Title: Profile of upper limb recovery and development of secondary impairments in patients after stroke with a disabled upper limb: an observational study.

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ABSTRACT

Purpose: To investigate, in patients after stroke with a very weak upper limb, the profile of recovery for upper limb activity over the first 12 weeks, and whether early secondary impairments predict later upper limb activity. **Method:** Forty patients within 2 weeks of stroke with a very weak upper limb (<4/6 Item 6 of Motor Assessment Scale) were observed longitudinally. Upper limb activity (Items 6, 7 and 8 of Motor Assessment Scale), shoulder subluxation (vertical distance), shoulder pain (visual analogue scale) and upper limb range of motion were measured at 2, 6 and 12 weeks post-stroke. **Results**: By Week 12 upper limb activity was 1.0/18 (IQR 3.0). Shoulder subluxation was 42 mm (SD 8), 7 mm > the intact side at 2 weeks. Pain at rest was 0.1/10 (IQR 1.1); and pain during movement was 2.6/10 (IQR 4.5). Passive shoulder external rotation was 23 degrees (SD 34), 30% of the intact side at 2 weeks. Shoulder pain during movement at 2 weeks predicted shoulder pain during movement at 6 and 12 weeks after stroke ($p = 0.05$). **Conclusion**: Most patients with a disabled upper limb after stroke have little recovery of activity in the first 12 weeks. Shoulder pain on movement at 2 weeks should be flagged as a predictor of future pain.

Key Words: glenohumeral subluxation, contracture, shoulder pain, stroke, upper extremity

INTRODUCTION

Stroke is a leading cause of disability worldwide (Feigin et al, 2010), and independence in activities of daily living depends largely on motor recovery, in particular, recovery of upper limb activity (Veerbeek et al, 2011). Prospective cohort studies suggest that 30-66% of people with a very weak and disabled upper limb initially do not recover upper limb activity by 6 months after stroke (Kwah, Harvey, Diong & Herbert, 2013; Nijland et al, 2010; Sunderland et al, 1994; Wade et al, 1983). People without active finger extension (Nijland et al, 2010; Smania et al, 2007) and shoulder abduction (Nijland et al, 2010) within a week after stroke have poor upper limb activity at 6 months after stroke (Smania et al, 2007; Nijland et al, 2010). While recent research has shown that a small subset of these people do make a better recovery if they have an intact cortico-spinal tract (Stinear et al, 2017), a systematic review concluded that overall, more severe impairment and activity limitations in the upper limb early after stroke are significantly associated with poorer upper limb outcomes (Coupar et al, 2012). Even with considerable rehabilitation, most people after stroke who have a very weak and disabled upper limb appear to have poor upper limb recovery. For example, very weak people acutely after stroke who were provided with electric stimulation to 4 upper limb muscle groups for 60 minutes daily for 4 weeks did not demonstrate any improvement in upper limb activity (Dorsch, Ada & Canning, 2014), and people with weak finger extension, and shoulder abduction, and no intact cortico-spinal tract had limited or no benefit from 30 minutes of daily upper limb rehabilitation over 4 weeks in the first 6 weeks after stroke (Stinear et al, 2012).

People who have substantial ongoing weakness of the upper limb are at risk of developing secondary impairments including shoulder subluxation, pain and contracture (Allison et al, 2015). Shoulder subluxation is reported to occur in 56-81% of people after stroke with no upper limb strength, (Miglietta, Lewitan & Rogoff, 1959; Najenson, Yacubovich & Pikieln, 1971), and shoulder pain is reported in 22-90% of people after stroke with a very weak or disabled upper limb (Roosink et al, 2011; Bohannon, 1988). Contracture of the muscles in the upper limb is reported to occur in 25% of people after stroke (Kwah, Harvey, Diong & Herbert, 2013). These impairments appear to occur with greater frequency in people after stroke who have significant weakness and upper limb disability (Allison et al, 2015).

