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Comprehension of pictograms for pain quality and pain affect in adults with Down syndrome

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ABSTRACT

Background Adults with Down syndrome (DS) are at risk for age-related painful physical conditions, but also for under-reporting pain. Pictograms may facilitate self-report of pain, because they seem suitable for the global visual processing in DS and for iconic representation of abstract concepts.
Method Participants ($N = 39$, M age = 41.2) assigned pain qualities to pictograms, rated pain affect levels in facial scales (pictograms vs. drawn faces), and performed cognitive tests.
Results Recognition of all intended pain qualities was above chance level. Pain affect levels of both facial scales were ordered equally well. Both facial scales were preferred equally well. Comprehension of the 3 scales was positively associated with mental age, receptive language ability, and verbal memory. Most participants (74%) had pictograms in their direct environment, mainly to communicate activities or objects.
Conclusion Using pictograms may optimise communication about pain for a subgroup of cognitively higher functioning adults with DS.

KEYWORDS
pain assessment; pictograms; communication tools; Down syndrome; adults

Introduction

It is worthwhile examining the possibility of people with intellectual disability using self-reporting scales for pain, because these people may have difficulty understanding and communicating abstract concepts such as pain (Tuffrey-Wijne & McEnhill, 2008) but may be able to use simple self-reporting scales for pain (Herr, Coyne, McCaffery, Manworren, & Merkel, 2011; Zabalia, 2013). The search for appropriate self-reporting scales is especially relevant for adults with Down syndrome (DS) for four reasons. First, DS is a common genetic cause of intellectual disability. According to a study with data from 14 countries (Australia, Canada, United States, Israel, Mexico, and European countries) for the period 1993–2004, the mean prevalence of DS was 8.3 per 10.000 total births (Cocchi et al., 2010). Population-based studies show that a moderate level of intellectual disability is most prevalent in DS (Coppus et al., 2006; Glasson et al., 2002; Holland, Hon, Huppert, Stevens, & Watson, 1998). Therefore, people with DS form a relatively large subgroup who might have the intellectual abilities to understand simple self-reporting scales for pain. Second, accelerated ageing (Nakamura & Tanaka, 1998) combined with an increased life expectancy (Glasson et al., 2002) make adults with DS extra vulnerable for age-related painful conditions. Detecting painful physical conditions and monitoring pain experience are thus important. Third, a clearly visible scale is needed, because ocular disorders are common in DS (Hestnes, Sand, & Fostad, 1991). Fourth, it is known that people with DS scarcely complain about pain (Smith, 2001). A self-reporting scale may therefore facilitate communication about pain.

Possible explanations for the clinical observation that people with DS rarely complain about pain are deficits in expressive language abilities (Laws & Bishop, 2004; Næss, Lyster, Hulme, & Melby-Lervåg, 2011), aberrant coping such as expressing medical problems as behavioural problems (Smith, 2001), aberrant pain responses (Mafrica, Schifilliti, & Fodale, 2006), and a possible lower pain experience resulting from neural abnormalities (de Knegt & Scherder, 2011; Risser et al., 1996). Due to the various possible causes mentioned in this paragraph, it is worthwhile to attempt stimulating self-reporting of pain in adults with DS by using scales.

The question arises whether facial scales for self-reporting pain could be used in individuals with DS.

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These individuals show deficits in recognising facial expressions of fear, surprise, anger, sadness, and a neutral state (Hippolyte, Barisnikov, & Van der Linden, 2008; Hippolyte, Barisnikov, Van der Linden, & Detraux, 2009; Kasari, Freeman, & Hughes, 2001; Porter, Coltheart, & Langdon, 2007; Williams, Wishart, Pitcairn, & Willis, 2005; Wishart & Pitcairn, 2000; Wishart, Cebula, Willis, & Pitcairn, 2007). However, other researchers have not found statistically significant abnormalities in performance on emotion-matching tasks by children with DS (Martínez-Castilla, Burt, Borgatti, & Gagliardi, 2015; Pochon & Declercq, 2013). As far as we know, only one study has included an emotion recognition task in the context of pain assessment of people with intellectual disability (Zabalia & Corfec, 2008). In that study, no statistically significant difference was found between people with DS and typically developing children in recognising happiness, anger, fear, sadness, surprise, and disgust in facial expressions. The group with DS was more able to use the Faces Pain Scale Revised (i.e., a facial pain scale starting with a neutral face) for rating pain intensity in pictures of painful situations than the Visual Analogue Scale (i.e., VAS; a vertical line from no pain to extreme pain; Zabalia & Corfec, 2008).

