dysfunction	Global function (BI)	Weakness (3-point scale)
Watkins <i>et al.</i> , 2002	MAS (wrist, elbow)	(aphasia, confusion or inattention)	Side of stroke Premorbid function (mRS)	Gender Diabetes
Lundstrom <i>et al.</i> , 2010	MAS (shoulder, elbow, wrist, fingers, hip, knee and ankle)	Stroke severity (NIHSS)	Weakness (ssNIHSS)	Sensation (ssNIHSS)
Lundstrom <i>et al.</i> , 2008	MAS (all arm and leg joints)	NA		
Moura <i>et al.</i> 2009	MAS (unclear which joint assessed)	Weakness (MST) Pain (any report)	Gender	Age
Pandyan <i>et al.</i> , 2003	MAS (wrist)	Arm function (ARAT)		
Picelli <i>et al.</i> , 2013	MAS (shoulder, elbow, wrist and fingers)	Motor control (items of European Stroke Scale)		
Sommerfeld <i>et al.</i> , 2004	MAS (all arm and leg joints)	NA		
Urban <i>et al.</i> , 2010	MAS (all arm and leg joints)	Sensation (LT-MAND)	Weakness (BMRC)	
Van Kujik <i>et al.</i> , 2007	AS (elbow and wrist)	Arm control (FMMA) Apraxia (clinical observation)	Global function (BI) Inattention (MAND)	Sensation (LT & FTT)
Pain				
Appelros, 2006	Pain-open question at assessment	Stroke severity (NIHSS)	Sensation (ss NIHSS)	Motor function (ss NIHSS)
Lundstrom <i>et al.</i> , 2009	Pain-VAS	None		
Sommerfeld and Welmer, 2012	Pain-interview	Sensation light touch (perceiving touch with cotton wool)	Motor control (BL) Spasticity (MAS)	Global function (BI) Proprioception (FTT)
Aras <i>et al.</i> , 2004	Pain-MAND	NA		
Bohannon 1988	Pain-reported during examination	NA		
Cheng <i>et al.</i> , 1995	Pain-MAND	NA		
Gamble <i>et al.</i> 2002	Pain-VAS	Mood (HADS) Weakness (ssNIHSS)	Sensation (LT)	Global function (BI)
Gamble <i>et al.</i> , 2000				
Hadianfard and Hadianfard, 2008	Pain-VAS	Global function (Kenny)	Aphasia (any problem with speech)	Visual field (MAND)
		Motivation (MAND)		Mood (symptom checklist)
		Sensation (NSAS and LT)		
Kuptniratsaikul <i>et al.</i> , 2013	Pain-MAND	NA		
Lindgren <i>et al.</i> , 2012	Pain-VAS	Side of hemiplegia	Stroke severity (NIHSS)	
Lindgren <i>et al.</i> , 2007	Pain-VAS	Side of hemiplegia	Stroke severity (NIHSS)	
O'Donnell <i>et al.</i> , 2013	Pain-self report;	Stroke severity (NIHSS)	Gender	Depression ('feeling sad')

(Continues)

Table 2. (Continued)

