Responsiveness of children with PIMD:

Two New Lines in the Study of Sensory Stimulation

Mariely Gestosa Lima

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RESPONSIVENESS OF CHILDREN WITH PIMD: TWO NEW LINES IN THE STUDY OF SENSORY STIMULATION

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Orientadora - Doutora Liliana de Sousa
Professora Associada
Instituto de Ciências Biomédicas Abel Salazar

Co-orientadora - Doutora Isabel Amaral
Professora Coordenadora
Escola Superior de Saúde do Instituto Politécnico de Setúbal
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“You can come to know us, and about our life satisfaction. (The term doesn’t make much sense, but quality of life and happiness do!) It’s mostly about doing fun things with the people that we like. It’s the same for us as it is for other children, but we show how we think and feel in very personal ways. Different people see us in different ways. It depends on how they understand our disabilities. People who don’t know us usually only see our disabilities, and not how we are feeling, learning and growing. We are individuals in what we do and how we feel. We are children and we have a future, but we live mostly in the here and now. A good life is having happiness and contentment each day, although contentment can be as simple as just taking it all in. It’s about being in balance. A good life is also about comfort and wellbeing. Many of us have lots of pain, so our health is very important. Just having a good day is great. Being friends is most important. It’s about belonging. A good life is also about our favorite things. This can be just caring and sharing, or doing special things with special people, or playing in water, or just joking around!

It can be hard to come to know us, and this is hard to explain. It’s the same as with other children, but takes longer. Eventually the penny will drop for you! To come to know us, you need to do three things at the same time. First, you need to watch and listen for how we show our feelings, for ‘patterns’ in what we do every day, and for changes in these patterns. You need to be able to look back to learn to understand the little things we do. Second, you need to spend time being with us and playing with us. You need to do the usual, simple things with us, and then put yourself in our shoes. What you do will probably be just trial and error, but you can experiment on purpose. Third, you need to talk to the people who know us, and ask the right questions, and work together with others. This takes some planning. How well you come to know us depends upon how well you know yourself. Your time with other children can help, and how you feel about us and what you presume about us is important. For some people this is hard, and for others it’s easy. Some people have just got it!”
Para vocês,
Príncipes e Princesas
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“O amor não pode estar muito longe de um coração grato e de uma mente agradecida…”.

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E por isso ser-vos-ei … Para sempre grata
LISTA DE MANUSCRITOS RESULTANTES DA TESE

Artigo experimental aceite para publicação em revista com arbitragem científica:


Artigos experimentais submetidos para publicação em revistas com arbitragem científica:


Artigo de opinião submetido para publicação em revista com arbitragem científica:

ABSTRACT

The quality of the sensory stimulation that is provided to individuals with profound intellectual and multiple disabilities (PIMD) has recently emerged as a major topic of investigation as a lack of, or inappropriate, stimulation is detrimental to their, already compromised, health and emotional well-being. Studies have been stressing the need for highly individualized sensory interventions that take into consideration each person’s responsiveness profile. To date, however, the methodologies that are most commonly used with individuals with PIMD to assess their reactions to sensory stimuli, as well as their potential appraisals of such stimuli, are mostly limited to behavioural observations.

The aims of the present thesis were i) to explore the combined use of behavioural and physiological measurements to capture objectively the actual responsiveness of four children with PIMD and infer about the hedonic value of the stimuli that are presented to them, and ii) to analyse the responsiveness of two children with PIMD to a certified dog, as a potential promoter of emotional well-being, and set a preliminary basis for future studies to explore the benefits of including such a multisensory live stimuli in a number of interventions.

