Needle-related pain and distress management during needle-related procedures in children with and without intellectual disability

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Abstract
Children with intellectual disability frequently undergo needle-related procedures for diagnosis or treatment. Nevertheless, only a few studies deal with pain and distress management during the procedure in this population of children. This study aimed to investigate the number of anxiety and pain management techniques performed during needle procedure in children with intellectual disability (cases) compared to a population of children without intellectual disability (controls). This multicenter cohort study was performed from July 2016 to January 2018 in the pediatric ward of four urban hospitals in Italy. Eligible subjects were children with and without intellectual disability, from 4 to 17 years old, who needed venipuncture or intravenous cannulation for diagnosis or treatment. Use of topical anesthesia, distraction techniques, and physical or verbal comfort during procedures were recorded. Pain and anxiety scores were also recorded. Forty-seven cases and 94 controls were recruited. Three pain- and anxiety-relieving techniques were performed during the procedure in 12 (25%) cases and in 10 controls (11%); two techniques were performed in 23 (50%) cases and in 26 (28%) controls; 12 (25%) cases and 52 (55%) controls received only one.

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Conclusion: In this series, children with intellectual disability received significantly more relieving techniques, but experienced more pain and anxiety when compared to children without intellectual disability.

What is Known:
- Children with intellectual disability experience more episodes of pain than cognitively healthy ones, and almost 10% of these episodes are due to medical procedures.

What is New:
- Children with intellectual disability despite receiving more relieving techniques during a needle-related procedure experienced more pain and anxiety when compared to healthy children.

Keywords Intellectual disability · Procedural pain · Procedural distress · Distraction

Abbreviations
CEMS Children’s Emotional Manifestation Scale
ID Intellectual disability
NCCPC-PV Non-Communicating Children’s Pain Checklist-Postoperative Version

Introduction

During their life, children with intellectual disability (ID) secondary to cerebral palsy or genetic syndromes experience more episodes of pain than cognitively healthy ones [3]. Children with more severe ID experience the worst pain, with a significant impact on the functioning [4]. In these patients, episodes of pain are mainly related to painful conditions associated with their syndromes or disorders, but almost 10% of these episodes are due to medical procedures [3].

Children with intellectual disability represent a challenge for the pediatricians due to their higher need for repeated procedures and the difficulties in recognizing and managing their fear, anxiety, and pain. Indeed, children with ID are often unable to verbalize their pain or may exhibit uncommon behaviors when they are in pain, leading to delayed recognition and management of pain [5, 8].

Moreover, while a massive amount of literature deals with procedural pain management, specifically with the efficacy of non-pharmacological and pharmacological techniques in healthy kids [19], little data are available for intellectually impaired patients.

Evidence shows that there is a discrepancy in pain management practices in children with ID and that children with ID may be at higher risk for having their pain undertreated if compared to cognitively healthy ones [12]. Nevertheless, to the best of our knowledge, no data are available comparing pain and distress management of children with and without ID during medical procedures in everyday practice.

As needle-related procedures are the most commonly performed procedures in hospitalized children [10], this study aimed to investigate the clinical approach to pain and distress management in children with and without ID during venipuncture or intravenous cannulation.

Patients and methods

This was a multicenter cohort study which was conducted from July 2016 to January 2018 in the pediatric wards of four urban hospitals in Italy:

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- Paediatric Palliative Care—Pain Service, Department of Women’s and Children’s Health, University of Padua
- Maternal and Child Department, Amas Civico, Palermo
- Pediatric Unit, Maternal and Child Department—Dolo Hospital, ULSS 3 Serenissima, Mestre

The study received approval from the Independent Bioethic Committee of each hospital. All of the children’s parents provided written informed consent before participation.

Every site enrolled patients subsequently for a period of 4 months, from Monday to Friday, during the morning hours when routine procedures were carried out. Cases and controls were enrolled in the same time frame.

The eligible subjects were children from 4 to 17 years of age, with or without ID who needed venipuncture or intravenous cannulation for diagnosis or treatment. Children with ID were recorded as test cases; children without ID were recorded as controls. For every case, two controls were consecutively recruited (case/control, 1:2), without matching.