	Outcome measures	Predictors of impairment which were assessed		
Pain				
	Neurologist assessed the cause (MAND)	Alcohol intake (no. of drinks)	Smoker	Previous exercise
Paci <i>et al.</i> , 2007	Pain-dichotomous response to pain at rest/ on mvt	Global function (mRS) Shoulder subluxation (palpation)	Motor control (FMMA)	Pain
Pong <i>et al.</i> , 2012	Pain-VAS	Motor control (BMR) Spasticity (AS)	ROM (goniometer)	Sensation (MAND)
Poulin de Courval <i>et al.</i> , 1990	Pain- reported during physical examination	NA		
Rajaratnam <i>et al.</i> , 2007	Pain- numerical rating scale	NA		
Ratnasabapathy <i>et al.</i> , 2003	Pain- questionnaire designed by study team	NA		
Roosink <i>et al.</i> , 2011	Pain-numerical rating scale at rest & on movement	NA		
Sackley <i>et al.</i> , 2008	Pain- reported during physical examination	NA		
Suethanapornkul and Kuptniratsaikul, 2008	Pain- MAND	Global function (BI) Spasticity (MAS) Proprioception (MAND)	Subluxation (MAND) Motor control (Brunnstrom)	Mood (HADS) Cognition (Thai mental state exam)
Wanklyn <i>et al.</i> , 1996	Pain- questionnaire designed by study team	NA		
Zorowitz <i>et al.</i> , 1996	Pain- VAS	NA		
Contracture				
Sackley <i>et al.</i> 2008	30% reduction in ROM (MAND)	NA		
Ada <i>et al.</i> 2006	ROM at elbow (measured from photograph- MAND)	NA		
Kwah <i>et al.</i> , 2012	Torque-controlled ROM at elbow wrist and ankle All other joints- 4 point scale of movement restriction	Spasticity (Tardieu)	Stroke severity (NIHSS) Pain (NRS)	Motor control (Mot Ass Scale) Strength (Manual muscle test)
Malhotra <i>et al.</i> , 2011	ROM at wrist with standardized force	Arm function (ARAT)		
Pandyan <i>et al.</i> , 2003	ROM wrist (goniometry with standard force)	Weakness (grip dynamometer)		

MAND = method of assessment not described; ARAT = action research arm test; AS = Ashworth scale; BMRC = British medical research council; BI = Barthel Index; BL = Birgitte Lindmark Motor Assessment; BMR = Brunnstrom motor recovery; FMMA = Fugl-Meyer motor assessment; FTT = Find the thumb; HADS = Hospital anxiety and depression scale; LT = light touch; MAS = Modified Ashworth Scale; Mod BI = Modified Barthel Index; MMSE = mini mental state exam; Mot Ass Scale = Motor assessment scale; mRS = Modified Rankin Score; MST = muscle strength test; NSAS = Nottingham Sensory Assessment Scale; NIHSS = National Institutes for Health Stroke Scale; ROM = range of movement; ssNIHSS = sub-scale of NIHSS; UEMI = Upper extremity motor index; VAS = visual analogue scale.

after stroke, five considered contracture and 22 examined pain.

Spasticity was most frequently measured with the Ashworth Scale, the Modified Ashworth Scale or Tone Assessment Scale, all of which grade the resistance to passive movement. Contracture was measured with a variety of methods including goniometry and

photography, but not all studies described the methods used. Of the studies that examined pain, nine used a visual analogue or numerical scales, and one used a dichotomous variable (pain was either present or absent at rest or on movement). The remaining studies of pain either used unvalidated tools or did not stipulate the methods of its measure.

Predictor measures

The studies examined a wide range of predictor variables including motor and sensory impairment, inattention, cognition, mood, global function and stroke severity (Table 2). Some used predictor measures that have well-established validity and reliability such as the Barthel Index, while other studies developed their own means of assessing predictors often without reference to psychometric testing.

Study designs

Characteristics of the study designs are summarized in Table 1. Twenty-eight of the studies were longitudinal and six were cross-sectional. All of the studies, with the exception of Pandyan *et al.* (2003) and Sackley *et al.* (2008), identified a single primary measure of a specific impairment after stroke and reported its incidence. Although a number of studies referred to evaluation of *predictors* of impairment, this term was interpreted in two different ways. Some studies followed a process where clinical tests were conducted at an early time point to then look at impact of these early predictors on disability or impairment in the longer term (for example, whether Barthel score at 7 days post-stroke predicted longer-term degree of spasticity). The remaining studies looked at the correlation between the selected outcome and related impairment at a single time point (for example, whether range of movement at a joint was correlated with pain). For this review, we included results that related only to early predictors and excluded reference to correlated impairments. A range of statistical analysis was used in the studies including univariate analyses, logistic regression and dividing participants into groups with specific impairments for comparison. In the synthesis of results, account was taken only of data related to incidence, change over time and evaluation of *early* predictors as these relate to the original research question.