This thesis includes three experimental studies and one personal view. A pilot study aimed at accurately describing the motor and physiological reactions of one child with PIMD to a set of stimuli commonly used during sensory interventions. The participant only exhibited consistent motor reactions to three out of the nineteen presented items, but showed consistent physiological reactions to a total of ten stimuli. Although exploratory, obtained results pointed to the importance of considering and combining different approaches to study the actual sensory responsiveness of individuals with PIMD. With the aim of further extending these findings, a second study was conducted, using a similar, although slightly modified, methodological approach. Results obtained seemed to corroborate the data from the pilot study in that all the three children that participated in this second study showed frequent consistent physiological reactions but only rarely exhibited consistent behavioural ones. Moreover, physiological data obtained pointed to the possibility of different appraisals of the presented sensory stimuli (namely in terms of novelty and pleasantness), even when no consistent behavioural reactions were observed.

The third experimental study aimed at quantifying the behavioural and physiological responses of two children with PIMD to a certified dog. As to better evidence the potential benefits of the animal for those children, their responsiveness to a familiar caregiver and to an artificial multisensory stimulus (specifically designed for sensory interventions) was
also assessed. Interestingly, both participants appeared to respond positively to the dog (in much a same way as they responded to the familiar caregiver), which appeared as a relaxing and pleasant stimulus with which participants seemed to engage. Prompted by these results, a personal view is also presented in this thesis as to stress the need for future studies to explore the potential of using certified dogs as stimuli in interventions aimed at training functional communicative skills in individuals with PIMD.

Overall, the studies presented in this thesis, although preliminary, may have important implications for the quality of support of individuals with PIMD, namely through an accurate selection of sensory items, and, thus, for their emotional well-being.
RESUMO

A qualidade da estimulação sensorial proporcionada a indivíduos com diagnóstico de multifeticiência emergiu, recentemente, como um importante tópico de investigação, na medida em que a falta ou a inadequação de informação sensorial pode comprometer o estado de saúde, já fragilizado, destes indivíduos, bem como o seu bem-estar emocional. Estudos realizados têm vindo a enfatizar a necessidade de implementar intervenções sensoriais individualizadas que contemplem o perfil de reactividade de cada indivíduo. No entanto, até à data, as metodologias mais frequentemente utilizadas na avaliação da reactividade sensorial dos indivíduos com multifeticiência, bem como da apreciação que estes podem fazer dos estímulos sensoriais, têm-se limitado a observações comportamentais.

Os objectivos desta tese foram: i) explorar a combinação do uso de medidas comportamentais e fisiológicas como forma de avaliar, de uma forma abrangente, a reactividade de quatro crianças com multifeticiência, e inferir sobre o valor hedónico dos estímulos sensoriais que lhes são apresentados e ii) analisar a reactividade de duas crianças com multifeticiência a um cão certificado, como primeiro passo para o estudo dos efeitos da presença deste tipo de estímulo, vivo e multisensorial, nas intervenções terapêuticas.

A tese aqui apresentada inclui três artigos experimentais e um artigo de opinião. Um primeiro estudo (piloto) teve como objectivo descrever as reacções motoras e fisiológicas de uma criança com multifeticiência a um conjunto de estímulos frequentemente utilizados nas intervenções sensoriais. O participante apenas exibiu reacções motoras consistentes a três dos dezenove estímulos apresentados, embora tenha manifestado reacções fisiológicas consistentes perante a apresentação de dez estímulos. Os resultados obtidos, embora exploratórios, parecem indicar a importância de considerar a combinação da análise comportamental e fisiológica como forma de estudar a reactividade sensorial de indivíduos com multifeticiência. Com o objectivo de corroborar estes resultados foi conduzido um segundo estudo, que seguiu uma metodologia similar, embora com pequenas alterações. Os resultados obtidos parecem confirmar aqueles apresentados no estudo piloto, na medida em que os três participantes mostraram um maior número de reacções fisiológicas consistentes quando comparadas com as reacções comportamentais. Além disso, os dados fisiológicos obtidos apontam para a possibilidade destes indivíduos realizarem distintas apreciações dos estímulos.
apresentados, nomeadamente em termos de novidade e de prazer, mesmo na ausência de reacções comportamentais consistentes.