Another study involving self-reporting scales for pain shows that children with intellectual disability who underwent venipuncture were able to respond on the VAS and 12.5% selected a sad face on the Wong–Baker Faces Scale (i.e., a facial scale starting with a smiling face), but the participants with DS needed enlarged body parts in the Eland Scale (i.e., the front and back side of a human figure) to mark their pain location (Benini et al., 2004). Although only 21% of children with intellectual disability completed a series of comprehension tasks for a numerical rating scale (i.e., magnitude and ordering tasks with numbered blocks and cards, a numeric rating task of pain intensity in three schematic faces, and explaining the pain level of the number zero), it was unclear how many participants failed to comprehend the facial task (Fanurik, Koh, Harrison, Conrad, & Tomerun, 1998). Adults with intellectual disability were able to recognise different pain intensity levels in photographs of facial expression by indicating the intensity levels with the Coloured Analogue Scale (i.e., CAS; a VAS that is coloured increasing red; Bromley, Emerson, & Caine, 1998). Only 65% of adults with intellectual disability comprehended the CAS in another study, but it was not specified how this was observed (LaChapelle, Hadjistavropoulos, & Craig, 1999).

As far as we know, pictogram scales have not been used in pain assessment studies, and such stimuli could be beneficial for adults with DS for three reasons. First, people with DS process visual stimuli in a holistic, global manner (Bellugi, Lichtenberger, Mills, Galaburda, & Korenberg, 1999) and tend to use a featural processing strategy of facial expressions (i.e., analysis of the main individual features of the face, such as eyes and mouth; Hippolyte et al., 2008). The black-and-white nature may help to discriminate the key elements, such as face and body, from the background and the schematic features within the key elements could aid visual processing. Second, pictograms may also facilitate communication about abstract pain-related concepts such as pain quality. Pain quality refers to how pain feels in subjective terms of somatosensory sensations such as burning (Jensen et al., 2006). Pictograms including symbols such as a fire combined with a facial expression of pain may form iconic, concrete representations (see Figure 1). Third, pictograms are already often used for communication by people with intellectual disability (Fujisawa, Inoue, Yamana, & Hayashi, 2011; Kåhlin & Haglund, 2009; Renblad, 2000) and could thus be easily integrated in their daily routines.

In sum, a next step to examine the possibilities for self-report of pain by people with intellectual disability is to develop pictograms and to test the comprehension of these in adults with DS. It is therefore important to examine whether a scale of facial pictograms (see Figure 2) and an existing self-reporting scale for pain with drawn faces (see Figure 3) are comparable concerning comprehension and preference.

The research questions for the present pilot study were (a) Do adults with DS recognise the intended pain qualities in pictograms above chance level and is this related to cognitive functioning or familiarity with pictograms? (b) Do adults with DS comprehend facial pictograms better than drawn faces for pain affect and is this related to cognitive functioning or familiarity with pictograms? and (c) Do adults with DS prefer facial pictograms over drawn faces for pain affect and is this related to familiarity with pictograms? Cognitive functioning included mental age, receptive language ability, and verbal memory, because mental age (Kasari et al., 2001) and receptive language ability (Hippolyte et al., 2008, 2009) seem related to emotion recognition in DS and verbal memory is impaired in DS (Jarrold, Baddeley, & Phillips, 1999; Næss et al., 2011).

Method

Ethical approval

The Medical Ethical Committee of the VU University Medical Center approved the study and informed consent procedure (file NL33540.029.11).

