Odour detection in children and young people with profound intellectual and multiple disabilities

Geneviève Petitpierre | Juliane Dind | Catherine De Blasio | Germaine Gremaud

Department of Special Education, University of Fribourg, Fribourg, Switzerland

Correspondence
Geneviève Petitpierre, Département de Pédagogie Spécialisée, Université de Fribourg, R. St Pierre Canisius 21, CH-1700 Fribourg, Switzerland. Email: genevieve.petitpierre@unifr.ch

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Abstract
Background: Olfaction provides information on very important dimensions of the environment; however the olfactory abilities of children and young people with profound intellectual and multiple disabilities (PIMD) remain largely unknown. This within-subjects study explores olfactory detection abilities in children with PIMD.

Method: Twenty-two children and young people with PIMD (7–18 years) were presented with 18 medium intensity odours and an odourless control stimulus. Odorants were presented one by one in a randomised order. The neutral stimulus was presented prior to each odorant. Participants’ responses were measured using 21 behavioural indicators.

Results: Results show that participants make a clear distinction between odorous and neutral conditions, between food and non-food, and between pleasant and unpleasant odours. The detection abilities are manifested by several behaviours, in particular by the duration of the head alignment on the odorant.

Conclusions: This study shows that participants detect the stimuli and act differently depending on the category.

KEYWORDS
odour, olfaction, profound intellectual and multiple disabilities, sensory behaviour

1 | INTRODUCTION

Since most persons with profound intellectual and multiple disabilities are also hindered by visual and/or auditory impairments, the olfactory function, if intact, can be a useful compensatory tool to strengthen interactions between these people and their environment. This sensory modality provides information on essential dimensions of physical and social contexts (Schaal et al., 2020; Stevenson, 2010). Observations about olfactory functioning in people with PIMD are nevertheless anecdotal. Roemer et al. (2018) found that people with PIMD use the sense of smell least of all the senses. An explanation for the lack of studies on olfaction in people with PIMD lies in the fact that conducting experimental studies involving this population is very challenging (Maes et al., 2021). People with PIMD form a heterogeneous group in terms of the origin of the disability (Nakken & Vlaskamp, 2007). Their participation is strongly impacted by profound or severe intellectual impairments combined with profound or severe motor disorders. They often have to deal with complex medical conditions and present frequent sensory impairments, especially visual and auditory (Dorche, 2021; Jacquier, 2021; van Blarikom et al., 2009). Understanding and communicating with them is often difficult as they mainly, if not exclusively, express in non-, proto- or pre-verbal ways, or via idiosyncratic communicative behaviours.
Knowledge available for other populations with neurodevelopmental disorders is not helpful as it remains ‘close to nothing’ (Dan, 2019). Atypical olfactory processing or morphological atypicalities have been indicated in a few syndromes (Lyons Warren et al., 2021; Manan & Yahya, 2021; Samat & Flores-Samari, 2017, 2019) but, except for autism spectrum disorders (Crow et al., 2020; Tonacci et al., 2017 for reviews), data are scarce and not conclusive. In people with cerebral palsy, no relationship was found between olfactory and gross motor functions (Nakashima et al. 2019).

With respect to evaluation, olfactory standardised tests, which are attentionally, verbally and cognitively demanding, can only be administered from 3 to 4 years of developmental age (Cameron, 2018). Hetero-evaluation could be used to explore the perceptive abilities of people with PIMD; however this is an indirect procedure, which may reduce the quality of the observations reported (Todorov & Kirchner, 2000). Notwithstanding its cost and high degree of technicality, psychophysical assessment offers the advantage of a direct and rigorous measurement. Commonly used to observe infants’ olfactory functioning, this procedure can be used beneficially in pre- or non-symptomatic populations difficult to study by other means.

With respect to intervention, the development of olfaction is rarely mentioned as an objective in personalised educational programs for people with PIMD (Petitpierre et al., 2017). Some incentives to use odours with this population do already exist to prevent sensory deprivation or boost cognitive functioning (Fröhlich, 2000; Government of Ireland, 2006; Haute Autorité de Santé, 2020; Hulsegge & Verheul, 1987). Olfactory stimuli are also used in multi-sensory books as a ‘safe’ alternative choice when the direct support persons are not sure or lack prior knowledge about a person’s tactile preferences or abilities (Ten Brug et al., 2012). However olfactory stimulation in daily care is currently only empirical. The support workers do not have guidelines to observe and interpret the responses manifested by people with PIMD, and their intervention usually takes place without any evidence, which is not very satisfactory. Educational staff report that olfactory functioning is one of the sensory dimensions for which they need more knowledge (Vlaskamp et al., 2007).

The present study has been developed at the request of a special needs education school for children and young people with PIMD. This school has used olfactory stimulation for several years and was wondering how to scientifically investigate how children and young people with PIMD respond to odours. The two following research questions were formulated in collaboration with the school: do children and young people with PIMD behave differently in the presence of an olfactory versus a neutral stimulus, and what detection behaviours do they exhibit? Because of the scarcity of studies on the topic, the hypothesis corresponding to the first question was formulated on a bilateral basis, that is, children and young people with PIMD were expected to behave differently in the presence of an olfactory versus a neutral stimulus, regardless of the direction of the difference. Moreover, since factors such as gender, age, mood, medication, as well as olfactory environment are likely to moderate odour detection and perception, exploratory analyses targeting the role of some observed, not manipulated, individual factors were also conducted.