Only children who have already been recognized with intellectual disability were enrolled. Intellectual disability was defined according to the most recent definition in DSM V: “a disorder with onset during the developmental period that includes both intellectual and adaptive functioning deficits in conceptual, social, and practical domains” [2]. Based on this definition, four degrees of severity were identified: mild, can live independently with minimum levels of support; moderate, independent living may be achieved with moderate levels of support, such as those available in group homes; severe, requires daily assistance with self-care activities and safety supervision; and profound, requires 24-h care. For this study, we considered severe and profound as an all the same category.
The exclusion criteria were as follows: the need for procedural sedation, the administration of an analgesic in the previous 8 h, a clinical diagnosis of a pervasive developmental disorder, the absence of the caregiver, and children already enrolled in this study.

Trying to assure an unbiased picture of procedural pain and distress management in hospitalized children with and without ID, every participating center designated an external observer, usually a nurse, for children recruitment and observations of the procedures. The external observer recruited both cases and controls. The external observer did not take part in the procedure or the child’s care. Both pediatric ward staff and children’s parents were informed that the external observer would record the children’s pain and distress during procedures, but they were blind to the registration of pain and distress management techniques.

In accordance with recent literature [23], for every procedure, we arbitrarily decided to record the following strategies: (1) use of topical anesthesia, (2) use of any distraction strategy (songs, doggerel, storytelling, soap bubbles, music, cartoons, tablet TV, etc.), and (3) use of physical (holding or touching the child) or verbal comfort adopted by caregiver or hospital staff during the procedure to cheer up the child.

For each enrolled child, the following variables were collected: age, gender, weight, presence and degree of intellectual disability, type of procedure (venipuncture or intravenous cannulation), the number of needle-related procedures that underwent in the last year before enrolment, and the need to physical restraint.

Procedural pain scores during procedures were collected by the external observer. For children with ID, the Non-Communicating Children’s Pain Checklist-Postoperative version (NCCPC-PV) was used. This scale is a validated pain assessment tool designed for children between 3 and 18 years of age with ID [2, 14]. This scale consists of 27 items. The observer establishes a score for each item on a five-step ordinal scale, according to the frequency of its occurrence: 0 = not at all, 1 = just a little, 2 = fairly often, 3 = very often, NA = not applicable. The total score (0–81) is obtained from the sum of each item’s score. A pain score < 11 indicates absence of pain or mild pain; a score of 11 or more indicates the presence of moderate to severe pain.

The same observer recorded pain in children without ID; for children between 4 and 7 years of age the Wong-Baker scale was used, while for older children (older than 7 years) the numerical rating scale was preferred [21, 22].

For the statistical analyses, scores from both scales were transformed into a numerical value from 0 for no pain to 10 for maximum pain, in accordance with previous studies, which demonstrated that scores reported using these two scales overlapped [9].

The external observer recorded the distress immediately before the procedure using the Children’s Emotional Manifestation Scale (CEMS) [11]. The CEMS defines five categories of behaviors (facial expression, vocalization, activity, interaction, and level of cooperation) rated by intensity from 1 to 5. The total score varies between 5 and 25 and it is obtained from the sum of each item. A higher score indicates higher distress. We consider two categories: no distress (5 points) and distress (> 5 points).

To the best of our knowledge, no validated tools are available for the assessment of distress in children with ID. Therefore, the observer asked the child’s caregiver a semi-quantitative judgment by giving three options: no distress, little distress, or a lot of distress.

The primary outcome of this study was to investigate the number of anxiety and pain management techniques performed during needle procedure in children with intellectual disability (cases) compared to a population of children without intellectual disability (controls).

The secondary outcomes included the pain and anxiety scores during the procedure in both groups, the relationship between pain and anxiety scores and the number of techniques employed in both groups, and the need for physical restraint during the procedure in both groups. By physical restraint, we mean the need to immobilize the child’s arms with a hand or one or more operators holding him still.

**Statistical analysis**

Differences in pain scores between groups were accessed with Mann-Whitney test while differences in categorical variables were accessed using chi-square test. A p value of less than 0.05 was considered as statistically significant. All the analyses were carried out with STATA 14 (StataCorp LLC, College Station, TX, USA).

**Results**

During the study period, we enrolled 47 children with intellectual disability and 94 children without intellectual disability. A convenience sample size of 60 children was stated. Since we were not able to find any previous similar study in the literature, this number was based on previous studies validating specific pain scales in children with ID [24]. Due to time restraints and difficulties in enrollment, the final number was 47. Sample size was not equally distributed between the centers, with one center enrolling 65% of patients and the other three centers equally enrolling the remaining 35%. Hospital staff in all centers previously received the same training in pain and anxiety management in children. Sixty-two patients were eligible for the study, seven patients refused to participate, and eight patients were not enrolled due to family’s or staff’s time restraints.

