# VALIDITY OF THE DISABILITY OF THE ARM, SHOULDER AND HAND PATIENT-REPORTED OUTCOME MEASURE (DASH) AND THE QUICKDASH WHEN USED IN DUPUYTREN'S DISEASE

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**Ethical approval:** This study was a minor element of a larger service evaluation project studying treatment outcome in Dupuytren's disease. In keeping with UK National Research Ethics Service guidance, it is exempt from ethical approval. Approval as service evaluation was prospectively obtained from all centres involved. Local information governance was prospectively obtained from all centres involved, including Caldicott Guardian approval where required.

1	VALIDITY OF THE DISABILITY OF THE ARM, SHOULDER AND HAND PATIENT-
2	REPORTED OUTCOME MEASURE (DASH) AND THE QUICKDASH WHEN USED
3	IN DUPUYTREN'S DISEASE
4	
5	SUMMARY
6	This study investigated aspects of the validity and reliability of the 30-item Disability
7	of the Arm, Shoulder and Hand patient-reported outcome measure (DASH) and its
8	relationship with the shorter 11-item QuickDASH in patients with Dupuytren's
9	disease.
10	Seven hundred and fifty nine DASH questionnaires were studied, covering pre and
11	postoperative patients undergoing different treatments for Dupuytren's disease.
12	Items related to pain rose early after treatment before returning to baseline,
13	suggesting that studying pain is relevant during postoperative recovery. Across all
14	759 sets of responses, the QuickDASH agreed closely with the DASH. In
15	exploratory factor analysis, the DASH was not unidimensional, questioning the
16	validity of the DASH summary score in Dupuytren's disease.
17	Further validation of existing PROMs for use in Dupuytren's disease is needed.
18	These data suggest that pain is a relevant symptom to study during postoperative
19	recovery following treatment for Dupuytren's disease.
20	Level of Evidence: III

22 23	INTRODUCTION
24	The use of patient-reported outcome measures (PROMs) in healthcare has been
25	promoted in the UK (Darzi, 2008, Department of Health, 2010); international
26	standards exist for their study (Mokkink et al., 2010). Several PROMs have been
27	used to evaluate Dupuytren's disease; the 30-item Disability of the Arm, Shoulder
28	and Hand tool (DASH) is the most popular (Ball et al., 2013). However it has been
29	suggested that the DASH may not be valid for use in Dupuytren's disease
30	(Beaudreuil et al., 2011, Packham, 2011), as it does not correlate closely with
31	angular deformity (Engstrand et al., 2009, Jerosch-Herold et al., 2011, Zyluk and
32	Jagielski, 2007); neither does the QuickDASH (Budd et al., 2011). Furthermore, both
33	include items that assess pain whereas it is claimed that Dupuytren's disease may
34	not be painful (Beaudreuil et al., 2011). Other groups suggest that pain may be
35	present (Hueston, 1963, Rodrigues et al., 2014, von Campe et al., 2012) and
36	treatment-related pain may affect postoperative recovery.
37	Much of the data describing the validity and reliability of the DASH was obtained from
38	mixed cohorts involving upper limb conditions widely accepted as painful (Kennedy
39	et al., 2011). Other than the recent publication of the secondary analysis of a
40	randomised controlled trial (Forget et al., 2014), there is limited data describing the
41	DASH's validity and reliability in Dupuytren's disease specifically.
42	Other PROMs that have been used to assess Dupuytren's disease (Ball et al., 2013),
43	include the Michigan Hand Questionnaire (MHQ) (Chung et al., 1998), the Patient
44	Evaluation Measure (PEM) (Macey et al., 1995), the Unité Rhumatologique des
45	Affections de la Main scale (URAM) (Beaudreuil et al., 2011), and the QuickDASH
46	(Beaton et al., 2005). In a study of patients with a range of hand conditions, the
47	DASH took longer to complete than the PEM, but was quicker than the MHQ (Dias et
48	al., 2008). Patients contributing to research, service evaluation or audit might be
49	asked to complete more than one outcome measure. For example, a specific PROM

and a generic measure to assess health-related quality of life, such as the EuroQol 5
D (EQ5D) (Herdman et al., 2011), may be required to facilitate cost effectiveness
analysis (NICE, 2008). As a result, using PROMs that are quicker for the patient to
complete may be more convenient and facilitate higher response rates.
The QuickDASH comprises 11 of the 30 items in the DASH, and should be quicker to
complete. However, it has not been used extensively in Dupuytren's disease (Ball et
al., 2013).

57 Consensus-based standards for the selection of health status measurement

58 instruments (COSMIN) have been developed (Mokkink et al., 2010). These define

59 different aspects of the validity of PROMs.

- Content validity assesses whether the items that comprise a PROM are an
   adequate reflection of what is trying to be measured. It involves assessing
   the relevance and comprehensiveness of the items in a PROM.
- Construct validity examines hypotheses about the PROM. Such hypotheses
   may relate to its structural validity (internal relationships between items),
- 65 hypothesis testing (assessing its relationship with other PROMs) and

66 differences between groups (cross-cultural validity).

- Internal consistency is the interrelatedness of the items within a PROM. This
  assumes that all of the items that contribute to a summary score actually
  assess the same underlying entity, or factor (e.g. impairment of structures in
  the hand versus restriction of function involving the shoulder), i.e. they are
  'unidimensional'.
- Criterion validity tests a PROM against a 'gold standard'. The only accepted
   methodology for this is the comparison of a shortened PROM against the long
   version (e.g. the QuickDASH against the DASH).