2 | METHODS

2.1 | Procedure

This within-subjects study has been authorised by the Geneva and Vaud ethics committees (ID: 2019-00234), which are part of the national organisation Swiss ethics. Written consent was obtained from parents or legal guardians before the start of the study. The researchers committed to postpone the session if the child’s state of alertness, fatigue or health required it and to permanently discontinue a participant’s participation if they showed discomfort after two sessions of odour exposure. A pilot study preceded the experimental study in accordance with the recommendations of Maes et al. (2021).

2.2 | Participants

The participants were recruited in three Swiss French-speaking special education schools. The referent teacher or support worker were asked to select participants meeting the three following criteria: (1) presenting the key characteristics described by Nakken and Vlaskamp (2007); (2) be aged between 7 and 18 years; (3) have attended the school for at least 6 months at the time of the recruitment. Children presenting food allergies or chronic airway problems with indication of daily respiratory therapy were excluded. Twenty-five children and young people met the criteria and were included in the convenience sample of the study. Three children left the study at the very beginning of data collection, two because of severe health problems, the third because he could not stand the experimental setting. The final sample of the study includes 22 participants aged between 7 and 18 years ($M = 13.0$ years; $SD = 3.4$), all attending a special educational school program. Demographic data (age of the child, gender, aetiology [if known], comorbidities, motor and communicative abilities) was collected from participants’ clinical files by their teachers and support workers. Participants were also assessed through the French validated version of the Mood, Interest and Pleasure Questionnaire [MIPQ] (Ross & Oliver, 2003; Verbel Sierra, 2013), and through questionnaires on gastrointestinal reflux and nausea responses (Senez, 2013a, 2013b). Participants’ characteristics are shown in Table 1.

2.3 | Stimuli

A total of 19 olfactory stimuli (18 odorous and 1 odourless solvent as control stimulus) were used (Table 2). Ten odors were food odours (basil, orange*, Nutella, green vegetables, rancid butter, cinnamon, cheese, strawberry, apple pie and garlic*), eight were non-food odours (chlorinated swimming pool water, pony, summer rain, rose*, lily of the valley, grass*, hand sanitizer [Sterilium®]** and sweat). Stimuli were selected in collaboration with the school that initiated the research. Odours that might be present in the context of everyday life
were preferred. The stimulus set corresponds to six of the eight categories of the typology identified by Castro et al. (2013). Four stimuli marked with a single asterisk (*) come from the ‘Sniffin’ Sticks test’, a standardised worldwide-used test manufactured by Burghart Messtechnik GmbH [http://burghart-mt.de/en/]. One odour, marked with the double asterisk (**), comes from a commercially available product. The other 13 stimuli were created on request by the Swiss perfume and aroma company Givaudan SA. All ingredients used in the production of the stimuli meet the Swiss standards in terms of regulation on cosmetic products (OCos of 16 December 2016; ODAIOUs of 16 December 2016). The calibration of the intensity of the stimuli created for the study required a three-step procedure. In the first step, eight members of the steering group (two researchers, three representatives of the school that initiated the research and three representatives of the funding foundation), assessed the intensity of a first set of stimuli using a Likert scale ranging from 1 (no odour at all) to 9 (extremely strong odour). The concentration of some stimuli was adapted to reach an average perceived intensity. The stimuli were then tested for intensity by 29 adult volunteers working in the Givaudan company. All odours reached an intensity range of 4.5–6.5 (neither too strong nor too weak), except summer rain and pool water. We nevertheless decided to keep these two odours in the panel because the intensity range was not excessively small. For the weakest odour (pool water), we discarded the idea of increasing the proportion of chlorine in the water beyond the proportion usually used in swimming pools for safety reasons. The volunteers were also asked to identify the stimuli and to categorise them hedonically into three categories (pleasant, unpleasant, neither pleasant nor unpleasant). As they usually rate odorants as being more intense and more pleasant than adults (Bensafi et al., 2007), it would have been better if children, not adults, had performed this pre-categorisation. However cross-cultural judgements about bad smells are highly convergent (Schaal et al., 1998) even from an early age (Wagner et al., 2013), probably because bad odours have an alarm function and too much variability under the effect of environmental factors would put the individual at risk (Bensafi & Rouby, 2021). For good odours, variability is usually greater (Ferdenzi et al., 2011). In our sample, only four children out of 22 are of non-European origin; the other 18 are European through at

