This chapter is under revision as:

ABSTRACT
The use of the Self-reporting Tool On Pain in people with Intellectual Disabilities (STOP-ID!), an online application developed by the authors to aid an autonomous self-reporting of pain, was evaluated in 40 adults with Down syndrome. Comprehension of the use of the tool (the ability to recognize representations for vocabulary and pain, and to navigate the tool interface), and use of the tool to self-report pain experience were investigated. Both a laptop and a tablet computer were used (crossover design). The results suggest that more participants recognized representations of pain location and pain affect than representations of pain intensity and pain quality. A small percentage of participants demonstrated comprehension of the tool without assistance (18% laptop, 18% tablet). Half of the participants reported without support information about at least one pain component of a current or remembered pain experience (50% laptop, 53% tablet). Ways to modify the design of the tool and improve performance are suggested.

Keywords: Down syndrome, pain assessment, augmentative and alternative communication, computer applications, adults.
Due to the subjective nature of pain [2], self-reporting is often considered to be the preferred method of pain assessment [16]. Intensity, affect, quality, and location are considered key aspects of pain to include into a self-report assessment [19]. Pain intensity is the term used to describe the perceived somatosensory severity of pain [25]. Pain affect refers to the perceived unpleasantness [34], which is related to pain tolerance and suffering from pain [38]. Pain quality refers to how pain feels in subjective terms of somatosensory sensations, such as burning or stinging [18]. Pain location is the term used to describe the ability to perceive the location of tissue injury, which is called for to report ‘where it hurts’ [40].

The ability to understand such terms and to evaluate one's own pain may require a certain level of cognitive functioning, including an understanding of associated vocabulary terms. People with intellectual disabilities could have difficulties in communication about health-related information, both in understanding what is being asked and in having a way to respond [31]. Factors such as fear of others’ reactions and not wanting to waste others’ time can result in hiding pain instead of self-reporting pain [3,9]. In addition, the display of atypical pain behaviours such as aggression and agitation could hamper caregivers in recognizing pain and asking about it [3,5,23].

The challenges in accurately assessing pain in adults with intellectual disabilities could be related to the indications for under-treatment of pain [2,6,33]. This is particularly alarming: like all individuals, they are in need of prompt medical attention in the event of illnesses and accidents. They also are at increased risk of suffering from pain due to painful physical conditions, such as gastroesophageal reflux disease [7] and musculoskeletal disorders (e.g., arthritis and spasticity) [24]. Because pain could negatively influence quality of life [41], it is crucial that reliable and valid methods are available to help people with intellectual disabilities report their pain experience. Further, techniques are needed that will allow people with intellectual disabilities to regularly report their pain experience, so the effectiveness of interventions can be assessed over time, and changes implemented (as needed) to obtain desired outcomes, and to prevent pain from becoming chronic. Ideally, these tools would support the independent reporting of pain so that individuals with disabilities would not be dependent
upon the presence of a trained caregiver. This is a screening of pain: further pain assessment should be performed by medical professionals. Such tools for self-reporting of pain also may provide individuals with intellectual disabilities a sense of self-determination, and a feeling of greater control over their lived experience. Following this line of reasoning, we argue that the development of techniques to support the self-reporting of pain might support the independent functioning of this population group.

Currently, the regular collection of information on pain from individuals with intellectual disabilities poses significant challenges for caregivers. Various behaviours are used by people with intellectual disabilities to express pain: verbal indicators, such as reporting the pain location, and non-verbal indicators, such as using sign language for “hurt”, pointing to or showing the injury, touching the hurting body part, changes in facial activity, crying or moaning, and withdrawn or aggressive behaviour [23]. It has been stated that the majority of people with intellectual disabilities may be able to report pain by using scales corresponding to their developmental level [17]. People with Down syndrome may have difficulty with localising pain [15], but this ability may improve by using a picture of a human body [4]. It has been found that 71% of adults with Down syndrome comprehended at least one of two scales for pain affect and pain intensity [22]. Adults with intellectual disabilities were unable to rate a statistically significant increase in pain on a coloured analogue scale during an injection compared to baseline [29]. As far as we know, no studies have been performed that address the ability of these individuals to report pain quality.

More insight is needed into the abilities of people with intellectual disabilities to report different aspects of pain. It is unclear how advanced the knowledge about self-reporting pain scales is in caregivers and medical professionals regarding the advantages and disadvantages of each scale and the standardized instructions for applying the scales. For example, suggestive or grammatically complex questions should be avoided [10] and at least comprehension of the scale extremes should be tested before the scale is applied to assess pain experience [22]. In conclusion, there is a need for a method to administer a set of various scales for self-reporting pain in people with intellectual disabilities, in which comprehension of the scale items is assessed and standardized instructions are used.
In developing a tool to support the self-reporting of pain, there are a number of important considerations. First, the tool must adequately address the needed content regarding the intensity, affect, quality, and location of their pain. Second, these terms (and response options) must be represented in a way that is quickly understandable and usable by the person with intellectual disability. For example, for individuals who are unable to read, instructions should be read aloud and/or represented with appropriate images [44]. Black-and-white pictograms, already often used for communication by people with intellectual disabilities [11,21,35], could be beneficial for the visual processing of key elements, and adding an increasing red colour to a numeric rating scale might enhance understanding. Third, there must be a reliable access method for the individual to select the needed vocabulary and concepts, so that the assessment can be carried out in an efficient manner both for the person with intellectual impairment, and can be repeated over time, as necessary. It would be important that it could be used with minimal training for staff, due to the high level of staff turnover in the field [13]. Finally, it would be important that the collection of information be efficient with respect to time for the caregiver. Professional caregivers of individuals with intellectual disabilities often have a heavy workload [14], with 25-33% of them experiencing high levels of stress [36]. The development of an efficient tool would increase the likelihood that it would be used on a regular basis, and thereby serve to support the effective monitoring and reporting of pain conditions.