- Responsiveness is the ability to detect change over time. This differs from
   'validity', in that responsiveness assesses a change score, whereas validity
   assesses a single time point score.
- 78 This cross sectional study assessed aspects of content validity, construct validity and
- reliability of the DASH in Dupuytren's disease, and studied its relationship with the
- 80 QuickDASH.
- 81

#### **METHODS**

# 84 Patient recruitment and data collection

The data presented in this study were gathered as part of larger service evaluation. Patient recruitment took place between September 2011 and April 2013. Exclusion criteria were cognitive impairment preventing informed consent and refusal of invitation to participate.

89 The inclusion criteria were primary or recurrent Dupuytren's disease and either

90 patients awaiting fasciectomy or dermofasciectomy at one UK hand surgery centre or

91 patients available for assessment at five UK hand surgery centres 1 year or 5 years

92 (+/- 2 months) after their surgery when the primary author (JR) was available.

93 Patients in the first inclusion criterion group, who were participating in an

94 observational cohort study, were recruited at a routine preadmission clinic visit prior

95 to surgery. Those who were eligible and consented to participate completed the

96 DASH in the clinic prior to surgery. These patients were also sent questionnaires for

97 completion by post at 3 weeks, 6 weeks and 1 year after surgery. Patients who were

98 scheduled for surgery to the left and right hand at different times during the study

99 recruitment period were eligible for recruitment twice. This happened on four

100 occasions.

101 Patients in the second inclusion criterion group, who were participating in a cross

102 sectional study of postoperative outcome, were invited to participate with a letter

103 explaining the project and inviting them to participate on a voluntary basis, with a

104 fixed stipend offered to cover travel expenses. A single surgeon (JR) assessed

105 those who consented to participate in a special clinic. The assessment included

106 collection of demographic data and completion of the 30-item DASH questionnaire.

### 107 Angular measurement: total passive extension deficit (TPED)

Patients who completed PROMs in a clinic (as opposed to completion by post –
which was the case for all 3 and 6 week postoperative measurements) had the

110 passive extension deficit of the treated digit assessed by a single examiner. Total 111 passive extension deficit (TPED) was calculated by adding the measured passive 112 extension deficits of the metacarpophalangeal joint and proximal interphalangeal joint 113 while the other joints of the digit were passively flexed. The distal interphalangeal 114 joint is rarely treated in our practice, and was not readily assessable with the model 115 of goniometer used in the study. The measurement thus minimised the influence of 116 dynamism, but is likely to have underestimated the total active extension loss 117 (Rodrigues et al., 2014).

118 Content validity: relevance of pain questions

The relevance of items assessing pain was assessed by extracting and analysing responses to question 24 of the DASH (which assesses pain, and is question 9 of the QuickDASH) and question 25 of the DASH (which assesses pain during specific activity; it is not in the QuickDASH) at different time points. It was hypothesised that if pain items were relevant, they would change significantly through the recovery period.

#### 125 **Construct validity and reliability**

126 The structural validity of the DASH was investigated by studying the internal 127 relationships of the 30 different items in the DASH (how they related to each other) to 128 assess whether the tool is "unidimensional". When used as instructed by the 129 developer, the DASH generates a single summary 'DASH score', using all of the 30 130 items. This is in contrast to other tools such as the Michigan Hand Questionnaire, 131 which generates several summary scores for different areas. For the single DASH 132 score to be valid, all items contributing to the score should measure, or 'reflect', the 133 same underlying entity or 'factor', in this case upper limb function, i.e. the tool should 134 be unidimensional (Mokkink et al., 2010). To evaluate whether the DASH is 135 unidimensional, exploratory factor analysis (EFA) was employed (Mokkink et al., 136 2010). EFA analyses the relationship between items when completed by different 137 people to identify underlying latent factors that explain the variance in scores; the

138 differences seen between individuals across a population. Some of the relevant 139 concepts involved are defined in Table 1. It was expected that the responses 140 obtained would have a tendency towards low scores and so not fit a normal 141 distribution. This was examined by calculating the kurtosis and skewness for items. 142 If the responses were not normally distributed, then logarithmic transformation of all 143 items would be performed (Pallant, 2010) and their distributions then reassessed 144 prior to factor analysis. EFA may be performed using different statistical methods. In 145 this study, principal axis factoring was used to extract latent factors that were being 146 reflected by the DASH's items, and the number of factors extracted was determined 147 and confirmed by using two different accepted techniques (scree plots and parallel 148 analysis, see Table 1) (Cattell, 1966, Horn, 1965, Patil et al., 2007). If the DASH is 149 unidimensional, then there should only be one factor extracted. Cronbach's alpha 150 was calculated to assess internal consistency. However, this must be interpreted 151 with caution if unidimensionality has not been confirmed.

#### 152 Relationship between the DASH and QuickDASH

153 The DASH summary score was calculated using the standard formula provided:

154

# DASH=((a/b)-1) x 25

Where "a" is the sum of the scores for the responses completed (each response could be scored between one and five), and "b" is the number of responses the patient completed.

The QuickDASH summary score was calculated by extracting the answers to the
relevant 11 questions. The score was calculated using the same formula as for the
DASH, only with these eleven items.

Parametric analyses were used to compare the summary scores of the DASH and
the QuickDASH as both have virtually continuous scales (each scored 0-100), and

- 163 the sample comprised a large number of independent observations. This is in
- 164 keeping with the central limit theorem (Norman, 2010). Pearson's correlation
- 165 coefficients were calculated between the total scores for the DASH and the

166 QuickDASH for a) the total sample and b) for different time point subgroups. If the 167 relationship between the QuickDASH and the DASH was not absolute and did not lie 168 on the line of equality (i.e. the correlation coefficient was less than 1, the maximum 169 possible correlation coefficient), then agreement was also studied. Agreement was 170 assessed by calculating 95% limits of agreement, using Bland-Altman analysis of the 171 difference between the QuickDASH and the DASH (Bland and Altman, 1986). The 172 responsiveness was studied by calculating the effect size (mean change in score / 173 standard deviation of baseline score).