| TABLE 1 | Participants’ characteristics |
|---------|-----------------------------|
| Demographics (%) |               |
| Gender |               |
| Male | 14 (36) |
| Female | 8 (64) |
| Aetiology |               |
| Cerebral palsy | 5 (23) |
| Syndromic aetiology | 6 (27) |
| Within degenerative syndrome (n = 1) |               |
| Cerebral palsy and syndromic aetiology | 1 (4.5) |
| Other | 10 (45.5) |
| Epilepsy |               |
| Seizures controlled by medication | 11 (50) |
| Seizures once a month | 2 (9) |
| Seizures once a week | 2 (9) |
| Seizures once a day | 1 (4.5) |
| Several seizures a day | 1 (4.5) |
| No epilepsy | 5 (23) |
| Visual impairment |               |
| Confirmed | 17 (77.5) |
| Presumed | 1 (4.5) |
| No visual impairment | 4 (18) |
| Auditive impairment |               |
| Confirmed | 1 (4.5) |
| Presumed | 2 (9) |
| No auditive impairment | 18 (82) |
| Missing | 1 (4.5) |
| Tactile impairment |               |
| Confirmed | 7 (32) |
| Presumed | 6 (27) |
| No tactile impairment | 9 (41) |
| Vulnerability to hypernausea reflex* |               |
| Often | 9 (41) |
| Sometimes | 12 (54.5) |
| Rare/not observed | - |
| Missing | 1 (4.5) |
| Vulnerability to gastroesophageal reflux |               |
| Often | 3 (14) |
|Sometimes | 17 (77) |
| Rare/not observed | 1 (4.5) |
| Missing | 1 (4.5) |
| Feeding mode |               |
| Blended food | 8 (37) |
| Chunky food | 3 (13) |
| Enteral feeding (partial) | 10 (45.5) |
| Enteral feeding (complete) | 1 (4.5) |
| Medication |               |
| Medicated | 21 (95.5) |
| Non-medicated | 1 (4.5) |

(Continues)
least one of the two parents, including eight Swiss children, which suggests a partially common ‘olfactory niche’ (Bensafi & Rouby, 2021). As we know that prior experience can affect olfactory responses (Martinec Nováková et al., 2018), the participants’ parents or legal guardians were asked to rate the familiarity of each odour on a binary scale (0 = unfamiliar or not familiar at all; 1 = somewhat familiar or very familiar). Unfortunately, we only received the responses of 15 families out of 22. Given this low response rate, we decided not to use this information as a moderator in the analyses. Odours were dispensed in Burghart pen-like devices 14 cm long and 1.3 cm diameter. These devices were chosen for their very hermetic closing mode which limits ambient olfactory ‘contamination’ and makes them reusable. During the odour presentation, a protective tip was added to the head of the stick in order to prevent the child’s skin or eye from coming into contact with the soaked wick.

2.4 | Measures

The Mood, Interest and Pleasure Questionnaire (MIPQ, Ross & Oliver, 2003; Petry et al., 2010) is a 23-item Likert questionnaire with three subscales: positive mood (nine items, max. 36 pts), negative mood (seven items, max. 28 pts) and interest and pleasure scale (seven items, max. 28 pts). Internal consistency of the French version (Verbel Sierra, 2013) is good (Cronbach’s alpha = .97 for the total scale; Pearson r = .90 for interrater reliability). The total score of the test is a maximum of 92 points. High scores denote high mood and interest and pleasure levels.

The gastroesophageal reflux disease observation form is a 19-item Likert questionnaire (Senez, 2013a). Items are scored on a 3-point scale (no problem; some problems; significant problems). The total score ranges between 19 (no signs) and 57 (all signs present). The Nausea responses observation form is a 10-item Likert questionnaire (Senez, 2013b). Items are scored on a 3-point scale (often; sometimes; never). The total score ranges between 1 (no signs) and 30 (all signs present). These two questionnaires are clinically recognised by speech therapists in France, but have never been scientifically validated.

2.5 | Procedure

The experiment was performed in the participants’ schools. Rooms were chosen for their quietness and good ventilation. All participants were sitting in their own seats during the presentation of the odours. Postural adaptations (such as headrest or support behind the neck if needed by participants to allow maximum mobility) were discussed with the children’s physio- or occupational therapists and tested before the experiment. A tripod with an articulated arm and a clip was used to bring the stimulus close to the participant’s nose. The device was used to prevent the researcher from bringing his or her arm or hand too close to the child’s face, which might have prompted the child to respond to the speaker’s proximity, rather than to the odour. The device also aims to prevent the adult’s hand from hiding the child’s face and to prevent olfactory interference with the researcher’s body odours.

Three people (two researchers and a direct support person) were present in the room during the sessions. Researcher 1 stood to the right or left of the child while presenting the stimuli. His or her position was switched from one session to another to avoid participant orientation bias. Cotton gloves were used to handle the sticks. Researcher 2 was in charge of interrupting the session if the participant showed any reaction of discomfort, according to the ethical commitments made by the researchers. They were also systematically asked if the child was in a stable behavioural and/or health condition for the experiment. When presenting the stimuli, Researcher 1 opened the stick, fixed it on the clip, presented it about 2 cm from the participant’s nose, between nose and chin (cf. Lima et al., 2011). To reduce the risk of fatigue and olfactory adaptation (i.e., temporary decrease in olfactory sensitivity following stimulation, Köster & de Wijk, 1991), the experiment was conducted in three sessions spaced about 1 week apart, during which a randomised sequence of six odorants and six neutral stimuli were presented. Odorants were presented successively in a randomised order (Hasard® software) for 15 s. In order to enable sufficient recovery of the sense of smell, the inter-odorant neutral stimulus duration was 30 s, during which only the last 15 s were coded, except for two indicators to observe the participant’s reaction 2 and 5 s, respectively, after the introduction of the stimulus. No words were addressed to the participant during the sequence. The total assessment time: 18 odorous stimuli * 15 s and 18 odourless stimuli * 30 s, was 13.5 min per participant.