Participants

Participants with DS were recruited via seven care centres for people with intellectual disability with

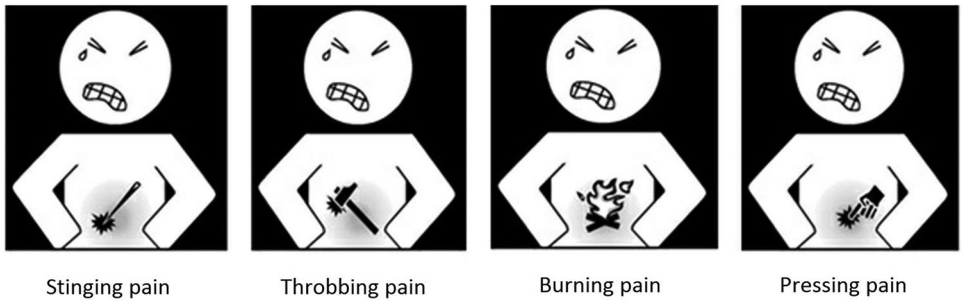

Figure 1. Pictograms for pain qualities. Participants were requested to match them with pain qualities that were read aloud (written descriptions not presented to participants). Developed by Sclera (http://www.sclera.be) in 2012.

permission from the management board of the care centres. Before the start of the study, the care centres' caregivers and behaviour specialists assessed inclusion and exclusion criteria. Inclusion criteria were 18 years of age or older, speaking and understanding Dutch, the capability to verbally answer simple questions, and a clinical impression of testability. This latter inclusion criterion implied that adults with DS of all levels of intellectual disability and adults with both DS and dementia could participate, as long as they could comprehend the instructions of at least some of the tests. Exclusion criteria were neurological disorders such as cerebrovascular accidents or tumors; the use of antipsychotics, anticonvulsants, or antidepressants due to possible neuropsychological side effects (Handen & Gilchrist, 2006; Stein & Strickland, 1998); and severe visual impairments or hearing loss. Participants had to provide informed consent by signing a form. If there was doubt regarding their capacity to provide informed consent, consent was also required from parents or guardians by signing a form. The characteristics of the final 39 participants with DS are presented in Table 1.

Stimulus materials

Pictogram scales

Four pictograms for pain qualities were developed by Sclera, a Belgian non-profit organisation for graphic design of pictograms to use with people with intellectual disability. The pictograms represented stinging pain, throbbing pain, burning pain, and pressing pain and included symbols for a needle, hammer, campfire, and finger, respectively (see Figure 1). These pictograms were modelled after the Pain Pictures Toolkit (McAuley, 2009), a set of schematic pictures developed for non-English speaking diabetic patients to communicate about neuropathic pain.

Three facial pictograms for pain affect were also developed by Sclera: a smiling face for "no pain," a face with raised inner corners of the eyebrows and a wavy mouth for "moderate pain," and a face with squeezed eyes, a tear, and clenched teeth for "extreme pain" (see Figure 2). These faces were modelled after the Facial Affective Scale (FAS; McGrath et al., 1996; McGrath, De Veber, & Hearn, 1985), which is described in the following section. The smiling face instead of the broad smiling face of the FAS was used as the model for "no pain," because that corresponded to our aim to make facial pictograms as basic as possible.

Figure 2. Facial pictograms. Participants were requested to arrange them from no pain to extreme pain (written descriptions not presented to participants). Developed by Sclera (http://www.sclera.be) in 2012.

Drawn faces for pain affect

The FAS was used because pain affect refers to the perceived unpleasantness (Rainville, Duncan, Price, Carrier,

Figure 3. Faces of Facial Affective Scale (FAS; McGrath, 1996). Participants were requested to arrange them from no pain to extreme pain (written descriptions not presented to participants). From *Pain in Children: Nature, Assessment and Treatment* by Patricia A. McGrath, 1990, New York, NY: Guilford. Copyright 1990. Reprinted with permission.

Table 1. Demographic and medical characteristics of the participants ($N = 39$).

Characteristic	*n*
Age in years	$M = 41.2$ ($SD = 11.8$), range: 20–62
Gender: male	19 (49%)
Living situation: in care centre, or with family	34 (87%), 5 (13%)
Estimated mental age	38 (97%), $M = 5.2$ ($SD = 1.6$), range: 2 years 10 months–9 years 0 months
Receptive language ability (number of words)	39 (100%), $M = 8.3$ ($SD = 1.5$), range: 5–10
Possible indication of dementia	3 (13% of $n = 23 \geq 40$ years)
Thyroid disorder	11 (28%)
Diabetes	1 (3%)
Present analgesics use	4 (10%)
Physical conditions possible pain/ discomfort (incl. changes)	17 (44%)
Characteristics of autism spectrum disorder	1 (3%)
Pictograms in direct environment	29 (74%)
Frequency of using pictograms	25 (86%) daily, 1 (3%) weekly, 3 (10%) less than weekly
Use of pictograms for activities or objects	24 (83% of $n = 29$)
Use of pictograms for activities/ objects and emotions	5 (17% of $n = 29$)
Drawn faces in direct environment	0 (0%)
Pictures or photos for communication in direct environment	24 (62%)