O terceiro estudo experimental apresentado nesta tese teve como objectivo quantificar as respostas comportamentais e fisiológicas de duas crianças com multifalência perante a apresentação de um cão certificado. De forma a tornar evidente os potenciais benefícios que podem advir da presença de um animal junto de indivíduos com este diagnóstico, foi igualmente avaliada a reactividade dos participantes a um cuidador familiar e a um estímulo artificial multisensorial (especificamente desenhado para as intervenções sensoriais). Curiosamente, ambos os participantes pareceram responder positivamente à presença do cão, tal como à figura do cuidador, pelo que o animal surge neste estudo como um estímulo aparentemente relaxante e agradável. Por fim, foi elaborado um artigo de opinião com o objectivo de realçar a necessidade de estudos futuros explorarem os benefícios que podem advir da utilização de cães certificados como estímulos em intervenções terapêuticas, nomeadamente nas que se destinam a promover comunicação funcional em indivíduos com multifalência.

De modo geral, os estudos apresentados nesta tese, embora preliminares, poderão ter implicações importantes para a qualidade do suporte oferecido a indivíduos com multifalência, nomeadamente através de uma escolha fundamentada de estímulos sensoriais, e como tal, para a garantia do seu bem-estar emocional.
RÉSUMÉ

La qualité de la stimulation qui est délivrée aux individus polyhandicapés a récemment émergé comme un important thème de recherche par le fait que le manque de stimulation, ou une stimulation inappropriée, est préjudiciable pour la santé et le bien-être émotionnel déjà compromis de ces individus. Diverses études ont souligné le besoin d'interventions sensorielles individualisées qui prennent en considération le profil de réactivité de chaque personne. A ce jour, cependant, les méthodologies les plus utilisées avec les personnes polyhandicapées pour estimer leurs réactions aux stimuli sensoriels se limitent essentiellement à des observations comportementales.

Les objectifs de cette thèse étaient i) d'explorer l'utilisation combinée de mesures comportementales et physiologiques pour estimer objectivement la réelle réactivité de quatre enfants polyhandicapés, et pour inférer de la valeur hédonique des stimuli qui leur sont présentés, et ii) d'analyser la réactivité de deux enfants polyhandicapés à un chien certifié, comme promoteur potentiel de bien-être, et établir une base préliminaire pour que des études futures explorent les possibles bienfaits qui peuvent être associés à l'inclusion de ce stimulus multisensoriel dans de nombreuses interventions.

Cette thèse comprend trois études expérimentales et un article d'opinion. La première étude (pilote) visait une description exacte des réactions motrices et physiologiques d'un enfant polyhandicapé à un ensemble de stimuli qui sont couramment utilisés pendant des interventions sensorielles. Le participant a montré des réactions motrices consistent à trois des dix-neuf stimuli présentés mais a montré des réactions physiologiques consistent à un total de dix objets. Bien que préliminaires, les résultats obtenus pointent vers l'importance de considérer la combinaison de différentes approches pour étudier la réactivité sensorielle des individus polyhandicapés. Avec l'objectif d'étendre ces résultats, une deuxième étude a été conduite, suivant une méthodologie similaire, en intégrant certaines modifications. Les résultats obtenus semblent corroborer les données de l'étude pilote par le fait que les trois enfants participant à cette deuxième étude ont montré de fréquentes réactions physiologiques consistantes mais seulement quelques rares réactions comportementales persistantes. Par ailleurs, les données physiologiques obtenues pointent vers la possibilité de différentes évaluations des stimuli présentés (notamment en termes de nouveauté et enjouement) même dans l’absence de réactions comportementales reproductibles.

Une troisième étude expérimentale est présentée qui visait quantifier les réponses comportementales et physiologiques de deux enfants polyhandicapées à un chien certifié. De façon à mieux mettre en évidence les bienfaits potentiels de l’animal pour ces enfants,
leur responsivité à un soignant familier et à un stimulus artificiel multisensoriel (conçu spécifiquement pour les interventions sensorielles) a aussi été analysée. De façon intéressante, les deux participants ont, apparemment, répondu positivement au chien (de la même façon qu'ils ont réagi au soignant familier), qui a paru un stimulus relaxant et engageant. Un article d'opinion est aussi ici présenté qui vise à souligner le besoin d'études qui explorent le potentiel d'utiliser, comme stimuli, des chiens certifiés pendant des interventions ayant pour but la définition de compétences communicatives chez des individus polyhandicapés.