# 174 Handling of incomplete responses

175 If more than three of the 30 responses are missing (i.e. fewer than 27/30 responses 176 provided), then the DASH cannot be calculated (Kennedy et al., 2011). If more than 177 one response of the eleven is missing (i.e. fewer than 10/11 responses provided), 178 then the QuickDASH cannot be calculated (Kennedy et al., 2011). If either occurred, 179 then that questionnaire was excluded from the study. Consequently, some of the 180 remaining questionnaires had some missing data (up to 3/30 responses missing for 181 the DASH, or 1/11 missing for the QuickDASH). As the study of the relationship 182 between the DASH and QuickDASH used summary scores, this was of no 183 consequence for that analysis. In the EFA, all such questionnaires were included for 184 analysis, but with missing responses excluded pairwise. 185 Approvals

This study was a minor element of a larger service evaluation project studying
treatment outcome in Dupuytren's disease. In keeping with UK National Research
Ethics Service guidance, it is exempt from ethical approval. Approval as service
evaluation was prospectively obtained from all centres involved. Local information
governance was prospectively obtained from all centres involved, including Caldicott
Guardian approval where required.

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#### RESULTS

#### 194 **Demographics**

195 768 DASH questionnaires were received. These described the preoperative or postoperative assessment of 527 different procedures. Nine cohort study 196 197 questionnaires were incomplete to the extent that calculation of a summary score 198 was not possible based on the guidance issued with the DASH or the QuickDASH. 199 and they were excluded from all analysis. Thus, 759 DASH questionnaires 200 describing 527 procedures on 523 patients were analysed (this is represented 201 graphically in the online appendix). The 527 procedures comprised 126 needle 202 aponeurotomies (fasciotomies), 327 fasciectomies and 74 dermofasciectomies. The 203 mean age at the time of assessment was 68 (range: 34 to 94) years; 403 of the 523 204 (77%) patients were men. The demographics of patients at time of completion of the 205 759 guestionnaires are shown in Table 2. TPED measurements were made at the 206 time of completion of the DASH scores in 522 of the 759 occasions (109

207 preoperative and 413 postoperative).

#### 208 **Content validity: relevance of pain questions**

209 Question 24 of the DASH (which is question 9 of the QuickDASH) requires 210 participants to rate pain experienced in the arm, shoulder or hand in the preceding 211 week on a scale from 1 (no pain) to 5 (severe pain). This question was completed in 212 750 of the 759 questionnaires studied. The median score for question 24 was 2/5 213 ("mild" pain) for the total study. This was also the case when preoperative responses 214 were studied alone. Sixty-eight patients provided answers to question 24 at each of 215 the preoperative, 3 weeks and 6 weeks time points. When these responses were 216 compared, there was a significant difference between them (p=0.003, repeated measures ANOVA test). Specifically, scores were lower at 6 weeks than at 3 weeks 217 218 (Tukey's multiple comparison test). Question 25 of the DASH (which is not part of 219 the QuickDASH) rates pain when performing a specific activity. It was completed in 220 745 of 759 questionnaires. The median score overall was again 2/5, and this was

also the case for preoperative responses. The median score was 3/5 at 3-weeks
postoperatively, falling back to 2/5 at 6 weeks postoperatively. Sixty-one patients
provided answers to question 25 at each of the preoperative, 3 weeks and 6 week
time points. Again, when these were compared, there was a significant difference
between them (p=0.003, repeated measures ANOVA test). Scores were higher at 3
weeks than preoperatively and were lower at 6 weeks than at 3 weeks (Tukey's
multiple comparison test).

#### 228 **Construct validity and reliability**

Across the subgroup that also had angular deformity measured, the DASH showed

weak correlation with TPED (Pearson's r = 0.30 (95% CIs: 0.22 to 0.38). The

231 QuickDASH also correlated weakly with TPED 0.29 (Pearson's r = 95% CIs: 0.21 to 0.37).

After logarithmic transformation, 29 of the 30 DASH items had normal distributions.

234 Most correlation coefficients between log transformed DASH items were over 0.3.

235 Some correlation between the items is required for EFA, as it studies their inter-

236 relationships. In the EFA, the presence of two major factors was suggested based 237 on all common tests for determining factor numbers (see online appendix, confirmed 238 by parallel analysis) (Cattell, 1966, Horn, 1965, Patil et al., 2007). Hence the DASH 239 was not truly 'unidimensional': its 30 items are likely to reflect two factors rather than 240 one, which is what might be expected given that all DASH items are combined into a 241 single score that is supposed to reflect 'upper limb function'. Thus, combining all 30 242 to make a single DASH score may not be appropriate. These two underlying factors 243 that were identified in the EFA explained 57.5% and 5.3% of variance respectively. 244 The results of the EFA are shown in Table 3, with the prerequisite tests in the footnotes (Kaiser, 1974). The outputs from an EFA, called factor loadings, are the 245 246 correlation coefficients between each item in the DASH questionnaire and the 247 mathematical derived factor generated by the analysis. Specific function items