Quality assessment and risk of bias within studies

Inter-rater agreement across reviewers for judging the quality of the studies was good with a kappa coefficient of 0.65 (Altman 1991). The areas of potential risk of bias identified in each of the studies are presented in Table 3. Methodological details reported in the papers were of

variable quality. Most of the studies described selection criteria, but many restricted recruitment. The most common shortcomings related to inadequate assessor blinding (detection bias) (if comparing outcomes to predictor measures), and the use of unreliable or unvalidated data collection tools (performance bias). For example, three of the studies that considered pain did not state a consistent approach to its measurement (Cheng *et al.*, 1995; Aras *et al.*, 2004; Suethanapornkul and Kuptniratsaikul, 2008). A further nine used either visual analogue scales or numerical rating scales, and although these may be considered the best tools available, Price *et al.* (1999) demonstrated that people with stroke are often unable to accurately complete them. Given this, and the lack of formal protocol for assessing pain in the majority of studies, the measurement of this outcome is a potential area of bias in all of the studies that examined pain. Equally, there is some debate about whether the measures used to record spasticity, such as the Ashworth scale differentiate between the neural and muscular components of resistance, and studies of reliability and validity have shown mixed results (Fleuren *et al.*, 2010). Nonetheless, they are widely used in both clinical practice and research trials.

Results of individual studies

Summary results of individual studies are presented in Tables 4, 5 and 6. For ease of interpretation, results are presented for distinct impairments and have been subgrouped into studies that recruited populations of all people with stroke against those who recruited only people with stroke who also had hemiplegia or weakness.

Synthesis of results

There were no studies that evaluated the natural course of development or potential predictors of difficulty caring for the arm after stroke in a systematic way. Three studies mentioned difficulty with hygiene and dressing (Lundstrom *et al.*, 2008; Kong *et al.*, 2010; Lundström *et al.*, 2010). However, reference to these difficulties was included within qualitative interviews with an overall rating of difficulty with care, active function and mobility, so it was not possible to extract data related to passive care of the arm. Therefore, the synthesis only considered studies that had examined the related impairments of pain, spasticity and contracture.

Table 3. Potential risk of bias in included studies (positive response indicates less risk of bias)

	Is sample representative of target population?	Are assessors blinded?	Are data collection tools reliable and valid?	Are withdrawals reported?	Were participants unlikely to receive an unintended intervention?	Was statistical analysis appropriate?
Appelros, 2006	Yes	No	No	Yes	Yes	Yes
Ada <i>et al.</i> , 2006	Yes	No	No	No	Yes	Yes
Aras <i>et al.</i> , 2004	No	No	No	Yes	No	Yes
Bohannon 1988	Yes	No	No	Yes	Yes	Yes
Cheng <i>et al.</i> , 1995	No	No	No	Yes	Yes	Yes
De Jong <i>et al.</i> , 2011	No	No	Yes	Yes	Yes	Yes
Gamble <i>et al.</i> 2002	Yes	No	No	Yes	Yes	Yes
Gamble <i>et al.</i> , 2000	Yes	No	No	Yes	Yes	Yes
Hadianfard and Hadianfard, 2008	Yes	No	No	Yes	Yes	No
Kong <i>et al.</i> , 2012	No	No	Yes	Yes	No	Yes
Kong <i>et al.</i> , 2010	Yes	No	No	Yes	No	Yes
van Kujik <i>et al.</i> , 2007	No	No	Yes	Yes	No	Yes
Kuptniratsaikul <i>et al.</i> , 2013	No	No	No	No	Yes	Yes
Kwah <i>et al.</i> , 2012	Yes	No	Yes	Yes	Yes	Yes
Leathley <i>et al.</i> , 2004	Yes	No	Yes	Yes	Yes	Yes
Lindgren <i>et al.</i> , 2012	Yes	No	No	Yes	Yes	Yes
Lindgren <i>et al.</i> , 2007	Yes	No	No	Yes	Yes	Yes
Lundstrom <i>et al.</i> , 2010	Yes	No	Yes	Yes	Yes	Yes
Lundstrom <i>et al.</i> , 2009	Yes	No	No	Yes	Yes	Yes
Lundstrom <i>et al.</i> , 2008	Yes	No	Yes	Yes	No	Yes
Malhotra <i>et al.</i> , 2011	Yes	No	Yes	Yes	Yes	Yes
Moura <i>et al.</i> 2009	No	No	No	Yes	No	Yes
O'Donnell <i>et al.</i> , 2013	No	No	No	Yes	Yes	Yes
Paci <i>et al.</i> , 2007	Yes	No	Yes	Yes	No	Yes
Pandyan <i>et al.</i> , 2003	No	No	Yes	Yes	Yes	Yes
Picelli <i>et al.</i> , 2013	No	No	Yes	No	Yes	Yes
Pong <i>et al.</i> , 2012	No	No	No	Yes	Yes	Yes
Poulin de Courval <i>et al.</i> , 1990	Yes	Yes	No	Yes	No	Yes
Rajaratnam <i>et al.</i> , 2007	No	No	No	Yes	Yes	Yes
Ratnasabapathy <i>et al.</i> , 2003	Yes	No	No	Yes	Yes	Yes
Roosink <i>et al.</i> , 2011	Yes	No	No	Yes	Yes	No
Sackley <i>et al.</i> 2008	Yes	No	No	No	Yes	Yes
Sommerfeld and Welmer, 2012	Yes	No	Yes	No	Yes	Yes
Sommerfeld <i>et al.</i> , 2004	Yes	No	Yes	Yes	No	Yes
Suethanapornkul <i>et al.</i> , 2008	Yes	No	No	No	No	Yes
Urban <i>et al.</i> , 2010	Yes	No	Yes	Yes	Yes	Yes
Wanklyn <i>et al.</i> , 1996	Yes	No	No	Yes	Yes	Yes
Watkins <i>et al.</i> , 2002	Yes	No	Yes	Yes	Yes	Yes
Zorowitz <i>et al.</i> 1996	No	No	No	Yes	No	Yes