In developing a tool for collecting information on pain, there may be benefits to the development of a computer-based approach, thereby providing an easily adapted user interface, and electronic collection of standardized information. In recent decades, applications for touch screens have been developed to support the communication of people with intellectual disabilities (for a review, see: [20]). Many individuals with intellectual disabilities can understand computer applications specially designed for them and can use touch screens. Although the use of a computer mouse requires larger cognitive and motor skills than the use of a touch screen [43], other research results show that a subgroup of people with intellectual disabilities is able to use a computer mouse for double clicking and dragging [30]. This suggests that traditional laptop computers may also be a viable approach, especially for people with mild intellectual disabilities [30].
developing a computer application for self-reporting pain, it is thus important to
determine which computer device would be most suitable to use for people with
intellectual disabilities, as this is a heterogeneous population group.

As part of a larger research project on pain experience, pain assessment, and
cognitive functioning in adults with Down syndrome, an online application was
developed to determine whether adults with intellectual disabilities (specifically
people with Down syndrome) can use a computer device that would enable them
to report information about their pain. The online application, called “STOP-ID!”
(Self-reporting Tool On Pain in people with Intellectual Disabilities) was designed
to support the communication of pain information by people with intellectual
disabilities. It features the use of graphic images and pictograms to represent key
issues of pain location, intensity, affect, and quality (see Figures 1-4 respectively),
and can be used on either a laptop or a tablet device.

FIG. 1
Graphic image showing the front and back of a human
body. Used to assess pain location. The image was used in
the comprehension test and, if 10 out of 10 locations were
correctly identified, also in supporting the participant’s self-
report of the pain location.
FIG. 2
Pictograms and graphic images used to assess pain intensity. To test comprehension, participants had to correctly indicate the larger of two numbers that were presented as pictograms on the screen (two questions of this type were asked: see Figure 2a and Figure 2b) and to correctly indicate on a 0-10 scale (see Figure 2c) which number would be used to represent no pain and which number would be used to represent a lot of pain. If 4 out of 4 answers were correct, then a 0-10 scale with an increasing red colour was used to self-report pain intensity (see Figure 2d).
FIG. 3
Pictograms used to assess pain affect. To test comprehension, participants had to correctly indicate which of three facial pictograms would be used to represent no pain (the middle face: pain affect level 0), a lot of pain (the left face: pain affect level 2), and a little bit of pain (the right face: pain affect level 1). If 3 out of 3 answers were correct, then participants were asked which of these pictograms represented their pain experience.

FIG. 4
Pictograms used to assess pain quality. To test comprehension, participants had to correctly indicate which of four pictograms represented stinging pain (the first pictogram), pressing pain (the second pictogram), burning pain (the third pictogram), and throbbing pain (the fourth pictogram). If 4 out of 4 answers were correct, then participants were asked which of these pictograms represented their pain experience.
The purpose of the present usability study was to provide a preliminary investigation of the use of STOP-ID! with the targeted group. The following questions were addressed: (1) Can adults with Down syndrome and mild to severe intellectual disabilities demonstrate comprehension (i.e., recognize the images used and navigate the interface) of the online tool? (2) If the answer to this question is yes, what kinds of information about a current or remembered pain experience are they able to report? (3) Do they require assistance and if so, what kinds of support are needed? (4) Do they prefer one computer device (e.g., laptop or tablet) over another?

MATERIAL AND METHODS

Study design and procedure

The usability study was conducted in 40 adults with Down syndrome on a laptop and a tablet. A crossover design was used to control for a possible order effect: the laptop preceded the tablet in 20 participants, while the tablet preceded the laptop in the other 20 participants.

The current study consisted of a single test session, in which the use of STOP-ID! was investigated with the participants. All tests were performed in a quiet room of the facility where the participants lived or worked. During this session, caregivers were asked about the experiences of the person with intellectual disabilities with laptops, computers, tablets, and pictograms, and about the participant’s ability to read. Demographic, medical, and language-related information about the participants that was collected earlier in an ongoing study about pain experience in adults with Down syndrome was used for the current study to avoid additional burden of caregivers and participants.

The average number of months between the initial collection of demographic, medical, and language-related information and the STOP-ID! testing was 14.6 months ($SD = 3.2$, range: 9 - 19), because time was needed to find financial support, to develop the STOP-ID!, and to obtain informed consent of the participants. Caregivers were asked whether changes in the medical and/or cognitive functioning of the participants had occurred in the previous year. Caregivers suspected a decline in cognitive functioning for nine participants.
and the development of a possible painful condition in one participant (i.e., sore throat due to reflux of gastric acid).

**Ethical approval**
The Medical Ethical Committee of the university to which the first author is related approved the study and the informed consent procedure.

**Participants and characteristics of the sample**
Forty individuals participated in the study. All belonged to a care organization for people with intellectual disabilities in a large city in the Netherlands. The following inclusion criteria were used: (a) 18 years of age or older, (b) ability to speak and understand Dutch, (c) a demonstrated willingness to participate in testing activities, (d) a diagnosis of Down syndrome. Exclusion criteria were neurological disorders such as cerebrovascular accidents or tumors, the use of antipsychotics, anticonvulsants, or antidepressants due to possible neuropsychological side effects [12,39], and severe visual impairments or hearing loss. The latter exclusion criterion was based on the estimation of the caregiver whether the participant would be able to see pictures clearly and to hear clearly what is being said. Severity of intellectual disability was not considered to be an exclusionary criteria – adults with Down syndrome of all levels of intellectual disability could participate as long as caregivers had reported that these adults had the motor ability to press on a touch screen and to use a computer mouse (i.e., move the mouse and press the button), and the cognitive ability to follow simple instructions (e.g., “Please sit down in front of this computer.”). Both individuals who were and who were not currently known to be experiencing pain were included in the study. The presence of painful or discomforting physical conditions according to medical information was not an exclusion criterion, because this is precisely the type of information the tool is meant to collect.