248 correlated well with Factor 1, whereas patient perception of impairment and

- 249 participation items (including pain-related items) generally correlated with Factor 2.
- The EFA was also rerun using raw, untransformed data, and generated the samepattern of results.
- 252 Cronbach's alpha for the DASH was 0.975. However its interpretation was limited by
- 253 finding that the DASH is potentially not unidimensional. Cronbach's alpha for the
- 254 QuickDASH was 0.933. Both results were consistent with there being redundancy of
- items within the scales.
- 256 Relationship between the DASH and QuickDASH
- 257 Across the entire study, the QuickDASH was higher than the DASH indicating
- apparently worse upper limb function (mean difference 1.6 (95%Cls: 1.3 1.8),
- 259 paired t test. However the QuickDASH correlated very well with the DASH,
- 260 (Pearson's *r* was 0.98 (95% CIs: 0.98 0.99), as shown in Chart 1).
- 261 Linear regression analysis was performed with the Y-intercept constrained to y=0, as
- the QuickDASH must equal zero if the DASH equals zero. Runs test confirmed that
- there was no significant deviation of the residuals from the model in the linear
- regression analysis (p=0.228). The slope for the relationship between the two was:
- 265 QuickDASH =  $1.054 \times DASH$ .
- 266 Similar correlations were seen in separate preoperative, 3 week, 6 week, 1 year and
- 5 year follow-up subgroup analyses (Table 4).
- 268 The 95% limits of agreement between the QuickDASH and the DASH were -5.8 to
- +8.9 (Chart 2). As relatively few differences were outside the 95% limits of
- agreement for mean scores under 30 (those with good upper limb function), further
- 271 Bland-Altman analyses were performed for scores considered asymptomatic
- 272 (<15/100), and scores considered symptomatic (>15/100) by the DASH's creators
- 273 (Kennedy et al., 2011). When the mean of the DASH and QuickDASH for a patient
- was 15 or less (asymptomatic upper limb function), the 95% limits of agreement were
- -3.3 to +5.5, and when the mean was over 15 (symptomatic upper limb function),
- 276 they were -7.8 to +12.3.

- 277 In terms of responsiveness from preoperative measurement to one year
- 278 postoperative measurement, the effect size for the DASH was 0.58, and the effect
- size for the QuickDASH was 0.64.

#### DISCUSSION

#### 283 Present study findings

This study assessed several aspects of the validity and reliability of the DASH in Dupuytren's disease, and its relationship with the QuickDASH.

286 PROM items covering pain have been previously criticised in Dupuytren's disease,

but our patients did report preoperative upper limb pain. This may have been due to

other comorbid upper limb conditions rather than Dupuytren's disease, nonetheless,

this would affect the overall function of their upper limb. Furthermore, pain levels

290 rose early after surgery, and then fell back. This is important to capture, as

291 postoperative pain may differ between treatments, and affect early recovery. Our

data support the relevance of assessing pain when treating Dupuytren's disease,

such that scales that do not measure pain (such as the Unité Rhumatologique des

Affections de la Main (URAM) (Beaudreuil et al., 2011)) may not provide a

295 comprehensive assessment as a result.

The DASH is not unidimensional, and its task-based items loading better with one construct, and patient perception items load best with another. Some other PROMs are multidimensional and generate separate subscale scores. Examples include the Michigan Hand Questionnaire, which has distinct subscales for entities that might be expected to behave as different 'constructs', such as function, pain and work (Chung et al., 1998). However, the DASH is designed to generate a single summary score, and its items were not necessarily selected to measure specific distinct constructs.

As a result, the different constructs identified here may not be easily interpreted. The items loading with Factor 1 might do so as they are all task-based items. The items that load with Factor 2 might do so as they are patient perception items. However,

an alternative explanation might be that items that load well with Factor 1 do so as

307 they reflect activities involving the entire upper limb activities, and the items that load

308 well with Factor 2 specifically reflect the patient's experience of Dupuytren's disease.

Either way, these data suggest that the DASH's single summary score may not be appropriate in Dupuytren's disease. However, Factor 2 accounted for relatively little variance, such that its presence may not completely preclude the use of the DASH and its summary score in Dupuytren's disease. If used in future studies, its selection as an endpoint for future studies should be carefully considered.

314 Interpreting whether agreement is adequate or not is a clinical decision (Bland and 315 Altman, 1986). Given that the minimum detectable change at the 95% confidence 316 level (MDC<sub>95</sub>) for the DASH is around 13, and that the MDC<sub>95</sub> of the QuickDASH is 317 around 16 (Kennedy et al., 2011), then the 95% limits of agreement seen here are of 318 similar magnitude to both tools' abilities to detect true change, though these MDCs 319 have not been specifically confirmed in Dupuytren's disease. Therefore, we believe 320 that the level of agreement seen here would support the use of the QuickDASH as 321 an alternative to the DASH if either were considered appropriate in Dupuytren's 322 disease. Furthermore, their responsiveness was similar.

Our large sample size allowed meaningful subgroup analysis, which demonstrated
that close correlation between the two tools was seen at preoperative, early
postoperative, and late outcome time points.

326 Limitations

327 There are limitations to this study. Our sample included some patients who provided

328 more than one measurement. However, multiple measurements over time

329 (preoperative, 3 weeks postop, 6 weeks post-operative and 1 year) do not constitute

330 replicate measurements. They are best considered as independent assessments, as

the patient's functional status is expected to be different at each time point, due to

332 treatment of disease and progressive recovery.

We used logarithmic transformation in an attempt to normalise the positive skew and kurtosis encountered. Suggested methods for handling skewed data vary (Ferguson and Cox, 1993, Floyd and Widaman, 1995). However, similar results were obtained when the EFAs were run using raw, untransformed data.

337 Here, as in previous studies (Gummesson et al., 2006, Niekel et al., 2009), the 338 QuickDASH and DASH were calculated from a single set of responses, on the DASH 339 guestionnaire's proforma. Intra-observer reproducibility, or test-retest reliability, is 340 not observed when using a single set of responses. However, we believe that our 341 chosen methodology was the most appropriate to fulfil the specific objective of this 342 study. This was to determine whether the QuickDASH formula demonstrates 343 acceptable criterion validity with the longer DASH formula for a given set of 344 responses, in keeping with the COSMIN checklist (Mokkink et al., 2010). 345 Test-retest reliability has been shown to be consistently high in previous studies, as 346 summarised in the user manual for the DASH and the QuickDASH (Kennedy et al., 347 2011). The question of the present study is: "For a given set of responses, does the 348 QuickDASH formula give you the same summary score as the DASH formula?" If 349 patients had completed the DASH and then the QuickDASH, then there would have 350 been additional error due to intra-observer reproducibility. This would have affected 351 the ability to answer the study question.