The main characteristics of the study population are reported in Table 1.
Children with intellectual disability were affected by genetic disorders in 18 cases (38%), cerebral palsy in 12 cases (26%), epileptic encephalopathy in 3 cases (6%), metabolic disease in 2 cases (4%), and other conditions in 11 cases (23%).

Children with intellectual disability had a mild intellectual disability in 11 cases (23%), a moderate disability in 16 cases (34%), and a severe disability in 20 cases (43%).

Forty-four children without intellectual disability (47%) were affected by a chronic disease (gastroenterological, endocrinological or reumatological disorders, or epilepsy). Oncological patients were excluded.

Both parents were present more often in children with ID (see Table 1). There was no difference in pain scores in the three subgroups (with mother, with father, or both) in children either with ID or without.

The number of pain- and anxiety-relieving techniques performed in both groups (main study outcome) was reported in Table 2. There was a statistically significant difference between the number of techniques employed between the two groups (p < 0.01).

EMLA cream was used in 25 children with intellectual disability (53%) and in 31 without intellectual disability (33%) (p = 0.021). Distraction techniques by hospital staff were employed in 22 cases (47%) and in 19 controls (20%) (p = 0.001). Twelve cases (25%) and 11 controls (12%) received both pharmacological and distraction techniques (p = 0.036).

Medical staff or parents carried out a physical or verbal/visual comfort in 47 children (100%) with intellectual disability and in 84 children (89%) without intellectual disability, and this difference was statistically significant (p = 0.03). Physical comfort (touching or rubbing the child) was observed in 45 cases (96%) and in 52 controls (55%) (p < 0.01).

Figure 1 shows the number of children with mild and with moderate to severe pain scores and the number of children with distress in both groups. The number of children with moderate to severe pain scores and with procedural distress was statistically significantly higher in children with ID (p < 0.01 and p = 0.002, respectively).

Moderate to severe pain is more frequently experienced by children with ID (Table 3, A). This difference remains in a stratified analysis based on numbers of used techniques: in each equal subgroup, children with ID experienced significantly more often moderate to severe pain (Table 3, B).

Pain scores increased with the level of disability as shown in Fig. 2: a median score of 12 (of 81; IQR 6–23) was recorded in children with mild disability, a median score of 20 (IQR 14.5–28.5) in those with moderate disability, and a median score of 23 (IQR 12–33) in those with severe disability (p 0.15).

In children with intellectual disability, a positive trend was also found between pain and anxiety scores with anxious children experiencing more pain: median pain score 12 (IQR 11–14) in those who do not experienced distress, 13 (IQR 6–23) in those who felt some distress, and 28 (IQR 17–39) in those who registered marked distress, by judgment of parents (see Supplement 1).

The number of children who needed immobilization to perform the procedure was 25 (53%) in the test group, and 14 (15%) in the control group, and this difference was significant (p < 0.001).

### Discussion

This study shows that even though children with intellectual disability receive more pain-relieving techniques, they still experience more pain and anxiety during needle-related procedures when compared to typically developing pairs of children.

Needle-related procedures are one of the most common sources of distress in hospitalized children [10], with international guidelines warranting a prompt pain and anxiety management [6].

Topical anesthesia, distraction, and physical and verbal comfort are useful techniques for pain and anxiety management during needle-related procedures, with substantial evidence supporting their use in everyday practice [16, 18, 20, 23, 25]. Frequently, children with intellectual disability undergo needle-related procedures for diagnosis or treatment. Many of these children are unable to communicate their pain verbally. Nevertheless, this impairment should not negate the possibility of an appropriate pain-relieving treatment [7].