& Bushnell, 1997), which is related to pain tolerance and suffering from pain (Scherder, Sergeant, & Swaab, 2003). The FAS has been used by children (Ellis et al., 2002; Goodenough, van Dongen, Brouwer, Abu-Saad, & Champion, 1999; McGrath et al., 1996; Nilsson, Finnström, Mörelius, & Forsner, 2014; Perrott, Goodenough, & Champion, 2004) and by people with dementia (Scherder & Bouma, 2000). The authors of the original FAS have attributed values to the faces that range from 0.04 (maximum positive affect) to 0.97 (maximum negative affect). To make the comparison between the two scales achievable for the participants, three out of the nine FAS faces were used (see Figure 3). These faces represented no pain (.37), moderate pain (.75), and extreme pain (.97). Because a broad smiling face represents the positive extremity of the original FAS, it has to be mentioned that the smiling face represented "practically no pain" in the original FAS.

Measures

Estimated mental age

The subtests Block Design and Vocabulary of the Wechsler Preschool and Primary Scale of Intelligence – Revised version (WPPSI-R; Wechsler, 1989) were administered, in which the participant has to construct patterns with blocks within a limited time and describe verbally the meaning of words, respectively. The dyad of these two subtests has an excellent reliability ($r = .87$) and a high correlation ($r = .83$) with the Full IQ of the WPPSI-R (Sattler, 2001). Afterwards, the age equivalents in years and months corresponding to the raw scores of both subtests were retrieved from the *Manual Psychodiagnostics and Limited Ability* (Kraijer & Plas, 2006) to calculate the mean age equivalent. A modified version of the Vocabulary WPPSI-R subtest was used. Through forward-backward translation based on guidelines (Ohrbach et al., 2013), we discovered that our Dutch translations of three of the 12 words on the WPPSI-R Vocabulary test were different (possibly easier) from the official translations. Data collection was, however, too far advanced to reach a consensus about the deviating translations. Consequently, the age equivalents for Vocabulary and the estimated total intelligence level might have been slightly too high.

Receptive language comprehension

Receptive vocabulary and syntactic comprehension were screened by the two sample sentences and the first 10 sentences of Sentence Comprehension, a subtest of the Foundation Aphasia Netherlands test (Dutch; Zinsbegrip subtest, Stichting Afasie Nederland test; Deelman, Koning-Haanstra, Liebrand, & Van den Burg, 1981). The participant chose drawings corresponding to sentences that were read aloud by the examiner in a neutral tone. When the examiner noticed that the participant chose randomly, then the instructions were repeated. Possible scores in this study ranged from 0 to 10. It has been suggested that receptive vocabulary and syntactic comprehension are relevant for emotion-recognition tasks in people with DS (Hippolyte et al., 2009). Using a subtest of a test battery to assess the language abilities in people with intellectual disability has been done before by other researchers (Pennington, Moon, Edgin, Stedron, & Nadel, 2003).

Verbal memory

The ability to imprint verbal information was screened by the first condition of the Eight Words Test from the Neuropsychological Test series for Elderly with Mild Intellectual Disability (Verberne, 1998). A list with eight words was read aloud and participants were asked to recall the words, irrespective of the order. This was repeated four times (total score 0–40). It is recommended to assess short-term memory in studies on emotion recognition in people with intellectual disability (Moore, 2001). Imprinting of verbal information was the most relevant in the present study due to the test instruction that was read aloud. Using a subtest of a test battery to assess verbal short-term memory in people with intellectual disability has been done before by other researchers (Pennington et al., 2003).