Globalement, les études présentées dans cette thèse, bien que préliminaires, peuvent avoir d'importantes implications pour la qualité du support des personnes polyhandicapées, notamment par une rigoureuse sélection de stimuli sensoriels, et, donc, pour leur bien-être émotionnel.
Chapter 2

1 List of the sensory stimuli presented to the participant.
2 Motor movements exhibited by the participant and corresponding descriptions.
3 Consistent motor and physiological reactions exhibited by the participant during and following stimuli presentation. The changes presented refer to mean differences from baseline.

Chapter 3

1 List of the stimuli presented to the participants.
2 Behaviours exhibited by the participants and corresponding descriptions.
3 Consistent reactions exhibited by participant 1 including i) behavioural reactions [mean change in frequency (± SD); mean change in duration (± SD)], ii) EDRs [mean amplitude (± SD) and mean latency (± SD)], and iii) biphasic changes in HR [mean change (± SD)]. All changes refer to mean differences from baseline.
4 Consistent reactions exhibited by participant 2 including i) behavioural reactions [mean change in frequency (± SD); mean change in duration (± SD)], ii) EDRs [mean amplitude (± SD) and mean latency (± SD)], and iii) biphasic changes in HR [mean change (± SD)]. All changes refer to mean differences from baseline.
5 Consistent reactions exhibited by participant 3 including i) behavioural reactions [mean change in frequency (± SD); mean change in duration (± SD)], ii) EDRs [mean amplitude (± SD) and mean latency (± SD)], and iii) biphasic changes in HR [mean change (± SD)]. All changes refer to mean differences from baseline.

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3 Mean values of (A) HR and (B) SCL exhibited by participant 1 during baseline and period 2 of each experimental condition. Significant differences between baseline and period 2 are marked with an asterisk (*). Error bars represent standard errors.

4 Mean durations of (A) body rocking, (B) frowning, and (C) moaning behaviours exhibited by participant 2 during baseline and period 2 of each experimental condition. Significant differences between baseline and period 2 are marked with an asterisk (*). Error bars represent standard errors.

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6 Mean values of (A) HR and (B) SCL exhibited by participant 2 during baseline and period 2 of each experimental condition. Significant differences between baseline and period 2 are marked with an asterisk (*). Error bars represent standard errors.
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1. Individuals with profound intellectual and multiple disabilities (PIMD)

1.1. Characterization

Individuals with profound intellectual and multiple disabilities (PIMD) are characterized, according to the working definition developed by the International Association of the Scientific Study of Intellectual Disabilities (IASSID), by profound intellectual disabilities combined with profound neuromotor dysfunctions, sensory impairments and general health problems (Van Der Putten & Vlaskamp, 2011).