Because of the variation in reporting of data (most studies reported *p* values for predictors in isolation of other statistics), and heterogeneity of the included studies, a decision was made not to attempt meta-analysis of the data. Therefore, the synthesis is narrative.

a. Difficulty caring for the arm

There were no studies that evaluated the natural course of development or potential predictors of difficulty caring for the arm after stroke in a systematic way, although three studies mentioned

difficulty with hygiene and dressing in a broader context. Kong, Chua *et al.* (2010) included interviews with people with stroke or their carers and identified 'symptomatic spasticity' as occurring when people reported difficulty with passive function, active function, pain or associated reactions. Lundström *et al.* (2010) and Lundstrom *et al.* (2008) defined 'disabling spasticity' as that which affected any movement, function or social experience and identified this from interviews and unstructured examinations. In all of these studies,

Table 4. Studies of spasticity: individual results

Study	Incidence of impairment	Reporting of change over time	Value of predictors
Studies which recruited a general population of people post stroke			
Kuptniratsaikul <i>et al.</i> , 2013	18% at 12 months	Not examined	Not examined
Leathley <i>et al.</i> , 2004	36% at 12 months	Not examined	1. Any degree of spasticity predicted by ↓ global function ($p < 0.001$) weakness ($p < 0.001$) 2. Severe spasticity predicted by: ↓ global function ($p < 0.001$) Right sided stroke ($p < 0.02$) 3. No relationship with higher cortical dysfunction, gender, diabetes and pre-morbid function
Watkins <i>et al.</i> , 2002	Severe spasticity in 20% at 12 months		
Lundstrom <i>et al.</i> , 2010	4% at up to 10 days, 27% at 1 month; 23% at 6 months	Not examined	1. Spasticity predicted by weakness (OR = 10; 95% CI: 2.1–48.4) stroke severity ($p = 0.002$) 2. No relationship with sensation or global disability
Lundstrom <i>et al.</i> , 2008	17% at 1 year 6% had 'disabling' spasticity in the arm	Not examined	Not examined
Moura <i>et al.</i> , 2009	26% at final timepoint	Not examined	1. Spasticity predicted by pain ($p < 0.0001$; OR = 107.0; 95% CI: 13.5–847.3), weakness ($p < 0.0001$; OR = 91.9; 95% CI: 12.0–699.4) 2. No relationship with gender or age
Sommerfeld <i>et al.</i> , 2004	20% at 1 week, 18% at 3 months	Prevalence decreased over time	Not examined
Studies which recruited a population of people post stroke with hemiplegia or weakness			
De Jong <i>et al.</i> , 2011	10% at 48 hours, 20% at 10 days, 42% at 3 months and 42% at 6 months	Some cases resolved at each time point with 1 new case at 6 months	Spasticity predicted by ↓ motor control ($p < 0.001$)
Kong <i>et al.</i> , 2012	33% at 3 months, 43% at 6 months and 47% at 1 year Severe spasticity in 17%	Some cases resolved at 12 months, with some new cases at 6 and 12 months	1. Moderate to severe spasticity predicted by: ↓ global function ($p < 0.001$) ↓ motor control ($p < 0.001$) stroke severity ($p < 0.001$) 2. No relationship with sensation
Kong <i>et al.</i> , 2010	78%, severe in 38%	Not examined	Not examined
van Kujik <i>et al.</i> , 2007	63% at any time point 55% at 26 weeks	Spasticity evident in 1 week, some cases resolved over all timepoints and few new cases at 26 weeks	No relationship between spasticity and arm control, global function, sensation, apraxia or Inattention