#### 352 Relationship to existing literature

Although the DASH is the most commonly used Patient Reported Outcome Measure
(PROM) tool in Dupuytren's research (Akhavani et al., 2015, Ball et al., 2013),

355 Dupuytren's-specific outcome measures are available, for example the URAM scale

356 (Beaudreuil et al., 2011) and the Southampton Dupuytren's score (Mohan et al.,

357 2014). Preoperative Dupuytren's disease is considered painless, and pain is not

assessed in the URAM (Beaudreuil et al., 2011). However, pain has been described

as a symptom, particularly related to Dupuytren's nodules (Hueston, 1963, Rodrigues

360 et al., 2014, von Campe et al., 2012). Our data demonstrated that pain was present

- 361 preoperatively, increased at 3 weeks post-operative compared to baseline, returned
- to the preoperative level by 6 weeks postop, and did not disappear completely.
- 363 The QuickDASH was produced from the DASH using item reduction methodology
- 364 (Beaton et al., 2005). The two showed good correlation in mixed cohorts of hand

365 surgery patients (Gummesson et al., 2006, Niekel et al., 2009). However, correlation 366 is not appropriate for studying agreement (Bland and Altman, 1986, Bland and 367 Altman, 1990, Schuck, 2004). We are not aware of any studies assessing 368 agreement between the DASH and the QuickDASH in Dupuytren's disease 369 specifically, with the technique recommended by Bland and Altman (Bland and 370 Altman, 1990). Studying the strength of relationship between the two correlated 371 measures, as has been done elsewhere, may conceal absolute differences in values 372 between them. Such differences are unmasked when agreement is studied using 373 other techniques, as has been done here (Bland and Altman, 1986, Bland and 374 Altman, 1990).

375 Poor correlation between angular deformity and the DASH has been previously

reported in Dupuytren's disease (Degreef et al., 2009, Engstrand et al., 2009,

377 Jerosch-Herold et al., 2011, Zyluk and Jagielski, 2007). This has led to the

378 suggestion that the DASH may not be valid for use in Dupuytren's disease

379 (Packham, 2011), although the basis for this claim was a series of only seven patient

380 interviews (Pratt and Byrne, 2009). Angular correction remains a very popular

measure of outcome in the treatment of Dupuytren's disease (Trickett et al., 2014,

Verheyden, 2015). However, such conclusions are dependent on angular deformity
being a 'gold standard' for assessing Dupuytren's disease, allowing the assessment
of criterion validity of outcome measures including PROMs. This is not appropriate:
hand function is a latent construct that cannot be measured directly, and so no true
'gold standard' for it can exist (Mokkink et al., 2010).

Concerns about the absence of the expected unidimensionality for the DASH have been previously reported. Forget and collagues investigated the structural validity of the DASH as the secondary analysis of a randomised controlled trial of splinting in Dupuytren's disease (Forget et al., 2014). EFA was performed on preoperative and early postoperative DASH scores from 153 fasciectomies and dermofasciectomies randomised to receiving postoperative splinting or no postoperative splinting. It also

393 demonstrated a lack of unidimensionality for the DASH. However the different 394 groups of DASH items that loaded well on different factors could not be as easily 395 explained as they can in the present study. The present study evaluated 396 preoperative, early postoperative and late postoperative outcomes of over 500 397 procedures, including needle aponeurotomies, and in contrast, our EFA showed a 398 clear division between task items and patient perception items. Another study that 399 investigated a heterogeneous cohort comprising a range of upper limb conditions 400 (mainly affecting the shoulder) and only a small minority with Dupuytren's disease 401 found the DASH items loaded on three distinct factors in EFA, and this was 402 confirmed using other techniques classified as confirmatory factor analysis 403 (Franchignoni et al., 2010). As with the present study, patient perception items 22 to 404 30 loaded separately from the main factor, but they also found that of the other items, 405 those relating to manual function loaded distinctly from those assessing shoulder 406 functions. Given that the equivalent of the present study's Factor 1 was two separate 407 factors in a mixed cohort including shoulder patients suggests that Factor 1 is not a 408 clean and reliable indicator of hand function in Dupuytren's disease. Our data further 409 question the structural validity of the DASH in Dupuytren's disease, and by 410 association, the QuickDASH. Due to this, we believe that it is neither necessary nor 411 appropriate to subject the DASH to further analyses such as Rasch analysis (Tesio, 412 2003). Instead, other PROMs that also assess pain, such as the MHQ or the PEM, 413 may be more appropriate for the study of Dupuytren's disease.

### 414 Summary

This study supports the assessment of pain when studying recovery from
Dupuytren's surgery. The QuickDASH show acceptable agreement with the full
DASH. However, the DASH may not be structurally valid for use in Dupuytren's
disease, and further study of existing PROMs for use in Dupuytren's disease is
needed.