The intellectual disabilities of these individuals are so profound that no existing standardized tests are applicable for a valid estimation of their level of intellectual capacity. As such, and as an alternative to an IQ definition, many clinicians consider, for classification purposes, a developmental framework that places the cognitive functioning of people with PIMD in a sensory motor stage, i.e., between 0 and 18 months (Vlaskamp, 2005). The neuromotor dysfunctions (e.g., spastic tetraplegia), in turn, commonly confine individuals with PIMD to a wheelchair, with little or no use of their hands or arms, along with difficulties in maintaining posture balance (Vlaskamp & Van Der Putten, 2009). In terms of sensory impairments, most people with PIMD experience cortical visual impairments, a condition related to damages in the visual cortex (Van Splunder, Stilma, & Evenhuis, 2003; Van Splunder, Stilma, Bernsen, & Evenhuis, 2006). Individuals with this condition have variable degrees of acuity loss caused by brain dysfunction and, thus, their ability to see is often inaccurate and inconsistent. Hearing impairments (e.g., Evenhuis, Theunissen, Denkers, Verschuure, & Kemme, 2001) and dysfunctions of taste and smell (e.g., Bromley, 2000) are also common in people with PIMD, but their prevalence is generally lower than that for cortical visual impairments. The tactile and cutaneous senses, including the receptors of touch, pressure, temperature and pain, are also thought to be impaired to some degree (Oberlander, O’Donell, & Montgomery, 1999). Finally, within the general health problems of individuals with PIMD, seizure disorders are a very frequently occurring form of co-morbidity (Tadema & Vlaskamp, 2010). Gastro-oesophageal reflux (which not only causes gastritis and leads to vomiting and feeding irritability, but also to recurrent pneumonia and other chronic respiratory disorders) (Tadema & Vlaskamp, 2010), sleep disorders (Didden, Korzilius, Van Aperlo, Van Overloop, & De Vries, 2002), constipation, osteoporosis and contractures (Veugelers, 2006) are also very common.
Individuals with PIMD form a very heterogeneous group with regard to their functional abilities. A common characteristic, however, is that they are heavily dependent on personal assistance in all aspects of their daily needs (Nakken & Vlaskamp, 2007). A caregiver has to constantly attend to their basic needs, such as eating, drinking and changing, and provide high levels of physical care, such as lifting and positioning (Tadema & Vlaskamp, 2010); the management of seizures, tube feeding and meeting other healthcare needs also require close attention (Tadema & Vlaskamp, 2010). Such a high level of dependence is reinforced by the fact that individuals with PIMD have little or no apparent understanding of verbal language and communicate mostly in a pre- or protosymbolic way, using an idiosyncratic repertoire of behaviours, including facial expressions, body movements, vocalizations, and changes in muscle tension, that are not always consistent (Vos, De Cock, Petry, Noortgate, & Maes, 2010a; Van Der Putten & Vlaskamp, 2011). This means that people with PIMD have to constantly rely on caregivers for the interpretation of their (unconventional) communicative signals, opening up the possibility of misinterpretation and inappropriate or even lack of responses (Hogg, Reeves, Roberts, & Mudford, 2001b). These responses from the caregivers can have serious implications in the quality of support for people with PIMD, and thus in their quality of life.

1.2 Domains of quality of life

The concept of quality of life has been increasingly used as a conceptual framework that guides quality enhancement interventions for people with PIMD (Neilson, Hogg, & Malek, 2000; Schalock et al., 2002; Lancioni, Singh, O’Reilly, Oliva, & Basili, 2005; Vos et al., 2010a, b). According to the World Health Organization's Quality of Life Group (WHOQOL Group), quality of life refers to the “individuals’ perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns” (WHOQOL Group, 1995; World Health Organization, 1997). This concept has a multi-element structure consisting of different domains that may apply to, or be experienced variously by different individuals or groups according to their special needs. That is, the operationalization of quality of life can be

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The current working definition of individuals with PIMD does not enable clinicians and researchers to draw absolute boundaries. While it is already difficult to precisely separate one group of individuals from another on a given characteristic, in the case of individuals with PIMD, multiple and varied characteristics make such separation even more difficult. As such, Vlaskamp and Nakken (2007) have suggested talking about a profound intellectual and multiple disability spectrum. This is comparable with the diagnostic description of individuals belonging to the autistic spectrum (Wing, 2001), in which there are also key characteristics and co-characteristics and, therefore, a variety of clinical descriptors.
different for different groups or individuals, namely those with profound disabilities (Schalock et al. 2002; Schalock, Verdugo, Bonham, Fantova, & Loon, 2008; Brown, Schalock, & Brown, 2009; Verdugo & Schalock, 2009).