The sessions were videotaped by two digital camcorders placed in front of the participant. One was focused on the participant’s face, the other on his or her body. See Petitpierre & Dind (under review) for a detailed presentation of the procedure.

2.6 | Coding

Twenty-one behavioural indicators were used to code the behaviours exhibited by the participants, of which 14 were coded on duration (milliseconds) and seven on occurrences. The grid was a two-level one. It specifies the operational definition of the indicator as well as behaviours that should not be coded (first level). The operational definition was sometimes accompanied by a video illustrating both the targeted or excluded behaviours. If necessary, it provides additional information for participants who express themselves in an idiosyncratic way (second level). The grid is presented in Appendix 1. We chose to code some behaviours on duration and others on occurrences and based our decision on methodological knowledge (i.e., Thompson et al., 2000), on newborns and young infants olfaction research (Schaal et al., 1998, 2002), and on the results of the pilot study. Two criteria have been considered: (1) the relevance and (2) the simplicity of the method of measurement for the target behaviour. Occurrence recording involves recording the number of times the target behaviour is observed to occur during a pre-defined observation period. It makes sense for shortly manifested
behaviours (‘sniffing’, ‘yawning’, ‘pouting’, ‘nausea reaction’, ‘rejecting the odorant’, ‘moving the head towards the stick’, etc.), but is not useful for behaviours that are not easy to count (e.g., maintaining a posture, in our case ‘head alignment on the stick’; ‘showing stereotypies’ and ‘being physically active’; ‘holds the stick by coordinating hand, nose, etc.’), nor for behaviours that occur very infrequently (e.g., ‘emotional outburst’). Duration recording involves recording the time that elapses between the moment when the target behaviour begins and when it ends. It makes sense for actions that involve a number of steps (e.g., in our case ‘exploring with one’s mouth’) or when the frequency of the behaviour is very rare (e.g. ‘emotional outburst’). For simplicity, we have also coded the duration for behaviours that the participant exhibits repeatedly, as long as the interval between two manifestations of the behaviour is not greater than 1 s (e.g., ‘positive/negative non-interpretable vocalisations’, ‘grinding’, ‘chewing’, ‘moving one’s lips’). The most difficult decision concerned the coding of smiles. Finally, we chose to measure the duration of the smile, which allows us to make statements about the percentage of time a person spends smiling while exposed to the odour or the neutral stimulus. In any case, either occurrence or duration recording requires very precise behavioural definitions; that is why we have included temporal specifications in the operational description of some of our behaviours. For the analyses, only raw values (occurrences and durations) were used.

The coding was carried out by a team of three coders. Two of them have a doctoral degree in special education, the third a MA degree. All coders were experienced in video analysis, two of them with the target group. Two coders were also experimenters. During the experiment, the stimuli and the neutral stimulus were designated by a number. The coders who were not experimenters were blind to the nature of the odorant, except for the neutral stimulus, whose more frequent presentation could be a potential cue. When the coder was one of the experimenters, we cannot exclude that he or she remembered the association between the number and the type of odorant. The coding was done both in continuous and real-time measurement. The coding procedure was set up in the EUDICO Linguistic Annotator (ELAN) Software version 5.9 (Max Planck Institute for Psycholinguistics, Nijmegen, The Netherlands). A refined reliability assessment procedure using EasyDIAg algorithm (Holle & Rein, 2015) was used to check both the observers’ agreement about presence/absence of a behaviour and the temporal overlap of their coding. The reliability value (kappa) was calculated on 20% of the data. The accuracy of the grid makes it possible to reach a satisfactory interobserver agreement for 13 indicators: head alignment on the stick ($k = .83$); sniffing ($k = .55$); chewing ($k = .89$); moving the lips or tongue ($k = .62$); smiling ($k = .72$); pouting ($k = .77$); yawning ($k = 1.00$); moving the hand or head towards the stick ($k = .81$); rejecting the stick ($k = .85$); being

| Stimuli                  | Food/non-food | Hedonicity | Perceived intensity | Familiaritya | Classificationb |
|-------------------------|--------------|------------|---------------------|-------------|----------------|
|                         |              |            | M (min.–max.) | SD  | M  | SD  |
| Orange (Burghart)       | F            | P          | 5.00 (2–6) | 0.92 | 0.48 | 0.48 | C1 |
| Nutella                 | F            | P          | 5.82 (4–7) | 0.61 | 0.48 | 0.51 | C7 |
| Cinnamon                | F            | P          | 6.10 (5–9) | 0.93 | 0.29 | 0.46 | C7 |
| Strawberry              | F            | P          | 6.00 (4–7) | 0.80 | 0.62 | 0.49 | C5 |
| Cheese                  | F            | U          | 5.07 (2–8) | 1.33 | 0.81 | 0.40 | C8 |
| Garlic (Burghart)       | F            | U          | 6.45 (5–9) | 0.91 | 0.38 | 0.49 | C8 |
| Green vegetables        | F            | N          | 5.85 (3–9) | 1.13 | 0.86 | 0.35 | C6 |
| Rancid butter           | F            | N          | 5.52 (3–8) | 1.09 | 0.05 | 0.22 | C8 |
| Apple pie               | F            | N          | 5.90 (4–8) | 0.90 | 0.48 | 0.51 | C7 |
| Basil                   | F            | N          | 6.04 (4–8) | 0.92 | 0.38 | 0.49 | C4 |
| Lily of the valley      | NF           | P          | 5.36 (4–9) | 1.02 | 0.13 | 0.35 | C4 |
| Summer rain             | NF           | P          | 3.59 (2–5) | 0.93 | 0.48 | 0.51 | C6 |
| Rose (Burghart)         | NF           | P          | 5.68 (4–8) | 0.81 | 0.20 | 0.41 | C4 |
| Hand sanitizer (Steriliumb) | NF   | U          | 5.21 (2–8) | 1.17 | 0.52 | 0.51 | C2 |
| Sweat                   | NF           | U          | 5.66 (2–8) | 1.44 | 0.33 | 0.48 | C8 |
| Swimming pool           | NF           | N          | 2.80 (1–6) | 1.21 | 0.70 | 0.47 | C2 |
| Pony                    | NF           | N          | 5.38 (4–7) | 0.98 | 0.14 | 0.35 | C8 |
| Grass (Burghart)        | NF           | N          | 5.62 (1–8) | 1.26 | 0.52 | 0.51 | C6 |
| Neutral                 | NF           | –          | –        | –   | –  | –  |