Possible indications for dementia

In accordance with national recommendations (Evenhuis, Kengen, & Eurlings, 2006; Working Group Network Behavioural Experts of Elderly People in District South, 2005), participants aged 40 years and older were screened for the possible presence of dementia. The scores of the Social Functioning Scale for Intellectual Disability (SRZ/SRZ-P; Kraijer & Kema, 2004; Kraijer, Kema, & de Bildt, 2004) and the Dementia Questionnaire for Intellectual Disability (DMR; Evenhuis et al., 2006) completed during the study were compared to scores of at least six months previously, derived from files of the care centres. The SRZ consists of the subscales Self Help, Communication, Persistence, and Social Skills. An example of an item is "Is able to use cutlery" (no/only spoon/only fork/both fork and knife). The SRZ correlates highly with the Vineland Adaptive Behavior Scales (de Bildt, Kraijer, Sytema, & Minderaa, 2005). The SRZ-P assesses abilities corresponding to a higher level of functioning and consists of the subscales Self Help I, Self Help II, Verbal-Numeric abilities, and Rest items. An example of an item is "Is able to use public transport unguided" (yes/no). The DMR consists of the subscales Short-Term Memory, Long-Term Memory, Orientation, Speech, Practical Skills, Mood, Activity and Interest, and Behavioural Disturbance. An example is "Knows which year it is" (yes/sometimes/no).

When the difference for both the SRZ/SRZ-P and the DMR was statistically significant according to cut-offs in the manuals, then we assumed that the individual possibly had dementia. When no old SRZ/SRZ-P and/or DMR scores were available in the files ($n = 11$), then the caregivers completed both the SRZ/SRZ-P and DMR after the test session and at least six months later to calculate the change in functioning over that time period.

According to guidelines in the manuals (Kraijer & Kema, 2004; Kraijer et al., 2004), the SRZ-P should be used if the SRZ total score is ≥ 100 and the maximal score of at least one subscale is achieved, whereas the SRZ should be used if the SRZ-P total score is ≤ 8. In two participants, the choice for the SRZ instead of the SRZ-P appeared incorrect. Although the use of a questionnaire with lower functioning abilities (SRZ) might have resulted in a ceiling effect (i.e., the functioning is too high to detect statistically significant decrease over time), a dementia indication was in any case not found for these participants, because the difference between measurements was not statistically significant for the DMR.

Medical information

Caregivers of participants used client files to provide the researcher with medical information. Reported medical information (physical conditions, complaints, and medication use) was used to determine a possible presence of discomfort or pain. One physiotherapist, one general physician, and two specialised physicians for people with intellectual disability rated whether the reported physical conditions were expected to cause possible pain or discomfort. A physical condition was ultimately rated as possibly causing pain or discomfort when at least two of the three types of professionals indicated that this could be the case.

Procedure

Study visits

At the first study visit, data were collected about demographic and medical information, and tests were performed for estimated mental age, receptive language ability, verbal memory, and comprehension of three self-reporting scales (i.e., pictograms for pain qualities, facial pictograms, and drawn faces). Five facial pictograms (comprehended by 54% of the participants) and three drawn faces were applied, but data about the comprehension of these scales could not be used. A second study visit was necessary for two reasons: (a) the number of faces needed to be equal to compare comprehension rates, and (b) it was not yet assessed which facial scale was preferred. Therefore, at the second study visit, three faces were used for both facial scales to test comprehension and preference of the scales. Because time elapsed between the two study visits ($M = 3.5$ months, range: .70–6.00 months), changes in the cognitive and/or medical situation were evaluated by asking the caregiver whether such changes occurred since the last visit. A change in cognitive functioning was identified in only one participant (i.e., general decline and disorientation). The medical situation changed in three participants, but resulted in a change from no discomfort to discomfort (i.e., severe dry skin) in only one participant.

Pictograms for pain qualities: Test of comprehension

The pictograms were introduced by asking participants to name the symbols (intended answers: "needle," "hammer," "fire," and "hand"). Participants were considered to comprehend the pictograms if they answered four questions matching with the intended pain qualities. The four questions were (a) "Which one is throbbing pain?", while showing stinging pain and throbbing pain; (b) "Which one is stinging pain?", while showing pressing pain and stinging pain; (c) "Which one is pressing pain?", while showing throbbing pain and pressing pain; and (d) "Which one is burning pain?", while showing burning pain and pressing pain. Repeating the question stimulated participants who did not respond. If a

participant gave an incorrect answer, then the examiner continued with the next question as usual (i.e., the four questions were always administered). The examiner provided no cues or feedback.