### Table 1 Main characteristics of the study population

<table>
<thead>
<tr>
<th></th>
<th>Children with intellectual disability (cases)</th>
<th>Children without intellectual disability (controls)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number</td>
<td>47</td>
<td>94</td>
</tr>
<tr>
<td>Age (years ± SD)</td>
<td>9.3 ± 3.9</td>
<td>9.7 ± 3.9</td>
</tr>
<tr>
<td>Male, n (%)</td>
<td>28 (60%)</td>
<td>45 (48%)</td>
</tr>
<tr>
<td>Weight (median, kg)</td>
<td>32.9</td>
<td>37.3</td>
</tr>
<tr>
<td>Caregiver:</td>
<td></td>
<td></td>
</tr>
<tr>
<td>• Mother</td>
<td>26 (55%)</td>
<td>49 (52%)</td>
</tr>
<tr>
<td>• Father</td>
<td>3 (6%)</td>
<td>23 (24%)</td>
</tr>
<tr>
<td>• Both</td>
<td>18 (38%)</td>
<td>22 (23%)</td>
</tr>
<tr>
<td>Number of needle procedures in the last year:</td>
<td></td>
<td></td>
</tr>
<tr>
<td>- Less than two</td>
<td>18 (38%)</td>
<td>52 (55%)</td>
</tr>
<tr>
<td>- Two or more</td>
<td>29 (62%)</td>
<td>42 (45%)</td>
</tr>
<tr>
<td>Type of intervention:</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Venipuncture, n (%)</td>
<td>38 (81%)</td>
<td>80 (85%)</td>
</tr>
<tr>
<td>Intravenous cannulation, n (%)</td>
<td>9 (19%)</td>
<td>14 (15%)</td>
</tr>
</tbody>
</table>
Specific assessment tools are available for measuring pain in this population of patients [14], so that the effectiveness of specific pain- and anxiety-relieving strategies in this setting could be evaluated. Despite this, very little data are available in the literature about the use and the effectiveness of pharmacological and non-pharmacological techniques to reduce pain during needle-related procedures in children with intellectual disability. In fact, only single experiences were published on this topic. Clowns, who are known to be effective in reducing children’s anxiety in hospital settings [7], were useful in distracting patients with a mild intellectual disability during botulinum toxin injections in a cohort of 45 children [1].

Distraction, counter conditioning, behavioral therapy, and topical anesthesia have been shown to be effective in a small cohort of intellectually impaired children requiring repeated procedures [17].

To the best of our knowledge, no data are available about how frequently pain- and anxiety-relieving strategies are used to manage children with intellectual disability in everyday practice. Moreover, no data are available regarding how the intellectual disability may influence the effectiveness of a specific pain- and anxiety-relieving technique. In this sense, while the relationship between per-procedural anxiety, pain experience, and memory of the procedural experience is well reported in healthy children [15], we are not aware of any study dealing with this issue in ID patients. The issue of anxiety may be particularly relevant since both clinical experience and parents’ reports suggest that these patients experience significant degrees of anxiety and fear during procedures. It could be hypothesized that many defensive mechanisms which require fully developed fronto-limbic connection and are based on rationalization, sense of belonging, and ability to search and receive comfort and distraction may be lacking in ID patients, making them more vulnerable.

Our work aimed to investigate if hospitalized children with intellectual disability would receive a different pain and anxiety management approach during needle-related procedures when compared to typically developing peers. Therefore, we decided to focus our attention to the strategies most frequently performed during needle-related procedures by hospital staff and parents, such as the application of a topical anesthetic, distraction, and physical and verbal comfort.

Our study showed that children with intellectual disability received more pain- and anxiety-relieving techniques, with almost everyone receiving at least one technique, thus suggesting increasing attention for these children of the hospitals’ staff. Nevertheless, children with intellectual disability experienced more procedural pain and anxiety when compared to typically developed peers, both in general and also when they were stratified for the number of techniques used. Remarkably, our data showed that observed pain was directly related to anxiety and to the degree of intellectual disability with the more compromised children experiencing the worse pain.

This study has some limitations. First, it was carried out in settings in which hospital staff is highly trained in the care of children with intellectual disability, and this may have influenced our findings. Moreover, even though operators and

### Table 2 Main study outcome

<table>
<thead>
<tr>
<th>Number of pain- and anxiety-relieving techniques performed during the procedure</th>
<th>Children with intellectual disability (cases)</th>
<th>Children without intellectual disability (controls)</th>
</tr>
</thead>
<tbody>
<tr>
<td>3</td>
<td>12 (25%)</td>
<td>10 (11%)</td>
</tr>
<tr>
<td>2</td>
<td>23 (50%)</td>
<td>26 (28%)</td>
</tr>
<tr>
<td>1</td>
<td>12 (25%)</td>
<td>52 (55%)</td>
</tr>
<tr>
<td>0</td>
<td>0</td>
<td>6 (6%)</td>
</tr>
</tbody>
</table>