The time course over which shoulder subluxation, pain and contracture develop in people after stroke has been reported, but is highly variable. Subluxation has been reported to develop within the first month of stroke (Fagrhi et al, 1994; Linn, Granat & Lees, 1999; Zorowitz et al, 1996), although Choolun, Kuys, & Bisset (2016) reported no shoulder subluxation in very weak and disabled people in the first 6 weeks after stroke. A systematic review indicated that shoulder pain is commonly reported to develop by 6 months after stroke (Allison et al, 2015), however, one study reported development of pain as early as 1-2 weeks after stroke (Ratnasabapathy et al, 2003), and another reported development of new shoulder pain 16 months after stroke (Lindgren, Jönsson, Norrving & Lindgren, 2007). Upper limb contracture has been reported to develop from as early as 2 weeks after stroke and to plateau at 9 weeks after stroke (Ada, O'Dwyer & O'Neill, 2006), although other studies report contracture does not develop until 6 weeks after stroke and continues to develop up to 24-32 weeks after stroke (Maholtra et al, 2011; Pandyan et al, 2003). However, it is possible that changes in rehabilitation practice over time, in line with best evidence, may have changed the time course over which shoulder subluxation, pain and contracture develop in people after stroke. As such, a current examination of the time course is merited.

Shoulder subluxation, pain and reduced range of motion may also have a negative impact on outcome of upper limb activity, although findings are inconsistent. Zorowitz et al (1995) found no correlation between upper limb activity and shoulder subluxation on x-ray at 6 weeks after stroke, but more recently Paci et al (2007) demonstrated that shoulder subluxation at baseline was highly correlated with upper limb activity at 2 months after stroke (Paci et al, 2007). Correlations between shoulder pain early after stroke and upper limb activity have also been reported, but range from no correlation to a strong correlation (Lindgren & Brogårdh, 2014; Roy et al, 1995; Zorowitz et al, 1996).

This study, therefore, aimed to investigate the development of secondary impairments including shoulder subluxation, pain and contracture in people after stroke with a very weak, disabled upper limb, and their relationship to upper limb activity. The research questions were, in people after stroke with a very weak, disabled upper limb:

1. What is the profile of recovery of upper limb activity, and development of secondary impairments such as shoulder subluxation, shoulder pain and upper limb range of motion over the first 12 weeks?
2. Do secondary impairments (including shoulder subluxation, pain or upper limb range of motion) at 2 weeks predict upper limb activity at 12 weeks?

METHOD

Design

A longitudinal observational study was carried out. People after stroke with a paralysed or very weak upper limb were recruited on admission to inpatient rehabilitation at 3 different centres, two in Australia and one in Norway. Outcomes were measured 2 weeks, 6 weeks and 12 weeks after stroke by a measurer blinded to the objectives of the study. Measurers were two qualified physiotherapists with training and experience in implementing all measures.

This study was undertaken within a broader randomised trial (Ada et al, 2017) that was approved by the relevant institutions' Ethics Committees and informed consent was obtained from all participants.

Participants

People after stroke were included if they were within 3 weeks of their first stroke, aged between 50 and 85 years, had a hemiparesis or hemiplegia, and had significant weakness and disability of the upper limb (defined as < 4 out of 6 on Item 6 of the Motor Assessment Scale for Stroke (Carr, Shepherd, Nordholm & Lynne, 1985). They were excluded if they had severe cognitive and/or language deficits which precluded them from giving informed consent, or if they had previous pathology of the shoulder. Information such as age, sex, side of hemiplegia, time to inclusion in the study, spasticity (Ashworth Scale scored 0 – 4, where 0 is no spasticity), sensory loss (Thumb Localisation Test, scored 0 -2, where 0 is no loss of sensation), and neglect (Line Bisection Test, where 0 is < 5 mm from the centre of the line) was recorded to describe the sample.