Abbreviations: C1, citrus, and so on; C2, disinfectants, alcohol, chemical, and so on; C3, mint, camphor, and so on (not presented); C4, flowers, plants, aromatics, and so on; C5, fruits except citrus, and so on; C6, leaves, wood, grass, and so on; C7, Honey, nuts, bakery, and so on; C8, Apple, acid, putrid rancid, garlic, and so on; N, neither pleasant, nor unpleasant; P, pleasant; U, unpleasant.

Table 2 Characteristics of stimuli.
physically active (k = .74); making positive vocalisations (k = .80); uninterpretable vocalisations (k = .44); all vocalisations (k = .68). Due to insufficient data in the random selected files, the interobserver coefficient could not be calculated for eight indicators. A total of 792 files (22 participants * 18 odorous + 18 neutrals) coded on 21 indicators results in a total of 8170 observations. The average number of observations per participant was 371.

2.7 Analyses

As the data were not normally distributed, non-parametric statistics have been used to perform the analyses. Data for odorous versus neutral, food versus non-food, pleasant versus unpleasant conditions, was analysed with Wilcoxon signed rank test (Z), alpha level at .05 (bilateral). Effect sizes for Wilcoxon were calculated according to Pallant (2020), that is, by dividing the test statistic by the square root of the number of observations. Generalised linear mixed models (t) were used to explore the role of personal characteristics on the olfactive response. All analyses were performed using SPSS software version 26.0.0 (IBM Corp.).

3 RESULTS

3.1 Response to odorous versus neutral condition

The first analysis aims to study whether participants responded differently to odour versus neutral condition. To test this hypothesis, the

| Table 3 Participants' behaviour in the odorous versus neutral condition |
|------------------------|------------------|-------------------|------------------|------------------|
| Variables              | Odorous versus neutral condition | Wilcoxon S-R test | Effect size r eta squared* |
|                        | Positive ranksA (N) | Sum of ranks | Negative ranksB (N) | Sum of ranks | TiesC (N) | Z | p bilateral | |
| Nose/head              |                      |               |                   |               |           |   |            | |
| Head alignment on the stick | 21                  | 252          | 1                 | 1             | 0         | -4.07D | .000 | .614A |
| Nose/hand on the stick | 7                    | 28           | 0                 | 0             | 15        | -2.36D | .018 | .356A |
| Sniffing               | 14                   | 131.50       | 2                 | 4.50          | 6         | -3.29D | .001 | .496A |
| Mouth                  |                      |               |                   |               |           |   |            | |
| Exploring with one's mouth | 6                   | 26.50        | 1                 | 1.50          | 15        | -2.12D | .034 | .320A |
| Grinding (teeth)       | 1                    | 1            | 1                 | 2             | 20        | -.447C | .655 |      |
| Chewing                | 11                   | 81           | 2                 | 10            | 9         | -2.48D | .013 | .374A |
| Moving one's lips/tongue | 14                  | 178          | 8                 | 75            | 0         | -1.67D | .095 |      |
| Nausea reaction        | 4                    | 10           | 0                 | 0             | 18        | -1.84D | .066 |      |
| Emotion                |                      |               |                   |               |           |   |            | |
| Smiling                | 13                   | 166          | 8                 | 65            | 1         | -1.75D | .079 |      |
| Pouting, making a face | 16                   | 176          | 3                 | 14            | 3         | -3.26D | .001 | .491A |
| Yawning                | 0                    | 0            | 7                 | 28            | 15        | -2.37D | .018 | .357B |
| Emotional outburst     | 1                    | 1            | 0                 | 0             | 21        | -1.00D | .317 |      |
| Hands/arms             |                      |               |                   |               |           |   |            | |
| Moving hand/head towards the stick | 16              | 183          | 3                 | 7             | 3         | -3.54D | .000 | .534A |
| Rejecting the stick    | 15                   | 120          | 0                 | 0             | 7         | -3.42D | .001 | .516A |
| Body                   |                      |               |                   |               |           |   |            | |
| Interrupting motor action | 4                   | 13           | 1                 | 2             | 17        | -1.49D | .136 |      |
| Being physically active | 8                    | 87           | 14                | 166           | 0         | -1.28D | .200 |      |
| Showing stereotypies   | 1                    | 1            | 1                 | 2             | 20        | -.447C | .655 |      |
| Vocalisations          |                      |               |                   |               |           |   |            | |
| Positive               | 9                    | 63           | 4                 | 28            | 9         | -1.22D | .221 |      |
| Negative               | 6                    | 42           | 8                 | 63            | 8         | -.655C | .510 |      |
| Uninterpretable        | 7                    | 55           | 11                | 116           | 4         | -1.32D | .184 |      |
| All vocalisations      | 11                   | 101          | 9                 | 109           | 2         | -1.49D | .881 |      |