To be included in the study, participants had to provide informed consent. If there was doubt regarding their capacity to provide informed consent, informed consent was also required from parents or guardians. In total, 40 participants were included, with an average age of 43.3 years ($SD = 11.7$, range 20-66 years) and of whom 40% was male. The median age equivalent of vocabulary abilities was 4;0 years ($IQR = 2;0$, range 2;1 – 10;1 years). Only three participants used
pain medication (acetaminophen and/or Diclofenac) and one participant had a possible indication of dementia. Most participants (68%) were unfamiliar with a laptop and tablet. Table 1 provides information about other characteristics of the sample. Of the participants, 23 (58%) were able to verbally report pain. Non-verbal pain behaviours were physical changes (including faces and posture), emotional changes (including crying), moaning, and pointing to the painful location. Table 2 provides more information about the pain behaviours of the participants as described by caregivers.

**Table 1**
Characteristics concerning demographic and medical variables, reading, use of pictograms, and use of computer-related devices (N = 40)

<table>
<thead>
<tr>
<th>Characteristics</th>
<th>n (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Level intellectual disability</strong></td>
<td></td>
</tr>
<tr>
<td>mild</td>
<td>17 (43%)</td>
</tr>
<tr>
<td>moderate</td>
<td>20 (50%)</td>
</tr>
<tr>
<td>severe</td>
<td>3 (8%)</td>
</tr>
<tr>
<td><strong>Living situation</strong></td>
<td></td>
</tr>
<tr>
<td>in care centre</td>
<td>36 (90%)</td>
</tr>
<tr>
<td>at home with family</td>
<td>4 (10%)</td>
</tr>
<tr>
<td><strong>Physical conditions that may cause pain/discomfort</strong></td>
<td>22 (55%)</td>
</tr>
<tr>
<td><strong>Ability to read according to caregivers</strong></td>
<td>23 (58%)</td>
</tr>
<tr>
<td><strong>Use of pictograms:</strong></td>
<td></td>
</tr>
<tr>
<td>in total</td>
<td>24 (60%)</td>
</tr>
<tr>
<td>daily</td>
<td>16 (40%)</td>
</tr>
<tr>
<td><strong>Familiar with at least one computer-related device</strong></td>
<td>20 (50%)</td>
</tr>
<tr>
<td><strong>Use of at least one computer-related device</strong></td>
<td>14 (35%)</td>
</tr>
</tbody>
</table>

*Note.* Computer-related device: laptop, tablet, regular computer, and smartphone.
Materials and methods for background characteristics of the sample

Information on the intellectual disability level, possible indication of dementia, vocabulary knowledge, and medical information of the participants was available from a larger, ongoing study in which the individuals had participated. Information on intellectual disability level was obtained from the Social Functioning Scale for Intellectual Disability (i.e., SRZ or SR-P; [26,27]). The SRZ and SRZ-P assess social and cognitive abilities and activities of daily living, in which the SRZ-P correspond to a higher level of functioning. The caregiver choose which questionnaire was more appropriate for the participant’s level of functioning. By using the population norms of the manual, the SRZ total score was converted into a standardized score, which was then converted into an estimated level of intellectual disability by using the “Manual of Psychodiagnostics and Limited

Note. $n =$ the number of participants displaying the pain behaviour. Because some participants displayed several pain behaviours and caregivers were asked to report all pain behaviours of a participant, the number of participants exceeds 40.

Table 2

Pain behaviour of participants as reported by caregivers

<table>
<thead>
<tr>
<th>Pain behaviour</th>
<th>$n$ (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Expressing pain never or rarely</td>
<td>5 (8%)</td>
</tr>
<tr>
<td>Expressing pain in an exaggerated way (perseverates or carries on too far)</td>
<td>3 (5%)</td>
</tr>
<tr>
<td>Expressing verbally</td>
<td>23 (37%)</td>
</tr>
<tr>
<td>Expressing vocally (moaning)</td>
<td>1 (2%)</td>
</tr>
<tr>
<td>Expressing physically (facial, posture, eating, sleeping, physiological reactions)</td>
<td>11 (18%)</td>
</tr>
<tr>
<td>Pointing to painful location</td>
<td>3 (5%)</td>
</tr>
<tr>
<td>Emotional changes (crying, mood, or behaviour such as withdrawal)</td>
<td>7 (11%)</td>
</tr>
<tr>
<td>No pain behaviour, while pain is suspected by caregiver</td>
<td>2 (3%)</td>
</tr>
<tr>
<td>Expressing pain, but unclear whether pain is present</td>
<td>1 (2%)</td>
</tr>
<tr>
<td>High pain threshold</td>
<td>6 (9%)</td>
</tr>
</tbody>
</table>

The scores for the intellectual level for the participants should be interpreted with caution as there is evidence that for at least two of the participants, the use of the SRZ-P instead of the SRZ appeared incorrect according to guidelines in the manuals.
Ability” [28]. To use only one variable of estimated intellectual disability level, the intellectual disability levels for all participants were based on the SRZ. Participants of whom only the SRZ-P was available were identified as having a mild level of intellectual disability according to the SRZ. To screen participants aged 40 years and older for a possible indication of dementia, scores of the SRZ or SRZ-P and the Dementia Questionnaire for Intellectual Disability (DMR; [8]) were collected for two moments in time (i.e., during the current study and old scores form the files) with at least six months between them to assess deterioration over time. A possible indication of dementia was considered to be present if the decrease in the total scores of the questionnaires was statistically significant according to criteria in the manuals.

Vocabulary level was estimated by using a modified version of the Vocabulary subtest of the Wechsler Preschool and Primary Scale of Intelligence – Revised (WPPSI-R) [42]. Participants were asked to provide a verbal description of the meaning of words (e.g., “Knife” and “Umbrella”), with the greatest number of points given for correct abstract descriptions according to the WPPSI-R manual (e.g., “A knife is a weapon” and “An umbrella keeps the rain off you”). Afterwards, the raw score was converted into an age equivalent2 in years and months using the “Manual of Psychodiagnos tics and Limited Ability” [28]. Medical information about the use of medication and the presence of physical conditions that may cause pain was obtained from a review of records by caregivers.