Pandyan <i>et al.</i> , 2003	Not reported	Spasticity evident in 1 week, and developed over 32 weeks	Spasticity predicted by ↓ arm function ($p < 0.01$)
Picelli <i>et al.</i> , 2013	44% had severe spasticity at 6 months	Not examined	Spasticity predicted by: ↓ motor control (OR = 0.45 95% CI 0.31–0.65)
Urban <i>et al.</i> , 2010	43% 16% had severe spasticity	Not examined	Spasticity predicted by weakness ($p < 0.001$) ↓sensation ($p < 0.001$)

difficulties with passive care were included within qualitative interviews, which also involved active function and mobility, so it was not possible to extract data related only to passive care of the arm. It

is interesting to note that all of these studies aligned difficulty with passive care within the construct of 'spasticity', although a clear correlation between these constructs has not been established.

Table 5. Studies of pain: individual results

Study	Incidence of impairment	Reporting of change over time	Value of predictors
Studies which recruited a general population of people post stroke			
Appelros, 2006	11% reported any pain at 1 year	Not examined	Pain predicted by: stroke severity (OR = 1.24 95% CI: 1.11–1.39) weakness (OR 1.8 95% CI: 1.3–2.7) ↓sensation (OR 3.2 95% CI: 1.5–6.5)
Gamble <i>et al.</i> 2002 Gamble <i>et al.</i> , 2000	25% developed shoulder pain at 2 weeks; 40% developed shoulder pain within 6 months	80% of cases had resolved at 6 months	Shoulder pain predicted by ↓sensation ($p < 0.001$) weakness ($p < 0.001$) No relationship with depression or global function
Hadianfard <i>et al.</i> , 2008	32% reported shoulder pain within first year	6% reported shoulder pain in first 2 months, 12% within 4 months and 11% within 6 months Occasional case reported after 6 months	Shoulder pain predicted by ↓sensation ($p < 0.0001$) aphasia ($p < 0.0001$) ↓ global function ($p < 0.0001$) depression ($p < 0.001$) ↓motivation ($p < 0.0001$) No relationship with visual field deficit
Kuptniratsaikul <i>et al.</i> , 2013	34% reported shoulder pain at 12 months	Not examined	Not examined
Lindgren <i>et al.</i> , 2012	22% reported shoulder pain within 4 months; 72% of these still had pain at 16 months	Few new cases at 16 months but resolved cases at all timepoints	Shoulder pain predicted by
Lindgren <i>et al.</i> , 2007			stroke severity ($p = 0.008$) left hemiplegia ($p = 0.01$)
Lundstrom <i>et al.</i> , 2009	21% report stroke pain at 1 year	Not examined	Not examined
O'Donnell <i>et al.</i> , 2013	10.6% report chronic pain	Not examined	Chronic pain predicted by: Stroke severity (OR = 1.07 95% CI: 1.05–1.09) Previous depression (OR = 1.67 95% CI: 1.47–1.89) Previous alcohol intake (OR = 1.37 95% CI: 1.11–1.7) Diabetes mellitus (OR = 1.18 95% CI: 1.05–1.33) Peripheral vascular disease (OR = 1.44 95% CI: 1.09–1.91) Female sex Statin use
Rajaratnam <i>et al.</i> , 2007	22% reported shoulder pain within 1 week	Not examined	Not examined
Ratnasabapathy <i>et al.</i> , 2003	17% at 1 week, 20% at 1 month, 23% reported shoulder pain at 6 months	Pain presented within 1 week, 72% of cases had resolved at 6 months	Not examined
Sommerfeld <i>et al.</i> , 2012	17% initially, 21% at 3 months, 17% at 18 months		Pain predicted by ↓sensation ($p < 0.05$) ↓mobility ($p < 0.05$) No relationship with spasticity, motor control or global function
Suethanapornkul and Kuptniratsaikul, 2008	19% developed shoulder pain	Pain resolved in 77% of cases	Pain predicted by: Shoulder subluxation (OR 2.06 95% CI: 1.08–3.95) No relationship with motor control, spasticity, proprioception, cognition, global function or mood