Setting

Of the three rehabilitation units in this study, two were referred patients directly from an acute stroke unit, and one was referred patients from an acute neurology ward. All units provided usual care of the upper limb for prevention of subluxation, muscle strength and/or co-ordination training of the upper limb, and practice of upper limb activities for up to 10 min/day, every week day.

Outcome measures

The outcomes measured were upper limb activity, shoulder subluxation, shoulder pain and upper limb contracture. Upper limb activity was measured using Items 6 (upper arm function), 7 (hand movements) and 8 (advanced hand activities) of the Motor Assessment Scale for Stroke (Carr, Shepherd, Nordholm & Lynne, 1985). Each item is scored out of 6. The three items were summed and reported as a score from 0 to 18, where 0 was no upper limb activity and 18 was very good activity. The upper limb items of the Motor Assessment Scale for Stroke have established validity (Carr, Shepherd, Nordholm & Lynne, 1985; Filiatrault, Arsenault, Dutil & Bourbonnais, 1992; Poole & Whitney, 1988) test-retest and inter-rater reliability (Carr, Shepherd, Nordholm & Lynne, 1985, Malouin et al, 1994) and have small to moderate sensitivity to change over time (English, Hillier, Stiller & Warden-Flood, 2006).

Shoulder subluxation was measured on x-ray and reported in mm. Participants were seated in a chair without armrests, with the arm hanging freely by the side and the elbow extended. A steel ball bearing of known size was taped halfway along the clavicle to act as a reference point. AP plain x-rays of both shoulders were taken at a focal field distance of 1 metre. Shoulder subluxation was quantified using the method described by Prévost, Arsenault, Dutil & Drouin (1987) which measures the shortest perpendicular distance in millimetres (i.e. vertical distance) between the most superior part of the head of the humerus and the inferior portion of the glenoid fossa of the affected arm. This method has demonstrated validity and reliability (Prévost, Arsenault, Dutil & Drouin, 1987). The amount of subluxation was determined by a radiographer subtracting the distance in the affected shoulder from that in the unaffected shoulder. Vertical distance in the intact shoulder was measured once at 2 weeks after stroke.

Pain was measured using a 10-cm visual analogue scale when the participant was at rest, as well as during passive external rotation of the shoulder, where 0 was no pain and 10 was extreme pain. The 10-cm visual analogue scale has adequate inter-rater reliability (Pomeroy, Frames & Faragher, 2000) for measuring pain in people after stroke.

Passive range of motion was measured for shoulder external rotation in supine, and forearm supination and wrist extension in sitting with the forearm on a table, because these are common sites of contracture in people after stroke. An inclinometer was used to measure range of motion of shoulder external rotation, forearm supination and wrist extension. Inclinometers have good inter-rater reliability (van de Pol, van Trijffel & Lucas, 2010) and good validity (Kolber & Hanney, 2012) for measuring upper limb range of motion. The inclinometer was set to zero when the shoulder, forearm and wrist were in the anatomical position and then the joints moved to their end of range of motion and measured in degrees. End of range was defined as the point when further movement was limited by pain or resistance. The intact side was measured once at 2 weeks after stroke.

Data analysis

Profile of recovery was examined using descriptive statistics (mean and standard deviation, or median and interquartile range). Correlation between upper limb activity and subluxation, pain and contracture was examined using Spearman’s correlation coefficient.

RESULTS

Flow of participants

Forty-six participants (26 male, 20 female) aged 69 (SD 10) years old were recruited. Outcomes were collected for 40 (87%) participants, except for subluxation where measures were collected for 38 (83%) participants at baseline and Week 6, and 33 (72%) at Week 12. Characteristics of the participants at baseline are presented in Table 1. Participants were recruited 15 (SD 10) days after a stroke and at this time had little spasticity, moderate sensory loss and little neglect.