As part of the present STOP-ID! study, a caregiver was asked about the participant’s ability to read, experience with pictograms in daily life, and experience with computer devices. When the caregiver of the living facility reported that the participant used a computer in the facility for work or activities, then the researcher contacted this facility. When the caregiver was not aware of the participant’s experience with pictograms or digital devices, then the caregivers asked the participant open questions in the presence of the researcher.

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2 The scores for language level should be interpreted with caution as they are based on adaptations made by the current research team in creating a modified Dutch version of the WPPSI-R Vocabulary test, which may have resulted in a slight over-estimation of their language ability (as reported using age-equivalent scores).
Self-reporting Tool On Pain in people with Intellectual Disabilities (STOP-ID!)

The concept of the online application STOP-ID! was designed by the authors and the STOP-ID! itself was developed by Stichting OOKJIJ (roughly translated as YOU TOO Foundation), a Dutch organization that has developed a website for people with intellectual disabilities to support safe use of the Internet. Participants could log in with their personal account consisting of a numeric code. After a successful log-in, the participant would see a photo of themselves and hear their name spoken aloud by the device.

For the laptop condition, a Latitude E5530 laptop™ was used that included Google Chrome™, a mouse with two buttons and a scroll wheel, and a mouse mat. For the tablet condition, an iPad™ 2 was used that included Google Chrome, a SIM card for 3G mobile internet™, and a Smart Case™ to be folded as a stand. Both devices had internet capability. Each participant had an opportunity to complete the STOP-ID! protocol twice during the same test session: once on the laptop and once on the tablet. The STOP-ID! test itself was identical on the two devices, but the devices differed from each other in screen size (i.e., 15.6 inch of the laptop versus 9.7 inch of the tablet) and response mode (i.e., computer mouse versus touch screen)

The STOP-ID! test consists of a number of pages (featuring graphics and/or text) that are presented to the participant on the device (laptop or tablet). All written text on a page of the test was read aloud by the computer.

In completing the test, the individual is first asked if they are experiencing pain on that day. In typical use, STOP-ID! automatically directs the participants without pain at the time of the assessment (i.e., individuals who are not presently experiencing pain) to the end of the test. Because of a concern that this would not have given the researchers enough opportunity to observe the use of the STOP-ID!, the procedure was modified in the current usability study, so that it could be used with all participants (regardless of their current pain status). First, patients were asked if they were currently experiencing pain – those who were continued.

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3 A Latitude E5530 laptop if a product of Dell Inc, Round Rock, TX.
4 Google Chrome is a product of Google Inc, Mountain View, CA.
5 An iPad 2 and a Smart Case are products of Apple Computers Inc, Cupertino, CA.
Participants without current pain were asked to think about pain that they might have experienced recently - they were asked to respond to questions on that basis.

As the participants answered questions, the system was navigated in one of two ways. Either the participant was automatically directed to the next page after clicking or touching an answer, or in some cases, an arrow as symbol for CONTINUE.

This investigation of STOP-ID! was focused on whether it could be used as a method of supporting the self-reporting of pain by people with intellectual disability. The testing procedures first centered therefore, on whether participants could demonstrate comprehension of the use of the tool. This was assessed by their performance on four comprehension tests, that required recognition of the representations for important features of pain (i.e., location, intensity, affect, and quality) and an ability to navigate to the correct answer by using the touch screen and computer mouse. Those participants who demonstrated passing (i.e., perfect) scores on one or more of the four comprehension tests were then asked to use the tool to report a current or recent pain experience. Support (e.g., repeating the question) was only provided if participants were unable to continue independently and was recorded by the researcher. Answers to questions in the STOP-ID! were not provided by the researcher. Additional details are provided below. All instructions and questions were provided by written text on the screen as well as recorded voice output of the computer device.

**Comprehension tests**

**Pain location**

To demonstrate recognition of the representation of pain location in STOP-ID!, participants were shown the front and back of a human body. They were then asked to locate the head, chest, belly, arms, and legs on this picture of the front side of a human body and the head, neck, back, arms, and legs on a picture of the back side of a human body (see Figure 1). A score of 10 out of 10 was required to earn a passing score.
**Pain intensity**

To demonstrate recognition of the representation of pain intensity in STOP-ID!, participants had to correctly indicate the larger of two numbers that were presented as pictograms on the screen (e.g., when asked, “Which is larger, 2 or 8?”, a correct response is to indicate the number 8). There were two questions of this type. They were also verbally asked to correctly indicate on a 0-10 numeric scale which number would be used to represent no pain (0 or 1) and which number would be used to represent a lot of pain (9 or 10). A total of four questions were used for this concept (see Figure 2). A score of 4 out of 4 was required to earn a passing score.

**Pain affect**

To demonstrate recognition of the representation of pain affect in STOP-ID!, participants had to correctly indicate which of three facial pictograms would be used to represent no pain (the middle face: pain affect level 0), a lot of pain (the left face: pain affect level 2), and a little bit of pain (the right face: pain affect level 1). A total of three questions were used for this concept (see Figure 3). A score of 3 out of 3 was required to earn a passing score.

**Pain quality**

To demonstrate recognition of the representation of pain quality in STOP-ID!, participants had to correctly indicate which of four pictograms represented stinging pain (the first pictogram), pressing pain (the second pictogram), burning pain (the third pictogram), and throbbing pain (the fourth pictogram). A score of 4 out of 4 was required to earn a passing score (see Figure 4).

**Report of a current or recent pain experience**

Those individuals who obtained perfect scores on one or more of the previous four tests of recognition and navigation also had an opportunity to provide information on a current or recent pain experience. The following instructions were provided by written text and voice output of the computer: “Now we want to know everything about your pain.”, “Where does it hurt?” (showing the image of the human body to assess pain location, see Figure 1), “Which number fits your pain?” (showing the 0-10 scale with an increasing red colour to assess pain intensity, see Figure 2), “Which face fits your pain?” (showing the facial pictograms
to assess pain affect, see Figure 3), and “Which picture fits your pain?” (showing the pictograms to assess pain quality, see Figure 4).