Table 6. Studies of contracture: individual results

Study	Areas of bias quality score (lower score = increased risk of bias)	Incidence of impairment	Reporting change over time	Value of predictors
Studies which recruited a population of people post stroke with hemiplegia or severe stroke				
Ada <i>et al.</i> , 2006	3/6	51% of those with hemiplegia developed contracture	Contracture evident by 2 weeks and plateaued by 9 weeks	Not examined
Kwah <i>et al.</i> , 2012	5/6	52% develop contracture	Not examined	Contracture predicted by stroke severity ($p < 0.01$) weakness ($p < 0.01$) ↓motor function ($p < 0.01$) No relationship with pain or spasticity
Malhotra <i>et al.</i> , 2011	5/6	100% of those without function develop contracture	Contracture evident by 6 weeks and plateaued by 24 weeks	Contracture predicted by: ↓function ($p < 0.01$)
Pandyan <i>et al.</i> , 2003	4/6	Not reported	Contracture evident by 6–8 weeks and developed over 32 weeks	Contracture predicted by Weakness ($p < 0.01$)
Sackley <i>et al.</i> , 2008	3/6	43% had contracture at 3 months, 56% at 6 months and 67% at 12 months	Not examined	Not examined

b. Spasticity

Incidence

In studies that examined general populations of people post-stroke, spasticity in muscles of the arm was present in 18% of participants at 3 months (Sommerfeld *et al.*, 2004) and 17% at 1 year (Lundstrom *et al.*, 2008). Populations of people who originally presented with weakness had a higher incidence of spasticity with rates between 33% at 3 months (Kong *et al.*, 2012) and 78% at 12 months (Kong *et al.*, 2010).

Time course

Spasticity was evident in some participants as early as 48 hours post-stroke (De Jong *et al.*, 2011). Although the course of spasticity was fairly dynamic, for the majority of cases, it was evident in most participants who would experience it by 3 months (van Kuijk *et al.*, 2007) and developed over at least 32 weeks (Pandyan *et al.*, 2003). There were some cases where early spasticity resolved.

Risk factors

The most frequent predictors of risk of spasticity were weakness (Lundström *et al.*, 2010; Moura *et al.*, 2009; Urban, Wolf *et al.* 2010; Leathley *et al.*, 2004) and

reduced motor control (Kong *et al.*, 2012; De Jong *et al.*, 2011; Pandyan *et al.*, 2003). Stroke severity (Kong *et al.*, 2012; Lundström *et al.*, 2010) and reduced global function (Leathley *et al.*, 2004; Kong *et al.*, 2012) were also positive predictors of risk in at least two studies. The impact of sensory loss on spasticity risk is not clear, with one study identifying a positive relationship (Urban, Wolf *et al.*, 2010) and three discounting this (van Kuijk *et al.*, 2007; Lundström *et al.*, 2010; Kong *et al.*, 2012). However, most of these studies did not clearly identify how sensation was quantified, making comparison difficult. Moura *et al.* (2009) identified early reports of pain as a predictor of risk of spasticity, but in this study, a significant number of areas of potential bias were identified on the quality assessment tool. Higher cerebral dysfunction including apraxia and inattention does not appear to increase risk (Leathley *et al.*, 2004; van Kuijk *et al.*, 2007).