Profile of recovery

Table 2 presents the outcomes at Week 2, 6 and 12, and Figure 1 presents the data normalised to the intact side or normal reference values. Upper limb activity did not improve much, being 2% of normal at 2 weeks, 8% at 6 weeks and 13% at 12 weeks. Only a small amount of subluxation developed, such that the vertical distance was 4 mm less than the intact side at two weeks, 6 mm at 6 weeks and 7 mm at 12 weeks. Pain at rest remained unchanged over time, at 0/10 (ie, no pain) at 2, 6 and 12 weeks. Pain during movement increased between 2 and 6 weeks, from 0.5/10 to 4.8/10, and decreased back to 2.6/10 by Week 12. Shoulder external rotation got worse over time, having decreased to 79% of the intact side at 2 weeks, 50% at 6 weeks and 30% at 12 weeks. Forearm supination decreased to 96% of the intact side at 2 weeks, 86% by 6 weeks and 78% at 12 weeks. Wrist extension also got worse over time, decreasing to 89% of the intact side at 2 weeks, 75% at 6 weeks and 65% at 12 weeks.

Relationship between shoulder subluxation, pain, contracture and UL activity

On average, upper limb activity only increased a median of 1 point out of 18 over the course of the study. At 6 weeks, 29 (73%) participants scored ≤ 1/18 for upper limb activity. At 12 weeks 27 (68%) were still only scoring ≤ 1/18 for upper limb activity, and only 3 (8%) participants scored > 9/18. Because so few participants recovered, we were unable to carry out a correlation between secondary impairments and upper limb activity. However, given that pain during movement increased substantially during the study, and that pain has a significant ongoing impact on quality of life, a post-hoc analysis was carried out to determine whether any secondary impairments predicted pain after stroke. Pain during movement at 2 weeks was the best predictor of pain during movement later on, with a Spearman's correlation coefficient of $r_s = 0.44$ ($p < 0.01$) at 6 weeks and $r_s = 0.31$ ($p = 0.05$) at 12 weeks.

DISCUSSION

In people after stroke with a very weak, disabled upper limb, there was very little increase in upper limb activity during the first 12 weeks. Only a small degree of shoulder subluxation became evident. Range of motion was already decreased in all three joints at 2 weeks after stroke, particularly in external rotation of the shoulder, and this continued to decrease over the first 12 weeks. Shoulder pain at rest was stable over the 12 weeks, while shoulder pain during movement increased markedly in the first 6 weeks, but not after that time. Pain during movement at 2 weeks predicted pain at 6 and 12 weeks after stroke.

Overall, upper limb activity did not improve significantly during the first 12 weeks after stroke in this group of stroke with a very weak, disabled upper limb, which is consistent with previous prognostic studies (Coupar et al, 2012; Kwah, Harvey, Diong & Herbert, 2013; Nijland, van Wegen, Harmeling-van der Wel & Kwakkel, 2010; Stinear et al, 2012). However, three people did make a good recovery and one of those made an excellent recovery. Recent research suggests that the few people with better outcomes, while not able to be predicted from their clinical presentation, would have been able to be predicted from the presence of an intact cortico-spinal tract (Stinear et al, 2017). Predicting outcome at an early phase is important so that appropriate rehabilitation goals can be set for people who have the potential to make a good recovery.

Despite the poor upper limb activity at 12 weeks, participants in this study did not develop much shoulder subluxation or pain. Linn, Granat & Lees (1999) reported an average of 63 mm of shoulder subluxation in the first 4 weeks after stroke in a similar group of very weak and disabled people after stroke. It is possible that the difference in these results is because all participants in our study were provided with some form of support to the shoulder as part of usual care, which may have minimized the development of subluxation, unlike the participants in Linn's study who were not provided with any support.