Information on Pain quality was not gathered, because pain location, affect, and intensity were deemed more clinically relevant as information for health care workers to determine whether further pain assessment and medical attention is necessary. The individual pain score that emerged in the database as a summary of the responses by the participant was for example ‘Back-side arms, 2/2, 9/10’, corresponding to pain location, pain affect, and pain intensity, respectively.

A key difference between the collecting of information on comprehension (i.e., recognition and navigation), and the reporting of a current or recent pain experience, was that it was difficult to determine the reliability of the responses. For recognition and navigation questions, the correct response is known to the researcher. For the self-reporting of pain, the correct response is only known to the participant, not the researchers.

Performance evaluation
A scoring form was developed and used by the first author to write down qualitative observations and to evaluate the participant's performance during the laptop version and the tablet conditions (see Supplementary Information 1). This form contains 7 statements in a yes/no format (e.g., presence of distraction or impulsivity), the actual time in minutes required to complete the test (i.e., without the wasted time during technical problems that was not caused by the participant), and the number of times that the instructions for the different steps needed to be repeated. After participants performed both versions of the STOP-ID!, they were asked which of the two tasks they liked best.

Statistical analysis
Statistical analyses were performed using Statistical Package for the Social Sciences version 21 (IBM SPSS Statistics 21). The categories of moderate and severe intellectual disability were combined for the analyses due to the small number of participants with severe intellectual disability, resulting in the two categories ‘mild’ (n = 17) and ‘moderate to severe’ (n = 23). The research questions were answered by using descriptive statistics, McNemar tests, independent-sample t-tests, a Wilcoxon signed-ranks test, and Chi-squared tests. For this questions for
which it was appropriate, the level of significance was set at $\alpha = .05$ with rejection of the null-hypothesis when two-sided $p < .05$.

RESULTS

Can adults with Down syndrome use an online application designed to report information about pain?

All participants finished the STOP-ID! in both the laptop and tablet conditions. The average performance time was 16.0 minutes ($SD = 8.1$) on the laptop and 14.6 minutes ($SD = 3.9$) on the tablet. This was timed from the starting screen up to the closing screen, including the comprehension tests and self-reporting of pain. Most participants were able to insert the account code in the laptop condition (69%) and tablet condition (85%) and all participants for whom a photo was available in the account ($n = 39$) recognized it in both conditions. It was observed by the researcher that during the study visit, some of the participants seemed at times distracted or bored (25% in laptop condition, 48% in tablet condition), and some of the participants appeared to answer at least a few questions impulsively (30% in laptop condition, 35% in tablet condition).

Recognition and navigation performance

The results of the comprehension tests are presented in Table 3. The average number of comprehension tests with a perfect score was 1.5 ($SD = 1.2$) in the laptop condition and 0.9 ($SD = 0.8$) in the tablet condition. In the laptop condition, most participants were successful in answering questions concerning pain location and pain affect (see Table 3). In the tablet condition, most participants obtained a perfect score for pain affect. These results in both conditions also applied to the participants who did not require support (see Table 3). The larger number of participants who were able to successfully answer questions concerning pain location with the laptop ($n = 22$ in total) than with the tablet ($n = 9$ in total) was statistically significant (McNemar test, $p = .007$, $\Phi = .01$). It was observed that the small displayed body parts easily resulted in touching a body part next to the target (e.g., head and neck) and that the sometimes slow internet connection on the tablet easily resulted in touching the same body part twice (i.e., risking an incorrect answer to the subsequent question).
The differences between participants with current pain and participants with remembered pain were not statistically significant concerning performance time of the test session (laptop: $t(38) = 0.27, p = .79, r = .04$; tablet: $t(38) = -0.10, p = .92, r = .02$) and the number of comprehension tests with perfect scores (laptop: $t(38) = 0.43, p = .67, r = .07$; tablet: $t(38) = 1.17, p = .25, r = .19$).

**TABLE 3**
Results of comprehension (recognition and navigation) tests ($N = 40$)

<table>
<thead>
<tr>
<th></th>
<th>Laptop n (% of 40) with perfect score</th>
<th>Tablet n (% of 40) with perfect score</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>In total</td>
<td>Without support</td>
</tr>
<tr>
<td>Pain location</td>
<td>22 (55%)</td>
<td>14 (35%)</td>
</tr>
<tr>
<td>Pain intensity</td>
<td>6 (15%)</td>
<td>3 (8%)</td>
</tr>
<tr>
<td>Pain affect</td>
<td>22 (55%)</td>
<td>10 (25%)</td>
</tr>
<tr>
<td>Pain quality</td>
<td>8 (20%)</td>
<td>8 (20%)</td>
</tr>
</tbody>
</table>

What kinds of information are adults with Down syndrome able to report with the online application?

The first question in the STOP-ID! was “Are you in pain today?”. The participant reported the presence of current pain by choosing YES ($n = 16$ with laptop, $n = 15$ with tablet). If the participant answered NO, then the researcher asked “Were you recently in pain?”. Some of the participants answered affirmative ($n = 14$ with laptop, $n = 7$ with tablet). Those individuals who obtained perfect scores on at least one comprehension test of recognition and navigation were asked to provide information about the current or remembered pain experience. Participants who had not reported current or recent pain were asked to think about pain that they might have experienced in the past ($n = 10$ in laptop condition, $n = 18$ in tablet condition). It was not within the scope of the present study to examine whether the reported pain was accurate (i.e., truly represented the experience of the participant). A McNemar test showed that the association between the self-reported presence of pain in the two conditions was statistically significant ($p < .001$), in which a Phi coefficient of .59 suggested a moderate to large test-
retest reliability. In total, 8 out of 40 participants reported the presence of pain differently in both conditions.