**Fig. 1** a Mild and moderate to severe pain in the two groups. b Presence of distress in the two groups.
parents were blinded about the recording of the pain management techniques during procedures, the presence of a watching external observer may have influenced their behaviors. Second, we are well aware that using more pain relief techniques during a procedure is not always synonymous of better care, but with the aim of delineating everyday practice, we needed an objective outcome to standardize this usual care. Topical anesthesia, distraction, and physical and verbal comfort could not be considered the only strategies available to reduce pain and anxiety during the procedure so that choosing to record only these techniques, we could have underestimated the number of techniques actually employed in both groups of children. Moreover, we could not exclude that other confounding factors, such as the quality of the environment, may have influenced the collected pain and anxiety scores during the procedure. Third, to evaluate pain, we compared parents’ observation with self-reports, but this limit could not be overcome due to impossibility of children with severe impairment to self-report their pain.

As well, a further limit is that, to record anxiety in children with intellectual disability, we used the semi-quantitative opinion of children caregivers, thus limiting the strength of this evaluation, but no validated tools are available for the assessment of anxiety during medical procedures in this peculiar population. No tool can reliably discriminate between pain and fear/anxiety and will capture observable behaviors and

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### Table 3

Number of children with moderate to severe pain in both groups compared for the number of techniques used during the procedure

<table>
<thead>
<tr>
<th>A. All patients</th>
<th>Children with intellectual disability (n = 47)</th>
<th>Children without intellectual disability (n = 94)</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Moderate/severe pain, number (%)</td>
<td>39 (83%)</td>
<td>19 (20%)</td>
<td>0.000</td>
</tr>
</tbody>
</table>

| B. Subgroups of children with the same number of techniques used | Children with intellectual disability | Children without intellectual disability | p value |
| Subgroup 0 techniques: | | | |
| Moderate/severe pain, number (%) | Children with intellectual disability (n = 0) | Children without intellectual disability (n = 6) | p value |
| Subgroup 1 technique: | | | |
| Moderate/severe pain, number (%) | Children with intellectual disability (n = 12) | Children without intellectual disability (n = 52) | p value |
| Subgroup 2 techniques: | | | |
| Moderate/severe pain, number (%) | Children with intellectual disability (n = 23) | Children without intellectual disability (n = 26) | p value |
| Subgroup 3 techniques: | | | |
| Moderate/severe pain, number (%) | Children with intellectual disability (n = 12) | Children without intellectual disability (n = 10) | p value |

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![Fig. 2](image) Relationship between pain scores and level of intellectual disability
distress. It can be postulated that observers’ pain assessment measured by the NCCP-PV was influenced by anxiety and fear, since some behavioral items may be triggered by emotions. In this perspective, parental judgment of children anxiety may attenuate this bias.

Future study should focus on this issue and develop a dedicated anxiety score for children with ID.

Finally, the choice of the NCCP-PV for pain assessment may be debatable since some studies reported the revised Faces, Legs, Activity, Cry, and Consolability Scale (rFLACC) as the most convenient for pain measurement in children with intellectual disability [13]. NCPCC-PV is a validated tool already used in several studies to collect pain in children with intellectual disability during needle-related procedures [1, 14, 24], and on the contrary of rFLACC, the NCCP has the advantage of not needing a previous interaction with the caregiver for the definition of the items. Therefore, trying to minimize as much as possible the interactions between external observers and hospital staffs and caregivers, we decided to use NCPCC-PV.

In conclusion, this study showed that a cohort of children with intellectual disability, undergoing a needle-related procedure, experience more procedural pain and anxiety when compared to cognitively healthy children, even though they receive more pain- and anxiety-relieving techniques. Recognizing and managing fear, anxiety, and pain in these children represent a challenge for physicians. Future trends and research should focus on increasing physician’s skills in recognizing, measuring, and managing with pain and anxiety in children with ID, defining the most effective pharmacological and non-pharmacological techniques.

Acknowledgements We would like to thank every health professional worker, especially the nurses, who took care of the hospitalized children.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Informed consent Written informed consent was obtained from all parents of all children included in the study.

Ethical approval The independent ethical committee of our Institutes and of every participated center approved the study protocol before enrollment of the first participant.

Ethical standards All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Animal studies This article does not contain any studies with animals performed by any of the authors.

References


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