Petry, Maes and Vlaskamp (2005) conducted a study aimed at verifying whether the five basic domains of quality of life (physical well-being, material well-being, social well-being, emotional well-being as well as development and involvement in activities), as described by Felce and Perry (1995), are relevant for people with PIMD, according to caregivers. Obtained results showed that the five domains were identified spontaneously by more than half of the respondents as being important for the quality of life of people with PIMD. However, Petry et al. (2005) found that the content of the domains, [as mentioned by Felce and Perry (1995)], differed in a considerable extent for people with PIMD. Caregivers named many indicators related to hygiene, nourishment, rest, technical aids, communication, basic security and individual attention. These indicators were not included as content of the domains in the model of Felce and Perry (1995) but they were considered especially important to people with PIMD because of their limitations and dependency on support in these particular areas. The caregivers, on the other hand, mentioned only few, if any, indicators regarding the indicators of fitness, personal safety, finances and income, stress, mental health and faith [which are all included in the model of Felce and Perry (1995)].

In the present thesis only the domain of emotional well-being, arising from tailored and pleasurable sensory stimulation, will be considered. Emotional well-being is here defined, as in Vos et al. (2010b), in terms of the relative balance of positive to negative emotional states (“hedonic level”).

2. Sensory stimulation as a source of emotional well-being

2.1 From theory to practice: the need for multiple approaches

Clearly, it is important that individuals with PIMD can develop their limited competencies, maintain their capabilities and avoid regression. Many authors defend the use of sensory stimulation as a program designed to reduce sensory deprivation and promote some level of perceptual development in individuals with disabilities, namely PIMD (e.g., Slevin & McCleland, 1999; Vlaskamp, De Geeter, Huijsmans, & Smith, 2003; Chan, Fung, Tong, & Thompson, 2005). The rationale is that as the human physical body requires nutrients, the brain demands positive forms of sensory stimuli at all stages of development, while also needing to be protected from different negative forms of stimulation (Lickliter, 2000; Selwyn, 2000). Moreover, studies have shown that frequent exposure to various sensory
stimuli may contribute to adaptive changes in the brain of individuals with brain damages, including a strengthening of existing synapses, the formation of new synapses, and recruitment of cortical tissue into the activated cortex that was not previously recruited (e.g., Amunts et al., 1997; Hotz et al., 2006; Desmurget, Bonnetblanc, & Duffau, 2007; Baroncelli et al., 2011). Nevertheless, note that no experimental studies have yet been conducted on the direct effects of sensory stimulation in the neural systems of individuals with PIMD.

According to several authors, sensory stimulation programs can also be implemented to promote emotional well-being and reducing problem behaviours in individuals with disabilities (e.g., Green & Reid, 1996; Green, Gardner, & Reid, 1997; Hogg, Cavet, Lambe, & Smeddle, 2001a; Chan et al., 2005; Maes, Lambrechts, Hostyn, & Petry, 2007). In a study evaluating the effects of multisensory therapy for individuals with developmental disabilities, Chan et al. (2005) reported an immediate impact in inducing relaxation and promoting positive emotions. In other studies, Green and Reid (1996) and Green et al. (1997), evaluated the degree to which the presentation of most preferred (i.e., most approached) and less preferred (i.e., most avoided) stimuli relates with emotional well-being of people with PIMD. In their studies, happiness was defined as “any facial expression or vocalization typically considered to be an indicator of happiness among people without disabilities, such as smiling, laughing and yelling when smiling”. Unhappiness, in contrast, was defined as “any facial expression or vocalization typically considered to be an indicator of unhappiness among people without disabilities, such as frowning, grimacing and yelling without smiling”. For each of the participants with PIMD, the higher frequencies of happiness indices were observed during the presentation of the most preferred sensory stimuli. In conclusion, the authors of this study reported that, subsequently, caregivers were able to increase happiness indices through presentation and contingent withdrawal of sensory stimuli.