Facial scales for pain affect: Test of comprehension

Three facial pictograms were presented in a standardised incorrect order and participants were asked to arrange the faces from no pain to extreme pain (see Figure 2). The examiner asked, "Could you place these faces in the correct order from no pain to a lot of pain?". Repeating the question stimulated participants who did not respond. When participants seemed to have finished, the examiner then asked, "Are you finished?". If participants said "No," then they were allowed to make changes to the placement of the faces. The examiner provided no cues or feedback. The same comprehension test was repeated with three FAS faces (see Figure 3). The intended response for both tests was placing the faces in the order of "no pain" – "moderate pain" – "extreme pain." To account for a possible bias due to the order of presentation, FAS stimuli were presented first in the first 20 participants whereas facial pictograms were presented first in the last 19 participants.

Facial scales for pain affect: Preference

After the comprehension tests, participants were presented with one row of FAS stimuli and one row of facial pictogram. Then they were asked which row of faces they liked best. The participant always responded by placing a hand before the preferred row or by pointing to all of the faces of the preferred row. The reason for the preferences was not assessed.

Communication tools in daily life

Caregivers were asked whether the participants used pictograms and drawn faces for communication in their daily life and how often. It was also asked whether pictures and photos were used for communication.

Statistical analysis

Statistical analyses were performed using SPSS Version 21. The level of statistical significance was set at $\alpha = .05$ (two-sided). The effect size calculator of Wilson (2001) was used to calculate Cohen's d and its 95% confidence interval for the independent-samples t tests and the odds ratio (OR), and its 95% confidence interval for the binomial tests, the chi-square test, Fisher's exact tests, and the McNemar test. Cohen's d was interpreted as follows: "insignificant" up to 0.20, "small" ≥ 0.20 to < 0.50, "medium" ≥ 0.50 to 0.80, and "large" ≥ 0.80 (Dunst & Hamby, 2012). OR was interpreted as follows: "insignificant" up to 1.68, "small" ≥ 1.68 to 3.47, "medium" ≥ 3.47 to 6.71, and "large" ≥ 6.71 (Chen, Cohen, & Chen, 2010). Table 2 describes the analyses (all inferential) and variables per research question.

Results

Pictograms for pain qualities

Of the participants, 51% ($n = 20$) chose pain qualities matching the intended answers for all four pictograms, 18% ($n = 7$) for three pictograms, 23% ($n = 9$) for two pictograms, 3% ($n = 1$) for one pictogram, and 5% ($n = 2$) did not succeed. A statistically significant difference was found between 51% and a test proportion of .06 (binomial test, $p < .001$, $OR = 16.31$ [3.78, 70.43]), which was based on the chance of all four responses matching with the intended answers.

The percentages of a matching response per pictogram were 74% for stinging pain, 80% for throbbing pain, 92% for burning pain, and 62% for pressing pain. Binomial tests showed that the difference between these percentages and a test proportion of .50 was statistically significant for three pain qualities (stinging pain, $p = .003$, $OR = 2.85$ [1.10, 7.37]; throbbing pain, $p < .001$, $OR = 4.00$ [1.47, 10.93]; burning pain, $p < .001$, $OR = 11.50$, [3.08, 42.89]), but not for pressing pain ($p = .20$, $OR = 1.63$ [0.66, 4.02]).

Compared to participants who did not recognise all intended pain qualities, participants who recognised all intended pain qualities performed better on tests for mental age, $t(36) = -4.57$, $p < .001$, $M = 6.10$ versus $M = 4.22$, $d = -1.48$ [−2.20, −0.77], receptive language ability, $t(37) = -4.17$, $p < .001$, $M = 9.10$ versus $M = 7.42$, $d = -1.34$ [−2.03, −0.64], and verbal memory, $t(34) = -2.18$, $p = .036$, $M = 20.79$ versus $M = 15.06$, $d = -0.73$ [−1.40, −0.05]. The association between recognition of all intended pain qualities and familiarity with pictograms was not statistically significant (Fisher's exact test, $p = .27$, $OR = 0.35$ [0.08, 1.62]).