Adey-Wakeling et al (2015) reported an average of 4/10 shoulder pain in patients measured at 4 months, unlike our participants who had an average of 0.1/10 pain at rest at 3 months after stroke. It is likely that the higher degree of pain reported by Adey-Wakeling is because people with previous shoulder pathology, and those who had shoulder pain from a previous stroke were included, unlike in our study. Like our participants, however, pain during movement became worse over time (Adey-Wakeling et al, 2015). In our study, pain during movement at 2 weeks significantly predicted pain during movement at 6 and 12 weeks after stroke, while shoulder subluxation did not predict any pain at rest or during movement at 6 and 12 weeks. Shoulder pain has a significant negative effect on patients' activities of daily living, participation and quality of life (Chae, Mascarenhas & Yu, 2007), as such, preventing the development of shoulder pain after stroke should be a priority. An in-depth assessment should be implemented in any people who have early shoulder pain during movement to identify and treat the cause.

Unlike shoulder subluxation and pain, loss of range of motion was large and developed very early after stroke (i.e. within 2 weeks). The loss of range of motion in shoulder external rotation was substantially greater than in any other joint. In people who are paralysed or very weak after stroke, the shoulder spends a lot of time resting in its shortest range (i.e. full internal rotation, with the hand resting on the lap), unlike the elbow and wrist which rest in mid-range (i.e. 90 degrees of elbow flexion, mid-pronation/supination, and a neutral wrist position). This may explain the relatively greater loss of range of motion in shoulder external rotation. It is estimated that passive shoulder external rotation in healthy older adults is 85 degrees (McIntosh, McKenna & Gustafsson, 2003), and that 59 degrees is required for everyday activities (Namdari et al, 2012). Participants in this study were already limited to 60 degrees of external rotation by 2 weeks after stroke, which decreased to 23 degrees by 12 weeks after stroke. This suggests that people after stroke with significant loss of shoulder external rotation will have insufficient range of motion to participate in daily activities, even if they were to develop sufficient strength to do so. Given there are some people with a very weak and disabled upper limb who do regain effective upper limb activity (Stinear et al, 2017; Nijland, van Wegen, Harmeling-van der Wel & Kwakkel, 2010), preventing loss of external rotation of the shoulder is important.

This study has some limitations. The participants in this study were part of a randomized trial, not an inception cohort, the sample size was small, and only 72% of data was available at 12 weeks. Therefore, the sample may not be representative of all patients with a very weak and disabled upper limb after stroke.

In conclusion, most people with a very weak and disabled upper limb after stroke have little recovery of activity in the first 12 weeks. These people develop secondary impairments quickly after stroke. Pain during movement is evident within 2 weeks of stroke, as is a loss of range of motion of the shoulder, forearm and wrist. External rotation of the shoulder decreases substantially over the first 12 weeks and has the potential to impact on activities of daily living, so should be managed early after stroke. Pain at 2 weeks is associated with shoulder pain later, so, if present, should be assessed thoroughly so the cause can be treated.

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DECLARATION OF INTEREST

The authors report no declarations of interest.

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Table 1. Baseline characteristics of participants and centres

Characteristic	Included	Excluded
	(n = 40)	(n=6)
Age *(yr)*, mean (SD, range)	69 (9, 53-87)	71 (16,51-90)
Sex, *n* males (%)	22 (55)	4 (67)
Side of hemiplegia, n left (%)	27 (68)	5 (83)
Time to inclusion in study *(d)*, mean (SD)	15 (10)	19 (10)
Spasticity *(0 to 4)*, med (IQR)	0 (1)	1 (2)
Sensory loss *(0 to 2)*, med (IQR)	1 (0)	1 (2)
Neglect *(0-2)*, med (IQR)	0 (1)	0 (1)

Table 2 Mean/med (SD, IQR) for all outcomes and % of intact side over time.