420

421 422	REFERENCES
423	ir L. A review of the classification of Dupuytren's disease. J Hand Surg Brit Eur. 2015,
424	40: 155-65.
425	Ball C, Pratt AL, Nanchahal J. Optimal functional outcome measures for assessing
426	treatment for Dupuytren's disease: a systematic review and recommendations for
427	future practice. BMC Musculoskelet Disord. 2013, 14: 131.
428	Beaton DE, Wright JG, Katz JN. Development of the QuickDASH: comparison of
429	three item-reduction approaches. J Bone Joint Surg Am. 2005, 87: 1038-46.
430	Beaudreuil J, Allard A, Zerkak D et al. Unite Rhumatologique des Affections de la
431	Main (URAM) scale: development and validation of a tool to assess Dupuytren's
432	disease-specific disability. Arthrit Care Res. 2011, 63: 1448-55.
433	Bland JM, Altman DG. Statistical methods for assessing agreement between two
434	methods of clinical measurement. Lancet. 1986, 1: 307-10.
435	Bland JM, Altman DG. A note on the use of the intraclass correlation coefficient in
436	the evaluation of agreement between two methods of measurement. Comput Biol
437	Med. 1990, 20: 337-40.
438	Budd HR, Larson D, Chojnowski A, Shepstone L. The QuickDASH score: a patient-
439	reported outcome measure for Dupuytren's surgery. J Hand Ther. 2011, 24: 15-20.
440	Cattell RB. The Scree Test for the Number of Factors. Mutlivar Behav Res. 1966, 1:
441	245-76.
442	Chung KC, Pillsbury MS, Walters MR, Hayward RA. Reliability and validity testing of
443	the Michigan Hand Outcomes Questionnaire. J Hand Surg Am. 1998, 23: 575-87.
444	Darzi A. High quality care for all: NHS Next Stage Review final report. London: TSO,
445	2008.
446	http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/PublicationsPolicyAnd
447	Guidance/DH_085825 (27th June 2012).

- 448 Degreef I, Vererfve PB, De Smet L. Effect of severity of Dupuytren contracture on
- disability. Scand J Plast Recons. 2009, 43: 41-2.
- 450 Department of Health. Equity and Excellence: Liberating the NHS. London 2010.
- 451 <u>http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/PublicationsPolicyAnd</u>
- 452 <u>Guidance/DH\_117353</u> (26th June 2012).
- 453 Dias JJ, Rajan RA, Thompson JR. Which questionnaire is best? The reliability,
- 454 validity and ease of use of the Patient Evaluation Measure, the Disabilities of the
- 455 Arm, Shoulder and Hand and the Michigan Hand Outcome Measure. J Hand Surg
- 456 Brit Eur. 2008, 33: 9-17.
- 457 Engstrand C, Boren L, Liedberg GM. Evaluation of activity limitation and digital
- 458 extension in Dupuytren's contracture three months after fasciectomy and hand
- therapy interventions. J Hand Ther. 2009, 22: 21-6.
- 460 Ferguson E, Cox T. Exploratory Factor Analysis: A Users' Guide. Int J Select Assess.
- 461 1993, 1: 84-94.
- 462 Floyd FJ, Widaman KF. Factor Analysis in the Development and Refinement of
- 463 Clinical Assessment Instruments. Psychol Assessment. 1995, 7: 286-99.
- 464 Forget NJ, Jerosch-Herold C, Shepstone L, Higgins J. Psychometric evaluation of
- the Disabilities of the Arm, Shoulder and Hand (DASH) with Dupuytren's contracture:
- 466 validity evidence using Rasch modeling. BMC Musculoskelet Disord. 2014, 15: 361.
- 467 Franchignoni F, Giordano A, Sartorio F, Vercelli S, Pascariello B, Ferriero G.
- 468 Suggestions for refinement of the Disabilities of the Arm, Shoulder and Hand
- 469 Outcome Measure (DASH): a factor analysis and Rasch validation study. Arch Phys
- 470 Med Rehabil. 2010, 91: 1370-7.
- 471 Gummesson C, Ward MM, Atroshi I. The shortened disabilities of the arm, shoulder
- 472 and hand questionnaire (QuickDASH): validity and reliability based on responses
- 473 within the full-length DASH. BMC Musculoskelet Disord. 2006, 7: 44.
- 474 Herdman M, C. G, Lloyd A et al. Development and preliminary testing of the new
- 475 five-level version of EQ-5D (EQ-5D-5L). Qual Life Res. 2011, 20: 1727-36.

- 476 Horn JL. A Rationale and Test for the Number of Factors in Factor Analysis.
- 477 Psychometrika. 1965, 30: 179-85.
- 478 Hueston JT. Principles of management. In: Hueston JT (Ed.) Dupuytren's
- 479 contracture. London, E &S Livingstone, 1963: 76-94.
- 480 Jerosch-Herold C, Shepstone L, Chojnowski A, Larson D. Severity of contracture and
- 481 self-reported disability in patients with Dupuytren's contracture referred for surgery. J
- 482 Hand Ther. 2011, 24: 6-10.
- 483 Kaiser HF. An index of factorial simplicity. Psychometrika. 1974, 39: 31-6.
- 484 Kennedy C, Beaton D, S S, S M, Bombardier C. *The DASH and QuickDASH*
- 485 Outcome Measure User's Manual, Third ed. Toronto, Institute for Work and Health,
- 486 2011.
- 487 Macey AC, Burke FD, Abbott K et al. Outcomes of hand surgery. British Society for
- 488 Surgery of the Hand. J Hand Surg Brit Eur. 1995, 20: 841-55.
- 489 Mohan A, Vadher J, Ismail H, Warwick D. The Southampton Dupuytren's Scoring
- 490 Scheme. J Plast Surg Hand Surg. 2014, 48: 28-33.
- 491 Mokkink LB, Terwee CB, Knol DL et al. The COSMIN checklist for evaluating the
- 492 methodological quality of studies on measurement properties: a clarification of its
- 493 content. BMC Med Res Methodol. 2010, 10: 22.
- 494 NICE. Guide to the methods of technology appraisal. London: NICE, 2008.
- 495 <u>http://www.nice.org.uk/aboutnice/howwework/devnicetech/technologyappraisalproce</u>
- 496 <u>ssguides/?domedia=1&mid=B52851A3-19B9-E0B5-D48284D172BD8459</u> (26th June
- 497 2012).
- 498 Niekel MC, Lindenhovius AL, Watson JB, Vranceanu AM, Ring D. Correlation of
- 499 DASH and QuickDASH with measures of psychological distress. J Hand Surg Am.
- 500 2009, 34: 1499-505.
- 501 Norman G. Likert scales, levels of measurement and the "laws" of statistics. Adv
- 502 Health Sci Educ Theory Pract. 2010, 15: 625-32.