Facial pictograms and drawn faces for pain affect

The difference between the comprehension rates for facial pictograms (56%, $n = 22$) and FAS stimuli (54%, $n = 21$) was not statistically significant (McNemar test, $p = 1.00$, $Phi = .85$, $OR = 160$ [13.28, 1,927.58]). A statistically significant difference was found between both percentages and a test proportion of .17 (binomial tests: FAS stimuli, $p < .001$, $OR = 6.21$ [2.18, 17.72]; facial pictograms, $p < .001$ [2.01, 16.32]), which was based on the chance of a response matched with the intended order. Of the participants, 51% ($n = 20$) comprehended both

Table 2. Statistical analyses and variables per research question.

Research question	Variables	Statistical analysis
1.1 Are the intended pain qualities in a set of pictograms recognised above chance level?	Comprehension test score of pain-qualities pictograms	Binomial test, with .06 chance on the four intended answers (.50 × .50 × .50 × .50)
1.2 Is cognitive functioning different between participants who comprehend this set and those who do not?	Comprehension test score of pain-qualities pictograms, mental age, receptive language ability, verbal memory	Independent-samples *t* tests
1.3 Is comprehension of this set associated with familiarity with pictograms?	Comprehension test score of pain-qualities pictograms, familiarity with pictograms	Chi-square test
2.1 Are facial pictograms better comprehended than drawn faces?	Comprehension test scores of facial pictograms and drawn faces	McNemar test Extra: binomial tests, with .17 chance on the intended order of three faces (.33 × .50 × 1)
2.2 Is cognitive functioning different between participants who comprehend these facial scales and those who do not?	Comprehension test scores of facial pictograms and drawn faces, mental age, receptive language ability, verbal memory	Independent-samples *t* tests
2.3 Is comprehension of the facial pictograms associated with familiarity with pictograms?	Comprehension test scores of facial pictograms and drawn faces, familiarity with pictograms	Chi-square tests
3.1 Are facial pictograms preferred over drawn faces?	Preference	Chi-square test
3.2 Is preference for facial pictograms associated with familiarity with pictograms?	Preference, familiarity with pictograms	Chi-square test

scales, 41% ($n = 16$) comprehended none of the scales, 5% ($n = 2$) comprehended only facial pictograms, and 3% ($n = 1$) comprehended only FAS stimuli. The two facial scales were comprehended by the participant with both DS and "cognitive deterioration" and by the three participants with both DS and a screening score of possible dementia.

Compared to participants who did not comprehend facial pictograms, participants who comprehended facial pictograms performed better on tests for mental age, $t(36) = -3.20$, $p = .003$, $M = 5.78$ versus $M = 4.30$, $d = -1.04$ [−1.72, −0.36], receptive language ability, $t(37) = -4.01$, $p < .001$, $M = 9.00$ versus $M = 7.35$, $d = -1.28$ [−1.97, −0.60], and verbal memory, $t(34) = -2.49$, $p = .018$, $M = 20.64$ versus $M = 14.07$, $d = -0.83$ [−1.51, −0.15]. Similarly, participants who comprehended drawn faces performed better on tests for mental age, $t(36) = -2.92$, $p = .006$, $M = 5.77$ versus $M = 4.41$, $d = -0.95$ [−1.62, −0.28], receptive language ability, $t(37) = -3.73$, $p < .001$, $M = 9.00$ versus $M = 7.44$, $d = -1.20$ [−1.88, −0.51], and verbal memory, $t(34) = -3.30$, $p = .002$, $M = 21.48$ versus $M = 13.33$, $d = -1.10$ [−1.80, −0.40], than those who did not comprehend drawn faces. The associations between comprehending facial pictograms and familiarity with pictograms was not statistically significant (Fisher's exact test, $p = .46$, $OR = 0.46$ [0.10, 2.13]). The same applied to drawn faces (Fisher's exact test, $p = .29$, $OR = 0.40$ [0.09, 1.86]).

Preference of facial pictograms or drawn faces

More participants preferred facial pictograms (56%, $n = 22$) over FAS stimuli (44%, $n = 17$), but the association between preference and facial scale was not statistically significant, $X^2(1) = 0.64$, $p = .42$, $OR = 1.68$ [0.68, 4.10]. The association between preference and familiarity with pictograms was not statistically significant (Fisher's exact test, $p = .28$, $OR = 2.46$ [0.56, 10.68]).