c. Pain

Incidence

Pain in any part of the body was reported by 10% (O'Donnell *et al.*, 2013) to 21% of participants

(Sommerfeld and Welmer, 2012) from a general population of people recovering from stroke, and incidence of shoulder pain occurred in 19% (Suethanapornkul and Kuptniratsaikul, 2008) to 40% (Gamble *et al.*, 2002). Higher incidences of shoulder pain were found in studies of people with hemiplegia, or who were receiving rehabilitation. Within this population, incidence varied from 22% (Roosink *et al.*, 2011) to 90% (Bohannon, 1988).

Time course

Pain was reported as early as 1 week post-stroke (Ratnasabapathy *et al.*, 2003), with new cases of pain still being reported at up to 16 months post-stroke (Lindgren *et al.*, 2007). The highest incidence appeared to be within the first 6 months post-stroke (Wanklyn *et al.*, 1996; Hadianfard and Hadianfard, 2008). The course of pain was fairly dynamic, with some participants reporting resolution of pain at all time points over the first year post-stroke (Wanklyn *et al.*, 1996; Lindgren *et al.*, 2007). However, one study found that 72% of people who experience shoulder pain at 4 months still had pain at 16 months (Lindgren *et al.*, 2012).

Risk factors

The most common predictor of increased risk of pain was reduced sensation (Gamble *et al.*, 2000; Appelros, 2006; Hadianfard and Hadianfard 2008; Sommerfeld and Welmer, 2012), with shoulder subluxation (Paci *et al.*, 2007; Suethanapornkul and Kuptniratsaikul, 2008), weakness (Gamble *et al.*, 2000; Appelros, 2006) and stroke severity (Appelros 2006; Lindgren *et al.*, 2007; O'Donnell *et al.*, 2013) also identified as potential risk factors. The significance of depression was not clear, with two studies identifying a positive link with pain (Hadianfard and Hadianfard, 2008; O'Donnell *et al.*, 2013), and two discounting this (Gamble *et al.*, 2002; Kong *et al.*, 2012). Equally, reduced global function was a predictor of pain in one study (Hadianfard and Hadianfard, 2008), but did not predict pain in two others (Gamble *et al.*, 2002; Sommerfeld and Welmer, 2012). Aphasia and reduced motivation (Hadianfard and Hadianfard, 2008), and reduced mobility (Sommerfeld *et al.*, 2004), had some predictive value in one study each. However, reduced motor control (Suethanapornkul and Kuptniratsaikul, 2008; Sommerfeld and Welmer, 2012), spasticity, proprioception and cognition (Suethanapornkul

and Kuptniratsaikul, 2008) and visual field loss (Hadianfard and Hadianfard, 2008) were not associated with increased risk of pain.

d. Contracture

Incidence

In a single study of a general population of stroke survivors, 52% of participants developed at least one contracture, with the most common joint affected being the shoulder (25%) and elbow (22%) (Kwah *et al.*, 2012). In those with hemiplegia or severe stroke, 51% of participants had elbow contracture (Ada *et al.*, 2006).

Time course

Contracture was detected within 2 weeks of stroke (Ada *et al.*, 2006) and continued up to 32 weeks (Pandyan *et al.*, 2003), although only one study examined this time point.

Risk factors

Contracture was most frequently predicted by weakness (Pandyan *et al.*, 2003; Kwah *et al.*, 2012) and reduced motor function (Malhotra *et al.*, 2011; Kwah *et al.*, 2012). It was linked with increased stroke severity but not degree of spasticity or pain (Kwah *et al.*, 2012).

Discussion

The purpose of this review was to identify the incidence and natural course of pain, spasticity, contracture and difficulty with passive care in the profoundly affected arm and to identify potential predictors of difficulty caring for the arm or these related impairments.