- 503 Packham T. Clinical commentary in response to: Severity of contracture and self-
- 504 reported disability in patients with Dupuytren's contracture referred for surgery. J
- 505 Hand Ther. 2011, 24: 12-4.
- 506 Pallant J. Part 3: Preliminary analyses: Manipulating the data. SPSS Survival
- 507 *Manual*, 4th ed. Maidenhead, Open University Press, 2010.
- 508 Patil VH, Singh SN, Mishra S, Donovan TD. Parallel Analysis Engine to Aid
- 509 Determining Number of Factors to Retain [Computer Software]. 2007.
- 510 <u>http://smishra.faculty.ku.edu/parallelengine.htm</u> (2nd August 2014).
- 511 Pratt AL, Byrne G. The lived experience of Dupuytren's disease of the hand. J Clin
- 512 Nurs. 2009, 18: 1793-802.
- 513 Rodrigues JN, Zhang W, Scammell BE, Davis TR. Dynamism in Dupuytren's
- 514 contractures. J Hand Surg Brit Eur. 2014, 40: 166-70.
- 515 Rodrigues JN, Zhang W, Scammell BE, Davis TR. What patients want from the
- 516 treatment of Dupuytren's disease -- is the Unité Rhumatologique des Affections de la
- 517 Main (URAM) scale relevant? J Hand Surg Brit Eur. 2014, 40: 150-4.
- 518 Schuck P. Assessing reproducibility for interval data in health-related quality of life
- 519 questionnaires: which coefficient should be used? Qual Life Res. 2004, 13: 571-86.
- 520 Tesio L. Measuring behaviours and perceptions: Rasch analysis as a tool for
- rehabilitation research. J Rehabil Med. 2003, 35: 105-15.
- 522 Trickett RW, Savage R, Logan AJ. Angular correction related to excision of specific
- 523 cords in fasciectomy for Dupuytren's disease. J Hand Surg Brit Eur. 2014, 39: 472-6.
- 524 Verheyden JR. Early outcomes of a sequential series of 144 patients with
- 525 Dupuytren's contracture treated by collagenase injection using an increased dose,
- 526 multi-cord technique. J Hand Surg Brit Eur. 2015, 40: 133-40.
- 527 von Campe A, Mende K, Omaren H, Meuli-Simmen C. Painful nodules and cords in
- 528 Dupuytren disease. J Hand Surg Am. 2012, 37: 1313-8.
- 529 Zyluk A, Jagielski W. The effect of the severity of the Dupuytren's contracture on the
- 530 function of the hand before and after surgery. J Hand Surg Brit Eur. 2007, 32: 326-9.

Appendix 1: Flow diagram demonstrating time points from which complete questionnaires were received



Cross sectional study







The curve flattens from factor 3 onwards. Based on Cattell's test, this supports the extraction of factors 1 and 2. This was confirmed using parallel analysis, and only these two factors had Eigenvalues over one.



Chart 1: Scatterplot of QuickDASH versus DASH (n=759).

Chart 2: Bland-Altman plot of total sample, with 95% limits of agreement shown as dotted lines



Bland-Altman plot of total (n=759)

### Table 1: Data handling and relevant statistical concepts

Aspect Studied	Method(s) Used	Concept(s)	Description
Content validity	Cohort study of pain	Comprehensiveness	Content validity considers whether PROM items are relevant to what is being measured, and
	items	& Relevance	whether the scale overall is comprehensive.
Distribution of item	Kurtosis & skewness	Kurtosis	Defines the sharpness of the peak of a distribution of data
responses			
		Skewness	Defines the amount of asymmetry of a distribution of data
Construct validity	Exploratory Factor	Unidimensionality	All items contributing to a summary score reflect the same underlying factor (some PROMs
(structural validity)	Analysis		generate more than one summary score, each describing a different subscale of the PROM)
		Factor	Factor analysis aims to describe the variation in measured items that correlate with each other in
			terms of fewer unobserved 'factors'
		Principal axis factoring	A form of factor analysis in which factors are extracted based on common variance, rather than
			total variance.
		Eigenvalue	Describes the amount of the total variance that is explained by a particular factor
		Catell's scree plot	Determines the number of factors to extract. All potential factors are plotted in order of
			Eigenvalues (see Chart 1). The turning point where the connecting line flattens sharply is the point
			at which the last significant factor has been passed, as further factors represent a flat level of
			background noise (Catell, 1966).

		Parallel analysis	Determines the number of factors to extract. A second set of Eigenvalues is generated, but which
			are based entirely on chance. All factors in the real model with Eigenvalues greater than their
			counterparts in the chance model are significant and are then extracted. Avoids the risk of bias
			that exists with scree plot interpretation (Horn, 1965).
		Kaiser-Meyer-Olkin	Assesses sampling adequacy. It lies between 0-1, describing the proportion of variance that is
		statistic	common variance. The minimum acceptable level for analysis being 0.6, and $>0.9$ being
			described as 'marvellous' (Kaiser, 1974).
		Bartlett's test of	Assesses whether an identity matrix would result if correlations between the items included were
		sphericity	studied, i.e. whether the correlation between all of the DASH items is zero. Some correlation
			between items is needed for EFA. A significant result is achieved if the data is suitable for EFA.
		Factor loading	The output of EFA. Described how closely an item correlates with a factor.
		Rotation	Presents the factor loadings in a manner that makes interpretation more straightforward, resulting
			in a pattern matrix. Only possible when more than one factor is extracted. When done, the
			majority of the items may each show strong loading with one of the factors, a situation described
			as simple structure.
Reliability	Cronbach's alpha	Internal consistency	Studies the inter-relatedness between items within a scale. However, it is dependent on the scale
			being unidimensional and reflective
Relationship between	Bland-Altman plots	Agreement	Studies the relationship between two variables that are expected to correlate. In such
DASH and			circumstances, reporting correlation is common, but may not be appropriate.
QuickDASH			