Outcome	Intact	Affected Week 2 (n=40)	Affected Week 6 (n=40)	Affected Week 12 (n=40)
Subluxation Vertical distance *(mm), mean (SD)*	49 (8)	45 (9) n = 38	43 (7) n = 38	42 (8) n = 33
Pain VAS *(0 to 10 cm), med (IQR)*				
Rest		0.0 (1.5)	0.0 (1.0)	0.1 (1.1)
Movement		0.5 (2.8)	4.8 (7.0)	2.6 (4.5)
Range of motion *(deg), mean (SD)*				
Shoulder external rotation	76 (13)	60 (17) 79%	38 (25) 50%	23 (24) 30%
Forearm supination	139 (36)	134 (38) 96%	120 (36) 86%	109 (46) 78%
Wrist extension	71 (12)	63 (12) 89%	53 (13) 75%	46 (16) 65%
Upper limb activity MAS Item 6+7+8 *(0 to 18), med (IQR)*		0.0 (1.0)	0.5 (2.0)	1.0 (3.0)

VAS = visual analogue scale; MAS = Motor Assessment Scale for Stroke

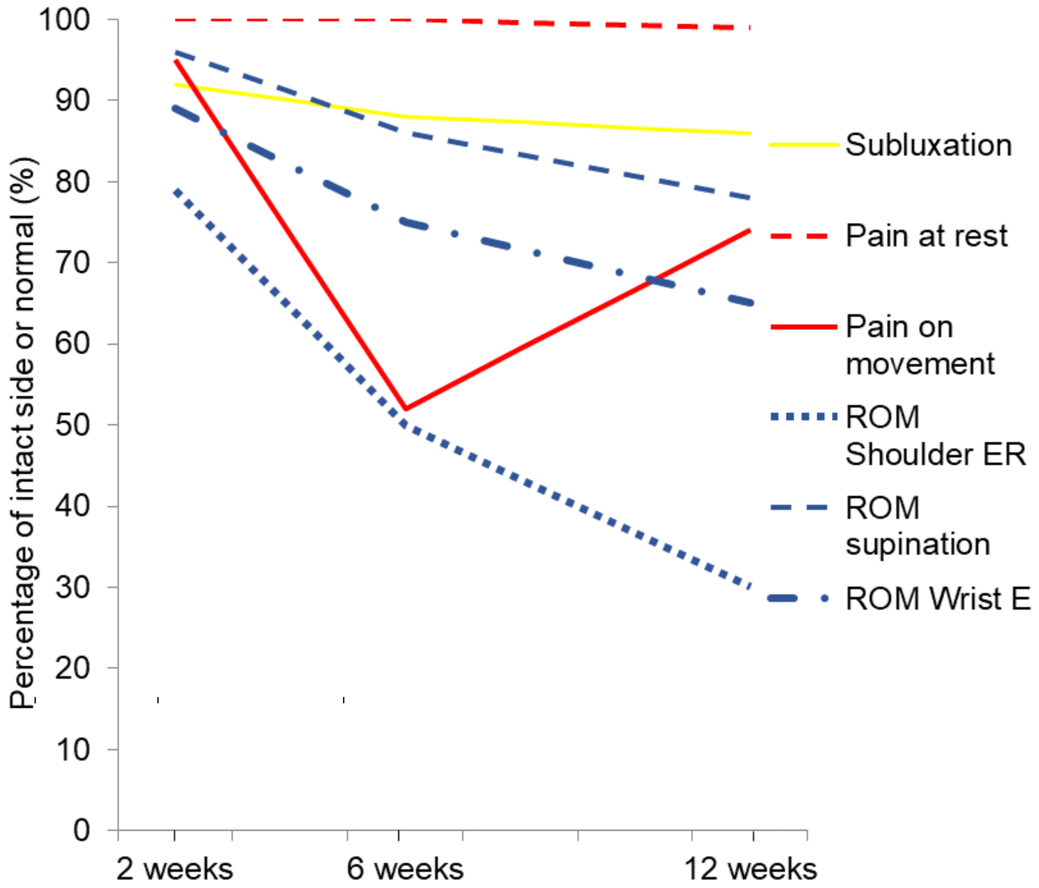

Figure 1. *Percentage of intact side or normal for subluxation, pain and contracture over the first 12 weeks after stroke. ROM = range of motion; ER = external rotation; E = extension.*