To date, there appear to be no studies that specifically examine the construct of difficulty caring for the profoundly affected arm after stroke. Although three of the studies identified in this review referred to difficulty with care of the arm (Lundstrom *et al.*, 2008; Kong *et al.*, 2010; Lundström *et al.*, 2010), this was included within the construct of problematic spasticity and was identified in interviews and examinations along with difficulties with active function, pain and mobility. It was therefore not possible to identify the incidence or time course of difficulties caring for the arm as a discrete construct, or identify potential predictors of this problem. There is increasing recognition

that in clinical practice, goals concerning the delivery of care to the arm are relevant, so future research in rehabilitation will need to examine this concept in detail.

Therefore, all of the studies included in this review focused on the impairments of spasticity, pain and contracture, which have been identified as having an association with difficulty caring for the arm. Risk of bias was fairly significant in most of the studies identified, particularly concerning the assessment tools used for quantifying both predictors and outcomes. Some studies included self-developed tools with no reference to psychometric testing, but even those using recognized tools had some risk of bias, as many accepted tools still have limited validity and reliability (Hobart *et al.*, 2007). Therefore, caution should be applied in drawing conclusions from the analysis.

There were higher incidences of pain, spasticity and contracture in people who originally presented with hemiplegia after stroke when compared with general populations of people recovering from stroke. In those with hemiplegia, the incidence of arm spasticity ranged from 33% to 78%, shoulder pain affected 22% to 90% and arm contracture was present in at least 50%. The incidence of both contracture and pain in the arm after stroke appears to be similar to that experienced by people following brain injury, where incidences of contracture between 44% (Yarkony and Sahgal 1987) and 84% (Moseley *et al.*, 2008) and incidences of pain between 52% and 58% (Lahz and Bryant 1996) have been identified.

Spasticity and pain were detected from as early as 1 week after stroke, with contracture apparent by 2 weeks. Many cases were dynamic in presentation, with spasticity and contracture continuing to develop beyond 3 months post-stroke and pain developing within 6 months of stroke. Therefore, clinicians may need not only to intervene early post-stroke but also to be prepared to act over a longer time period in managing disability. As interventions are developed, they will also need to take account of the longer-term evolution of these impairments.

Evaluation of the potential predictors of increased risk of impairment is limited, as many of the tools for quantifying the predictors were of limited quality. However, the most consistent risk factors for developing spasticity and contracture were weakness and reduced motor control, and the risk of pain is most commonly predicted by reduced sensation, shoulder subluxation, weakness and stroke severity. It is less

clear if there is a relationship with higher cerebral functions and depression.

Limitations of the review

A comprehensive literature search was undertaken as part of this review, but may be subject to retrieval bias. Notable omissions include the grey literature such as reports, conference proceedings and theses outside commercial publications and articles not published in English. In those studies that have been included, there are large variations within the populations of people with stroke studied, making synthesis of the results limited. Many of the studies themselves used data collection tools that may either not have been subjected to psychometric testing or, if they had, may still not be reliable in people with stroke with particular difficulties such as aphasia or inattention, adding further potential bias.

Implications for physiotherapy practice

There is currently no evidence to predict the risk of developing difficulty caring for the profoundly affected arm after stroke. However, related impairments such as spasticity, contracture and pain affect a significant number of survivors and can start developing within 1–2 weeks of stroke and may not stabilize for at least 6 months post-stroke.

There is no sufficient evidence for clinicians to develop targeted interventions at this stage. However, the research available suggests that clinicians may need not only to intervene early post-stroke but also to be prepared to act over a longer time period in managing disability. Further research is required to establish the relationship between impairments and difficulty caring for the arm and to investigate if predictors of impairment can be used to identify those at risk of developing difficulty caring for the arm. This review has informed the design of a longitudinal study Care of the Arm after Stroke to test a range of predictors of difficulty caring for the arm and to develop a profile of impairment in people with profoundly affected arm.

Ethical approval

No ethical approval was required.

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Conflict of interest

The authors report no declarations of interest.

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