# 1 Table 2: Subgroups and demographics of completed questionnaires

Subgroup	Number of complete	Procedure type	Mean age at	Proportion male
	questionnaires (incompletes)		assessment (range)	
Total	759 (9)	130 aponeurotomies	68 (34-94)	592/759 (78%)
		494 fasciectomies		
		135 dermofasciectomies		
Preoperative	114 (3)	N/A	67 (34-90)	88/114 (77%)
3 weeks postop	83 (0)	2 aponeurotomies	67 (34-90)	66/83 (80%)
		57 fasciectomies		
		24 dermofasciectomies		
6 weeks postop	85 (3)	2 aponeurotomies	68 (34-90)	68/85 (80%)
		62 fasciectomies		
		21 dermofasciectomies		

1 year postop	316 (3)	<ul><li>105 aponeurotomies</li><li>168 fasciectomies</li><li>43 dermofasciectomies</li></ul>	68 (35-91)	249/316 (79%)
5 years postop	161 (0)	19 aponeurotomies 126 fasciectomies 16 dermofasciectomies	71 (39-89)	121/161 (75%)

- 1 Table 3: Exploratory factor analysis output of log-transformed DASH items (Pattern
- 2 matrix with two-factor solution and oblimin rotation)
- 3 [The pattern coefficients may be considered similar to correlation coefficients
- 4 between the item and the factors was derived in EFA. They may range from -1
- 5 (perfect inverse correlation), through 0 (no correlation), to +1 (perfect correlation)]

Item	Item question	Pattern coefficient	Pattern coefficient
number		with factor 1	with factor 2
12	Difficulty changing a light bulb overhead	0.86	-0.05
14	Difficulty washing your back	0.84	-0.09
7	Difficulty doing heavy household chores	0.84	0.05
5	Difficulty pushing open a heavy door	0.82	-0.06
13	Difficulty washing or blow drying hair	0.82	-0.03
9	Difficulty making a bed	0.80	0.03
16	Difficulty using a knife to cut food	0.80	0.01
11	Difficulty carrying a heavy object (>10 lbs)	0.80	0.04
8	Difficulty gardening	0.80	0.08
6	Difficulty placing an object on a shelf above	0.78	-0.04
	head		
17	Difficulty with recreational activities that require	0.78	-0.05
	little effort		
4	Difficulty preparing a meal	0.77	0.07
18	Difficulty with recreational activities in which	0.73	0.13
	force is taken through the limb		
21	Difficulty with sexual activities	0.72	-0.11
19	Difficulty with recreational activities in which	0.71	0.12
	the arm moves freely		
3	Difficulty turning a key	0.69	0.03
15	Difficulty putting on a pullover sweater	0.68	0.09
20	Difficulty managing transportation needs	0.68	0.02
10	Difficulty carrying a shopping bag/briefcase	0.67	0.15
2	Difficulty writing	0.63	0.02
1	Difficulty opening a tight or new jar	0.61	0.15

To what extent has your problem interfered	0.45	0.37
with normal social activities?		
To what extent has your problem interfered	0.44	0.42
with work or other daily activities?		
Rate your arm, shoulder or hand pain	-0.06	0.90
Rate your arm, shoulder or hand pain when	-0.02	0.87
performing a specific activity		
Rate the stiffness in your arm, shoulder or	0.08	0.72
hand		
Rate the weakness in your arm, shoulder or	0.17	0.69
hand		
How much difficulty have you had sleeping	0.10	0.62
because of pain in the limb?		
Rate the tingling in the arm, shoulder or hand	-0.04	0.59
Is this true: I feel less capable, confident or	0.29	0.51
useful because of the limb problem?		
	To what extent has your problem interfered with normal social activities? To what extent has your problem interfered with work or other daily activities? Rate your arm, shoulder or hand pain Rate your arm, shoulder or hand pain when performing a specific activity Rate the stiffness in your arm, shoulder or hand Rate the weakness in your arm, shoulder or hand How much difficulty have you had sleeping because of pain in the limb? Rate the tingling in the arm, shoulder or hand Is this true: I feel less capable, confident or useful because of the limb problem?	To what extent has your problem interfered0.45with normal social activities?0.44To what extent has your problem interfered0.44with work or other daily activities?-0.06Rate your arm, shoulder or hand pain-0.02performing a specific activity-0.02Rate the stiffness in your arm, shoulder or0.08hand-0.17hand-0.17How much difficulty have you had sleeping0.10because of pain in the limb?-0.04Is this true: I feel less capable, confident or0.29useful because of the limb problem?-0.04

7 N.B. Items have been sorted by size of factor loading. Large factor loadings (>0.3)

8 are shown in bold.

9 The Kaiser-Meyer-Olkin statistic for the analysis was 0.97 and Bartlett's test of

10 sphericity was highly statistically significant (Kaiser, 1974).

TIME POINT	MEAN DASH	MEAN	PEARSON's r
	(95% CIs)	QuickDASH	(95% CIs)
		(95% CIs)	
Preoperative	27	28	0.98 (0.97-0.99)
	(23-31)	(24-32)	
3 week postoperative	37	41	0.97 (0.95-0.98)
	(33-42)	(36-46)	
6 week postoperative	22	24	0.98 (0.97-0.99)
	(18-26)	(20-28)	
1 year postoperative	11	12	0.99 (0.98-0.99)
	(9-13)	(10-14)	
5 years postoperative	11	12	0.97 (0.96-0.98)
	(9-13)	(10-15)	

1 Table 4: Mean DASH and QuickDASH scores and their correlations, by time point