Discussion

The main findings of the present pilot study were (a) adults with DS recognised pain qualities above chance level in a set of pictograms and pain affect levels in facial pictograms; (b) the comprehension of facial pictograms and drawn faces was comparable in adults with DS; (c) comprehension of quality-of-pain pictograms, facial pictograms, and drawn faces was related to cognitive functioning but not to familiarity with pictograms; and (d) a subgroup of adults with DS preferred facial pictograms, although this subgroup was not larger than those preferring drawn faces and preference was not related to familiarity with pictograms.

The percentages around 50% for all three self-reporting scales indicate that many adults with DS could not comprehend the scales. Possible explanations for this finding are a difficulty to recognise negative emotions in facial expressions (Kasari et al., 2001; Porter et al., 2007), the appeal to receptive language ability to recognise semantic complex expressions (Hippolyte et al., 2008), such as the qualities of pain, impairment in verbal short-term memory (Næss et al., 2011), and the mainly instrumental instead of expressive use of pictograms by the participants in daily life (i.e., objects and activities instead of emotions).

The question arises whether pictograms and drawn faces are suitable for people with DS to self-report pain. The results suggest that all these scales may be unsuitable for adults with DS with a lower cognitive functioning. It has been found before that verbally labelling emotions in facial expressions was positively related to mental age in children with DS, although emotion

recognition was not (Kasari et al., 2001). Research on the ability of people with DS to recognise pain and pain intensity levels in facial expressions is needed. The symbols on the belly in the quality-of-pain pictograms (see Figure 1) could make it difficult to comprehend that those pictograms also represent other pain locations besides abdomen pain. The Iconic Pain Assessment Tool may be an alternative, because symbols, such as an ice cube, are placed on a body map (Lalloo & Henry, 2011). Quality of pain is important to discriminate nociceptive from neuropathic pain (Lin, Kupper, Gammaitoni, Galer, & Jensen, 2011). Still, it remains an abstract concept requiring introspective ability and may be too difficult for people with intellectual disability.

The additional value of pictograms as self-reporting pain scale is the finding in the present study and other studies (Fujisawa et al., 2011; Kåhlin & Haglund, 2009; Renblad, 2000) that many people with intellectual disability already use pictograms in daily life for communication. However, only five participants used pictograms in daily life to express emotions. Adults with DS may be unfamiliar with recognising different levels of a certain emotion in facial pictograms and with the fact that pictograms could communicate more abstract feelings, such as pain qualities. Practice may improve comprehension of the pictogram scales used in the present study, which will increase the value of these scales for pain assessment. Adults with DS who have a higher cognitive functioning may benefit most from practice.

Limitations

To interpret how atypical the performance of the group with DS is, it would have been useful to include control groups matched on mental age and receptive language ability, as is often done in research on emotion recognition in DS (e.g., Hippolyte et al., 2008; Kasari et al., 2001). These groups could then also have performed a task for recognising emotions in facial expressions to evaluate how well pain is discriminated from other emotions (e.g., sad or angry). Further, exposure to the facial pain scales should have taken place during one study visit, as the two study visits may have influenced the results.

Recommendations for future research

For a better interpretation of the current results, recognition of pain in facial expressions as well as comprehension of pictogram scales for self-reporting pain should be examined in large samples of adults with DS, adults with other aetiologies of intellectual disability, and controls matched on mental age. The relationship between cognitive functioning and comprehension should be studied for various types of self-reporting scales (e.g., numeric rating scale and coloured analogue scale) to know whether the finding for the pictogram scales and drawn faces is atypical. More information is needed about whether number, size, colour, and arrangement of items influence comprehension of a self-reporting pain scale by people with intellectual disability. Further, it is unclear how many items from a facial pain scale can be used in an ordering task to assess comprehension without losing too much information about the interpretation of the scale. This is especially a problem for scales such as the original FAS (including nine faces), because ordering a large number of faces may be too difficult for people with intellectual disability.

Conclusion

The findings of the present pilot study suggest that adults with DS can recognise a set of pictograms for pain quality above chance level, are as able to comprehend facial pictograms as drawn faces for pain affect, and do not prefer one of these facial scales over the other. For adults with DS with a high mental age, a good receptive language ability, and an unimpaired verbal memory, the use of pictogram scales to self-report pain could be practised. This would be valuable, because pictograms are already often used for communication in their daily life. Research on emotion recognition and pain should be combined to aid pain assessment in DS.

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