Note: Bold values denote statistical significance at the p < 0.05 level; A, Odorous condition > Neutral condition; B, Odorous condition < Neutral condition; C, Based on positive ranks; D, Based on negative ranks.

*The effect size (r) was calculated from Pallant (2020).
data has been aggregated for all odorants and all neutral stimuli, respectively, and compared to one another for each indicator. The results show that participants respond significantly differently to the odour condition compared to the neutral. A difference is present in 9 of the 21 indicators (Table 3). The effect size, calculated according to Pallant (2020), is large or nearly large in four indicators. The head alignment on the stick is significantly longer in the odorous condition ($Mdn_{Odours} = 6.69$ vs. $Mdn_{Neutrals} = 4.34$ s; $Z = -4.07$, $p = .000$). The number of hand or head movements towards the stick is significantly higher in the odorous condition ($Mdn_{Odours} = .81$ vs. $Mdn_{Neutrals} = .42$ occurrences; $Z = -3.54$, $p = .000$), as well as the number of stick rejection behaviours ($Mdn_{Odours} = .06$ vs. $Mdn_{Neutrals} = 0$ occurrences; $Z = 3.42$, $p = .001$) and the number of sniffing behaviours ($Mdn_{Odours} = .17$ vs. $Mdn_{Neutrals} = 0$ occurrences; $Z = -3.29$, $p = .001$). The effect size is nearly large for pouting with four times more pouts in the odorous condition compared to the neutral ($Mdn_{Odours} = .22$ vs. $Mdn_{Neutrals} = .055$ occurrences; $Z = -3.26$, $p = .001$) and moderate for nose/hand coordination on the stick ($Z = 2.36$, $p = .018$), chewing ($Z = -2.48$, $p = .013$), exploring the stimulus with the mouth ($Z = -2.12$, $p = .034$), which appear significantly longer in the odorant condition than in the neutral condition. Conversely, the number of yawning occurrences is higher in the neutral condition ($Z = -2.37$, $p = .018$).

### 3.2 Response to food versus non-food odorants

The second analysis aims to study whether participants responded differently to food versus non-food odorants. Results indicate that participants sniff the edible odorants more than the inedible ones ($Z = -2.46$, $p = .014$, $r = .37$) and they coordinate their nose and hand on the stick more often ($Z = -2.19$, $p = .028$, $r = .33$). Chewing is only tendential ($Z = -1.78$, $p = .075$, $r = .27$).

With regard to food versus neutral stimuli, a significant effect was observed in 9 of the 21 indicators (see online Supplement 1 for details). The effect size was large ($r > .50$) for one indicator (head alignment on the stick). It was moderate ($r$ between .30 and .49) for six (nose/hand
coordination on the stick; chewing; pouting; moving hand or head towards the stick; rejecting the stick; sniffing) which prevail in the edible odorant condition versus neutral while two (yawning; being physically active) prevail in the neutral compared to the edible. The effect of non-food odorants versus neutrals was observed in 8 indicators out of 21 (see online Supplement 2 for details). Two indicators (head alignment on the stick; moving hand or head towards the stick) show large effect sizes ($r > .50$). Effect sizes were moderate ($r$ between .30 and .49) for the other six (nose/hand coordination on the stick; exploring the stimulus with the mouth; pouting; rejecting the stick; smiling; sniffing), which were all more prevalent in the presence of the odorant.

| TABLE 4 | Significantly manifested behaviours according to the type of stimuli |
|---------|---------------------------------------------------------------|
|         | All stimuli Scented versus neutral | Food versus non-food | Pleasant versus unpleasant |
|         |                              | Food versus neutral | Non-food versus neutral | Food versus non-food | Pleasant versus neutral | Unpleasant versus neutral | Pleasant versus unpleasant |
| Nose/head |                               |                     |                         |                      |                     |                           |                           |
| Head alignment on the stick | S > N | F > N | NF > N | P > N | u > n |
| Nose/hand on the stick | s > n | f > n | nf > n | f > nf |       |
| Sniffing | S > N | f > n | nf > n | f > nf | p > n |
| Mouth |                     |                         |                     |                      |                     |                           |                           |
| Exploring with one’s mouth | s > n |               | nf > n |           |               |                           |                           |
| Grinding (teeth) |                 |                     |               | p > u |
| Chewing | s > n | f > n |       |           |               |                           |                           |
| Moving one’s lips/tongue |         |               |       |               | u > n |
| Nausea reaction |           |               |       |               |           |
| Emotion |                     |                         |                     |                      |                     |                           |                           |
| Smiling |                     |                         | nf > n |           |               |                           |                           |
| Pouting, making a face | s > n | f > n | nf > n |           | U > N | P > U |
| Yawning |           |               | n > s | n > f |               |                           |                           |
| Emotional outburst |           |               |           |       |               |                           |                           |
| Hands/arms |                     |                         |                     |                      |                     |                           |                           |
| Moving hand/head towards stick | S > N | f > n | NF > N |       | u > n |
| Rejecting the stick | S > N | f > n | nf > n |       | u > n |
| Body |                     |                         |                     |                      |                     |                           |                           |
| Interrupting motor action |               |               |       |               |           |
| Being physically active |           |               | n > f |           |               |                           |                           |
| Showing stereotypes |           |               |       |               |           |
| Vocalisations |                     |                         |                     |                      |                     |                           |                           |
| Positive |                     |                         |               |           |               |                           |                           |
| Negative |                     |                         |       | n > p |               |                           |                           |
| Uninterpretable |         |               |       |               | All vocalisations | p > u |