It depended on the performance on the comprehension tests whether participants reached the self-report part of the test session in both the laptop and tablet conditions. Table 4 shows the available information about the self-reported pain experience. Twenty-nine participants (73%) in the laptop condition reported information about at least one pain component (n = 20 without support). Twenty-six participants (65%) in the tablet condition reported information about at least one pain component (n = 21 without support). In both tablet and laptop conditions, participants most frequently chose the highest level of pain affect and the abdomen was a frequently reported pain location (see Table 4). The average pain intensity on the 0-10 scale was moderate to severe, with $M = 6.7$ ($SD = 2.9$) in the laptop condition and $M = 7.8$ ($SD = 3.1$) in the tablet condition, but this was based on only the six participants who passed the comprehension test. Pain quality was not used to report pain experience, because pain location, affect, and intensity were deemed more clinically relevant as (alarming) information for health care workers to determine whether further pain assessment is necessary.

Do adults with Down syndrome require assistance and if so, what kinds of support are needed?

Some participants were able to use the tool independently ($n = 7$ laptop, $n = 7$ tablet), a few needed only assistance with the use of the device ($n = 0$ laptop, $n = 4$ tablet), some needed only assistance with the questions about the comprehension tests and/or self-report of pain ($n = 18$ laptop, $n = 16$ tablet), and others needed assistance with both the use of the device and the questions ($n = 15$ laptop, $n = 13$ tablet). Verbal and/or non-verbal requests of assistance were made by 68% of the participants in the laptop condition and by 70% of the participants in the tablet condition. A Wilcoxon signed-ranks test showed that the larger number of times that assistance was provided during the laptop condition than during the tablet condition was statistically significant, $z = -2.39$, $p = .017$, $r = -.38$, $Mdn_{\text{laptop}} = 6$, $Mdn_{\text{tablet}} = 5$.

Table 5 describes the parts that needed extra explanation. In both conditions, most participants required assistance to use an arrow as a symbol for CONTINUE. The comprehension test for pain intensity was the comprehension test that
### Table 4
Reported pain experience by means of the Self-Reported Tool on Pain in People with Disabilities ($N = 40$)

<table>
<thead>
<tr>
<th>Reported information</th>
<th>Laptop condition</th>
<th>Tablet condition</th>
</tr>
</thead>
<tbody>
<tr>
<td>None</td>
<td>11 (28%)</td>
<td>14 (35%)</td>
</tr>
<tr>
<td>Only pain location</td>
<td>6 (15%)</td>
<td>4 (10%)</td>
</tr>
<tr>
<td>Only pain affect</td>
<td>6 (15%)</td>
<td>12 (30%)</td>
</tr>
<tr>
<td>Only pain intensity</td>
<td>0 (0%)</td>
<td>2 (5%)</td>
</tr>
<tr>
<td>Pain affect and intensity</td>
<td>1 (2%)</td>
<td>3 (8%)</td>
</tr>
<tr>
<td>Pain location and affect</td>
<td>11 (28%)</td>
<td>4 (10%)</td>
</tr>
<tr>
<td>Pain location and intensity</td>
<td>1 (2%)</td>
<td>0 (0%)</td>
</tr>
<tr>
<td>Pain location, affect, and intensity</td>
<td>4 (10%)</td>
<td>1 (2%)</td>
</tr>
<tr>
<td>Pain affect:</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>0</td>
<td>3</td>
<td>3</td>
</tr>
<tr>
<td>1</td>
<td>9</td>
<td>5</td>
</tr>
<tr>
<td>2</td>
<td>10</td>
<td>12</td>
</tr>
<tr>
<td>Pain location:</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>head (front)</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>chest</td>
<td>3</td>
<td>1</td>
</tr>
<tr>
<td>abdomen</td>
<td>7</td>
<td>2</td>
</tr>
<tr>
<td>arms (front)</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>legs (front)</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>head (backside)</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>neck</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>back</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>arms (backside)</td>
<td>3</td>
<td>1</td>
</tr>
<tr>
<td>legs (backside)</td>
<td>5</td>
<td>0</td>
</tr>
</tbody>
</table>
required the most assistance in both conditions. Remarkably, a relatively high number of participants needed assistance with the question about the presence of pain (58% in the laptop condition and 38% in the laptop condition).

**TABLE 5**
Number of participants who needed extra instruction, specified for the parts of the Self-Reported Tool on Pain in People with Disabilities

<table>
<thead>
<tr>
<th>Use of device</th>
<th>Laptop</th>
<th>Tablet</th>
</tr>
</thead>
<tbody>
<tr>
<td>mouse</td>
<td>15 (38%)</td>
<td>-</td>
</tr>
<tr>
<td>touch screen</td>
<td>-</td>
<td>17 (43%)</td>
</tr>
<tr>
<td>Use of arrow (CONTINUE)</td>
<td>31 (78%)</td>
<td>29 (73%)</td>
</tr>
<tr>
<td>Presence of pain</td>
<td>24 (58%)</td>
<td>15 (38%)</td>
</tr>
<tr>
<td>Comprehension</td>
<td></td>
<td></td>
</tr>
<tr>
<td>pain location</td>
<td>17 (43%)</td>
<td>17 (43%)</td>
</tr>
<tr>
<td>pain intensity</td>
<td>26 (65%)</td>
<td>18 (45%)</td>
</tr>
<tr>
<td>pain affect</td>
<td>10 (25%)</td>
<td>7 (18%)</td>
</tr>
<tr>
<td>pain quality</td>
<td>8 (20%)</td>
<td>7 (18%)</td>
</tr>
<tr>
<td>Self-report of pain</td>
<td></td>
<td></td>
</tr>
<tr>
<td>pain location</td>
<td>7 (18%)</td>
<td>1 (3%)</td>
</tr>
<tr>
<td>pain intensity</td>
<td>1 (3%)</td>
<td>1 (3%)</td>
</tr>
<tr>
<td>pain affect</td>
<td>3 (8%)</td>
<td>5 (13%)</td>
</tr>
<tr>
<td>pain quality</td>
<td>0 (0%)</td>
<td>2 (5%)</td>
</tr>
</tbody>
</table>

**Do adults with Down syndrome prefer one computer device over another?**

The reasons that most participants gave for their preference could be categorized into 'easier to use' (40% for laptop, 25% for tablet) and 'more attractive' (10% for laptop, 18% for tablet). Some participants gave several reasons. Two participants did not have a preference. The preference for the type of computer to perform the STOP-ID! was almost equally distributed: 53% (n = 20) for the laptop and 47% (n = 18) for the tablet, $\chi^2(1, n = 38) = 0.11, p = .75$. Having a mild level of cognitive impairment was related to preference of the laptop (71%) and having a moderate...
to severe level was related to preference of the tablet (62%), $\chi^2(1, n = 38) = 3.98$, $p = .046$, $\Phi = .32$.

DISCUSSION

The results provide evidence that the use of tools such as STOP-ID! appears to be a promising approach for the reporting of pain information by some adults with Down syndrome in the presence of a trained caregiver. The moderately high percentage of participants who demonstrated the ability to report information by using STOP-ID! (73% with laptop, 65% with tablet) suggests that the online application has potential for self-reporting pain, especially as most participants (68%) had never used a laptop or tablet before. However, the finding that only seven participants (18%) were able to perform the entire test session without assistance indicates that the additional value of the STOP-ID! is tentatively limited in terms of increasing individual capabilities for autonomous self-reporting of pain and in reducing the workload of caregivers.

Although 58% of the sample expressed pain verbally in typical pain situations, the use of the STOP-ID! may facilitate the communication about more pain components than only the presence. To optimize the potential, the application may benefit from additional changes. For example, it may be of benefit to concretize the question about the presence of pain could be by using ‘now’ instead of ‘today’: the use of explicit terms might help to reduce underreporting of pain. The use of the arrow as symbol for CONTINUE often needed assistance: it may be preferable that participants automatically continue to the next page after the text has been read aloud (introduction) or an answer was selected. Displaying a larger human figure may reduce the risk of erroneously selecting the adjacent body part; it has been previously found that people with Down syndrome benefit from a human figure with enlarged body parts to report their pain location [4]. Also, additional work is needed to address challenging issues related to representation. Our results show that comprehending a numeric pain scale is more difficult than a facial pain scale, and that comprehending quality-of-pain pictograms is difficult, possibly because pain quality is an abstract concept. A numeric rating scale is appropriate for a developmental level of eight years, because comprehension of the quantitative significance of numbers needs to be developed [1]. Quantity of
pain can be represented in more simplified ways than numbers, such as cubes of increasing size [4] or the number of poker chips [1].

More instruction prompts were needed in the laptop condition (\(Mdn = 6\)) than in the tablet condition (\(Mdn = 5\)). Many participants required assistance in manoeuvring the mouse, clicking without moving the cursor, and using only the left mouse button. The task of mastering these motor skills and the ability to remember them makes the use of a computer mouse more complex than the use of a touch screen, as has been described in literature [43]. The participants’ own preferences also confirmed the difficulty of using a laptop: most of the participants with mild intellectual disabilities gravitated towards the laptop, whereas the majority of those with moderate to severe intellectual disabilities preferred the tablet. Still, some difficulties with using the tablet were also observed by participants (e.g., pressing too long). It was observed that particularly the participants with severe intellectual disabilities had difficulties in using a computer device and in comprehending the questions.

It took participants an average of 14.6 minutes to complete the STOP-ID! in the tablet condition and 16.0 minutes in the laptop condition. The observed distraction, impulsivity, and relatively long performance time may be explained by the difficulty of the questions, the observed difficulty with using a computer mouse (sometimes also with using a touch screen), and the slow internet connection of the tablet. This indicates the need to closely examine the number of questions, and the representations used for the answers, which may increase the usability of the STOP-ID!. Reducing the length of the STOP-ID! will strengthen its practical use as a screening tool for pain. Using a 4G internet connection and offering a short basic training for using a computer mouse may further facilitate the use of the STOP-ID!

Our usability study was an essential first step towards developing a novel solution for technology-aided pain assessment aimed at reducing the undertreatment of pain in people with intellectual disabilities. Future research should also explore whether individuals can be trained to use the STOP-ID! on a regular basis by examining whether autonomous use of this tool increases over time. It is important to determine whether this is possible, as that would signify that frequent, autonomous use is an attainable goal and that tools such as STOP-ID!
are suitable for broad implementation in clinical practice to complement pain diagnostics through its inclusion, among other things, in the care plans of care-providing facilities.

Another important area for further research is the concurrent validity of the STOP-ID! as a screening tool for pain assessment. For example, the STOP-ID! could be compared with the same self-reporting pain scales assessed person to person, with other self-reporting pain scales assessed person to person (e.g., Coloured Analogue Scale or Facial Affective Scale: [32]), and/or with proxy ratings of pain intensity (e.g., NRS: [6]). Careful evaluation of the implementation is required to determine whether (regular) use of the STOP-ID! improves pain assessment and management in clinical practice.

**Limitations**

The findings reported here should be interpreted with caution as they represent the initial investigation of an innovative tool for the assessment of a challenging topic (i.e., pain) by people with intellectual disabilities. Participants without current pain were asked to think about pain that they might have experienced recently, so that use of all parts of the STOP-ID! could be evaluated in each participant. However, questions about recent pain appeal more to memory than questions about current pain and could therefore be more difficult to answer [37]. Although our results suggest that this had not biased the use of the STOP-ID! (i.e., performance time and number of perfectly scored comprehension tests were comparable), the reported information about a remembered pain should be interpreted cautiously, as there is no information on the concurrent validity of this tool (i.e., would the same information have been collected if it is was collected in other formats, by example verbal report?). Although it was not the aim of the current study, it was not possible to verify if the self-reported pain reflected the actual experience of the participants. More research is also needed on the test-retest reliability of the STOP-ID!, because eight participants reported inconsistently the presence of their pain in the two conditions.