Note: Capital letters indicate a large effect size (EF > .05). Lower case letters indicate a moderate effect size (0.3 > EF > .05).

Abbreviations: F or f, food; N or n, neutral stimuli; NF or nf, non-food; P or p, pleasant; S or s, scented stimuli; U or u, unpleasant.
3.3 | Response to odors a priori rated as pleasant or unpleasant by typical adults

A comparative analysis was carried on four pleasant (Nutella, lily of the valley, rose and strawberry) versus four unpleasant (garlic, sweat, hand sanitizer and cheese) stimuli. Since pleasant odours that are too strong can become unpleasant and vice versa, it was essential to ensure that, for this analysis, the two categories of stimuli had a rather similar average intensity. The latter amounts to 5.71 for stimuli rated as pleasant and 5.59 for those rated as unpleasant, respectively. A stimulus pleasantness effect was confirmed in 3 of the 21 indicators. Participants made significantly more pouts when they were exposed to stimuli rated as unpleasant ($Z = -3.54$, $p = .000$), the effect size being large ($r = .53$). In contrast, they spent more time exploring the stimuli with their mouth ($Z = -2.26$, $p = .024$), and vocalising ($Z = -2.16$, $p = .038$), in the pleasant condition, the effect sizes being moderate ($r = .34$ and $r = .33$, respectively).

Significant effects of the four aforementioned pleasant stimuli versus neutrals were observed in 4 of the 21 indicators (see online Supplement 3 for details). The effect size was large ($r > .50$) for one indicator (head alignment on the stick) and moderate ($r$ between .30 and .49) for three others (smiling; sniffing; negative vocalisations). Head alignment, smiling and sniffing prevailed in the pleasant condition compared to the neutral one; conversely negative vocalisations prevailed in the neutral condition. The effect of unpleasant versus neutrals was observed in 5 indicators out of 21 (see online Supplement 4 for details). One indicator (pouting) shows a large effect size ($r > .50$). Effect sizes were moderate ($r$ between .30 and .49) for the four other indicators (head alignment on the stick; moving the lips or tongue; moving the hand or head towards the stick; rejecting the stick).

3.4 | Influence of moderators on the olfactory response

The influence of moderators like gender, age, aetiology, presence of epilepsy, feeding mode, vulnerability to nausea, as well as the link with MIPQ scores, was explored. The analyses were performed on the nine behaviours significantly highlighted in the first analysis. No effect of age, aetiology, presence of epilepsy, feeding mode, vulnerability to nausea, was found. In contrast, a simple gender effect was found on the alignment of the head on the stick, since girls stay aligned longer both in the odorous and in the neutral condition, but both boys and girls stay longer on the odorous sticks than on the neutrals ($t = -2.72$, $p = .013$). Feeding mode moderates the participants’ frequency of head or hand movements towards the stick ($t = -2.50$; $p = .021$). Participants eating blended or chunky food make more movements towards the sticks than participants who are fed with a tube, either partially or totally. However, all participants, no matter how they eat, make more movements towards the odorous sticks compared to the neutrals (Figure 1a). Participants assessed with a high positive mood score make more movements towards the sticks (Figure 1c) than participants who exhibit low positive mood ($t = -2.14$, $p = .045$). On the contrary, participants assessed with a high positive mood score (Figure 1d) or level of interest (Figure 1b) exhibit shorter head alignment on the stick behaviours than those reported with lower positive mood ($t = 2.82$, $p = .010$) and interest level ($t = 5.02$, $p = .000$). However, all align the head longer on the odorous stimuli compared to the neutrals.

4 | DISCUSSION

Despite the importance of olfaction in human functioning, only very few studies have explored the olfactory behavioural responses of people with PIMD. The scope of this research was to systematically observe how children and young people respond to odours and whether certain factors were likely to influence their response. The overall results show that participants respond significantly differently to odorous compared to odorless stimuli. They also show significant differences according to the types of stimuli, whether food or non-food, pleasant or unpleasant. Table 4 summarises their responses under the various conditions.