In addition, the use of qualitative observations of only one rater may have biased the results due to the lack of inter-rater agreement data. Further, the exclusion criterion of severe visual impairments and hearing loss was only checked by asking the caregiver during the selection procedure to estimate whether the
participant was able to see pictures clearly and the hear clearly what is being said. The tablet was more susceptible to poor performance in the presence of weak wireless signals and was, consequently, sometimes slower than the laptop. This could have influenced the preference of participants for the laptop.

**Conclusion**

The two objectives for developing the STOP-ID! were (1) to enable people with intellectual disabilities to self-report pain and (2) to provide a standardized information gathering tool that covers multiple aspects of pain experience. As a result of the study, we can conclude that the STOP-ID!, if modified, might accomplish these objectives, however more research is needed to establish reliability and to validate the tool by comparing with other means of pain assessment. The presence of a trained caregiver is recommended for the current version. Modifications in length of the tool, selection of graphic images, and phrasing of questions are needed to improve the autonomous use of the STOP-ID! and hence its additional value for clinical practice as a screening tool for pain.

**REFERENCES**


[7] Böhmer CJM, Niezen - De Boer, M.C., Klinkenberg-Knol EC, Devillé WLJM, Nadorp JHSM, Meuwissen SGM. The prevalence of gastroesophageal reflux disease in institutionalized


[23] De Knegt NC, Pieper MJ, Lobbezoo F, Schuengel C, Evenhuis HM, Passchier J, Scherder EJA. Behavioral pain indicators in people with i-


**SUPPLEMENTARY MATERIAL**

**Scoring form pilot study STOP-ID**

**Observational Checklist STOP-ID!: Comparison between Laptop and Tablet**

Participant number: __________________________

Date today: __________________________

Age: __________________________

Gender: [ ] Male [ ] Female

Order of exposure: [ ] Laptop -> Tablet [ ] Tablet -> Laptop

Instructions: You will do two tasks. I would like to know if you have any pain today and if so, how it feels. [natural speech]

Questions for the caregiver about medical and cognitive change since the first test session

1. Did medical changes occur in the previous year that are still present? YES/NO
2. If so, what are they? ……………………………………………………………………………………………
3. Did cognitive changes occur in the previous year that are still present? YES/NO
4. If so, what are they? ……………………………………………………………………………………………

Questions for the caregiver about ability to read and familiarity with pictograms

1. Is the participant able to read? YES/NO
2. Does the participant use pictograms for communication? YES/NO
3. If so, with which frequency? [ ] Less than daily [ ] Daily [ ] Weekly
4. Do these pictograms mainly consist of facial expression to communicate? YES/NO

Questions for the caregiver about LAPTOP

1. Is the participant familiar with a laptop? YES/NO
   (Has he/she ever used a laptop? Is he/she able to switch it on and start to work without support?)
2. On average, how many hours per week does the participant use a laptop? …………
3. Is the participant familiar with a regular computer? (for ‘familiar’: see question 1) YES/NO
4. On average, how many hours per week does the participant use a regular computer? …………

Questions for the caregiver about TABLET

1. Is the participant familiar with a tablet? YES/NO
   (Has he/she ever used a tablet? Is he/she able to switch it on and start to work without support?)
2. On average, how many hours per week does the participant use a tablet? …………
3. Is the participant familiar with another kind of touch screen? YES/NO
4. How many hours per week (average) does the participant use this touch screen? …………
Question for the participant at the end: Preference ☐ Laptop ☐ Tablet
Reason for preference: ....................................................................................................................

(We have completed two tasks now. Which task did you like best? Why? [natural speech])

STOP-ID test on LAPTOP: Participant reports pain ☐ Yes ☐ No
Participant is able to insert own account-number YES/NO
Recognizes own photo YES/NO
Total time (minutes) needed for participant to complete STOP-ID! (incl. support): ............
(Total time = from the starting screen up to and including the closing screen)
How many times in total does the instruction need to be repeated?* ............
Participant asks for help verbally or says that he/she does not know what to do YES/NO
Participant asks for help non-verbally (looks confused around room/ at researcher) YES/NO
Participant seems distracted/ bored YES/NO
Participant is responding impulsively (takes no time to think well about the answer) YES/NO
Participant is able to finish the test YES/NO

*Parts that needed extra explanation: ...........................................................................................................
.................................................................................................................................................................

Extra observations:
.................................................................................................................................................................
.................................................................................................................................................................
.................................................................................................................................................................
.................................................................................................................................................................
.................................................................................................................................................................
<table>
<thead>
<tr>
<th>Activity</th>
<th>YES/NO</th>
</tr>
</thead>
<tbody>
<tr>
<td>STOP-ID test on TABLET: Participant reports pain</td>
<td></td>
</tr>
<tr>
<td>Participant is able to insert own account-number</td>
<td></td>
</tr>
<tr>
<td>Recognizes own photo</td>
<td></td>
</tr>
<tr>
<td>Total time (minutes) needed for participant to complete STOP-ID (incl. support):</td>
<td></td>
</tr>
<tr>
<td>(Total time = from the starting screen up to and including the closing screen)</td>
<td></td>
</tr>
<tr>
<td>How many times in total does the instruction need to be repeated?*</td>
<td></td>
</tr>
<tr>
<td>Participant asks for help verbally or says that he/she does not know what to do</td>
<td></td>
</tr>
<tr>
<td>Participant asks for help non-verbally (looks confused around room/ at researcher)</td>
<td></td>
</tr>
<tr>
<td>Participant seems distracted/ bored</td>
<td></td>
</tr>
<tr>
<td>Participant is responding impulsively (takes no time to think well about the answer)</td>
<td></td>
</tr>
<tr>
<td>Participant is able to finish the test</td>
<td></td>
</tr>
</tbody>
</table>

*Parts that needed extra explanation: .................................................................

Extra observations:

.................................................................................................................................

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