In children and young people with PIMD, the reaction to odours is mainly manifested by the alignment of the head on the odour, sniffing, pouting, nose/hand coordination on the stick, moving the hand or head towards the stick and rejecting the stick. When the scent is presented to them, the participants significantly align their head on the stick, whether it is food, non-food, pleasant or even unpleasant odours. Odorous stimuli, except those rated as unpleasant by typical adults, have an effect on the number of sniffs, which are more numerous in the odorant condition than in the neutral one. To a lesser extent, exploring with the mouth, chewing and moving the lips or tongue, show that the two conditions are approached differently. Smiling is significantly more manifested in the presence of non-food stimuli, as well as with stimuli rated as pleasant. On the contrary, pouting appears more often in the presence of unpleasant stimuli. Pouting also differentiates between odours rated by typical adults as pleasant and unpleasant. Yawning appears significantly more often in neutral conditions, which may suggest boredom in the absence of interesting events. Being physically active and making uninterpretable vocalisations, significantly more manifested during neutral conditions, can likewise be seen as a means to react to the vacuum of stimulation or to the disappearance of the interesting stimulus. Results show a parallel between movements to bring the stimulus closer or further away. Both occur in the presence of various stimuli, including those rated as unpleasant. These oriented movements suggest that the person identifies the location of the stimulus and, perhaps seeks to act actively on it. The nose/hand coordination on the stick is an attempt to explore it by stabilising both the head and the odorous source. Behaviours are stable and consistent between and within the various odorous conditions.

The results, showing behavioural differences between pleasant versus unpleasant, food versus non-food groups of stimuli, imply the possible existence of an early implicit categorisation ability in the participants. Along with discrimination, categorisation is the second basic process investigated in infant olfactory perception (Schaal et al., 2002). Discrimination is the ability to respond selectively to
specific individual odours, while categorisation means to ‘group discriminable properties, objects, or events into classes (...)’ (Rakison & Lawson, 2013, p. 4) and to respond to groups of odours. A large number of studies highlight the possibility that categorical abilities exist already at the very onset of development and may be expressed ‘in concept-like behaviours’ (Alessandroni & Rodríguez, 2020, p. 2). In the present research, the setting was not specifically designed to observe the participants’ categorical abilities and it would be unsuitable to overreach the findings in terms of mechanisms underlying the behaviours observed; even so, given the clear differential reactions between both groups of stimuli, and by analogy with the developmental data, the presence of prelinguistic categorisation abilities cannot be excluded.

With regard to whether moderating variables affect responses, we find girls align the head longer on the stimuli. Head alignment however significantly reflects the ability to detect odorous versus neutral stimuli in both genders. It was also interesting to observe that attempts to get closer to the stick are more numerous in the odorous compared to neutral conditions and that capture attempts differ according to the participant’s feeding mode. Several explanations are possible regarding this result. The meal is a situation with a great wealth of odours in which children are often encouraged to grasp (the food or the utensils). Compared to neutral stimuli, it is possible that our odorants, which included a lot of food odours, generated reaching behaviours by analogy to what happens during mealtimes. We can also assume that, even if they do not handle the spoon themselves, spoon-fed children have a more sustained experience of contact with food than enteral-fed children. However, most children, enteral-fed children included, have an experience with spoon-feeding. This could explain why the odorous condition elicited more reactions than the neutral, though to a lesser extent in children who were artificially fed at the time of the study. We were surprised to find that participants’ head alignment duration was inversely related to the interest and the positive mood sub-scores, respectively. A positive relationship between the two dimensions would have been more logical. On the other hand, our results are based on direct observations, while the MIPQ is a reported retrospective measure; the nature of the data therefore differ considerably. In addition, positive mood behavioural manifestations, as well as interest ones, may be more or less salient from one person to another. It is possible that some children express their interest in a very discreet way, by simply positioning themselves in front of the source of a stimulation, while others express themselves through more easily observable behaviours. The difference in procedure sensitivity may explain this result, which however does not challenge the fact that the two groups express a clear difference between the two stimulation conditions, regardless of their level of interest or positive mood.

This research is not free from limitations. It focuses on odour detection using only a single concentration, which is a rather simplified way to observe human olfactory functioning. Prior olfactory experience should have been controlled better. Finally, group analyses are reductive and do not provide access to individual clinical realities. We observed, for instance, a nauseous response in one participant. This response occurs twice while presenting the same single odorant (strawberry) 2 weeks apart. An anamnesis revealed that this participant, fed by gastric tube at the time of the study, had taken strawberry-odorous food supplements a few years before. Her reaction suggests the integrity of an olfactory memory, which is very interesting from a clinical point of view.

However, the study has the merit of drawing attention to a neglected sensory modality in people with PIMD. On the research level, many questions remain open. The function of odours for this population should be further investigated, for example whether some odorants are calming, whether they provide a feeling of security or, on the contrary, evoke an unpleasant experience as in the anecdotal observation reported. The influence of medication should also be studied because some drugs act on olfaction. Studying children’s and young people’s reactions to familiar versus unfamiliar people’s odours, or to their own odour versus that of peers, could be interesting in order to provide access to their self- and other awareness capacities. On the educational level, the results support recommendations to use odorants with this population and provide guidelines to observe and interpret their responses.

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DATA AVAILABILITY STATEMENT
Data available on request due to privacy/ethical restrictions.

ORCID
Geneviève Petitpierre https://orcid.org/0000-0002-3353-897X
Juliane Dind https://orcid.org/0000-0002-8378-5944

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SUPPORTING INFORMATION

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