Curiously, when analyzing the sensory stimulation programs offered in day centers for individuals with PIMD, different authors have found that little time is scheduled for planned and structured interventions and that the majority of them focus on the group rather than on the individual (e.g., Brodin & Renblad, 2000; Vlaskamp, Hiemstra, & Wiersma, 2007; Van Der Putten & Vlaskamp, 2011). This may pose a problem as people with PIMD are known to have specific, idiosyncratic thresholds for stimuli, with some reacting in a hypersensitive way, and subsequently trying to ignore certain stimuli or parts of stimuli, and others needing very strong stimuli before they can actually respond (Vlaskamp & Cuppen-Fonteine, 2007). The sensory stimulation programs that are commonly implemented in day-centers are, therefore, very frequently not tailored to each individual’s specific characteristics (namely functional abilities) and are generally provided
without a prior objective assessment of each individual’s sensory responsiveness. Why is that?

Assessment of the sensory responsiveness of individuals with PIMD, and of their potential appraisals of environmental stimuli (namely pleasantness), is found to be very difficult as we encounter a wide range of barriers to the conventional types of assessment that are feasible with more able individuals (Vlaskamp & Cuppen-Fonteine, 2007; Vos et al., 2010a,b). In practice, such assessments are mostly limited to i) questionnaires directed to caregivers and ii) observation and analysis of the behaviour of people with PIMD (Tadema, Vlaskamp, & Ruijssenaars, 2005, 2007; Vlaskamp & Cuppen-Fonteine, 2007). There are, however, serious questions about whether these are valid approaches. Caregivers gradually build up practical knowledge about the sensory responsiveness of individuals with PIMD and the hedonic value of some of the items that are presented to them during interventions, but such knowledge remains predominantly intuitive, fragmented and unused, and ends up being lost when significant people move out of the individuals’ life (Bradley et al., 1997; Zijlstra, Vlaskamp, & Buntinx, 2001). Moreover, results of several studies indicate that preference rankings based on caregivers’ opinions do not consistently coincide with the results of a systematic observational approach (e.g., Reid, Green, & Parsons, 2003). The latter itself is not problem-free as the “reading” of the behavioural responses to stimulation of individuals with PIMD is very difficult. In many cases, the neuromotor impairments are so profound that individuals are impeded to perform conspicuous reactions to sensory stimuli and, thus, look relatively calm even when experiencing significant arousal (Vlaskamp et al., 2007). Moreover, even when individuals can display observable behaviours in response to stimulation, those behaviours are rarely consistent and not easy to decode (Hogg et al., 2001b; Lancioni et al., 2005). To overcome this caveat, some researchers have recently proposed the use of complementary physiological measures to go beyond the fairly restricted and ambiguous range of behavioural indices, which hardly allow to infer about the actual hedonic value of the stimuli (i.e., whether they are appraised as pleasant or unpleasant) presented to individuals with PIMD during sensory interventions (e.g., Lancioni et al., 2005; Brinkman, 2009).

2.2 Appraising the novelty and pleasantness of sensory stimuli: theoretical background

Since 1980, appraisal theories have become a major perspective in the study of behavioural and physiological indicators of emotional well-being (Scherer, 1984, 1999, 2001, 2009; Ellsworth & Scherer 2003; Delplanque et al., 2009). The central tenet of these
theories is that the elicitation and differentiation of an emotion are determined by appraisals, that is, by continuous evaluation of sensory stimuli (Delplanque et al., 2009). According to Scherer (1984, 2001) as well as Ellsworth and Scherer (2003), these appraisals can occur at different levels of information processing, (sensory motor, schematic and conceptual representational; Sander, Grandjean, & Scherer, 2005) and are organized in four major types of information that an organism needs to process to adaptively react to a salient sensory stimulus: (a) How relevant is this stimulus for me? (relevance); (b) What are the implications or consequences of this event and how do they affect my well-being and my immediate or long-term goals? (implications); (c) How well can I cope with or adjust to these consequences? (coping potential); (d) What is the significance of this event for my self-concept and for social norms and values? (normative significance). These major classes of appraisals are themselves organized more finely in sub-evaluations or sub-checks. For instance, in relevance detection, a first sub-evaluation is related to novelty detection in that any change in the ongoing flow of processed stimuli could require attention and demand further processing (Delplanque et al., 2009). Note here that studies in this area suggest that a large number of factors may affect novelty detection, including stimulus characteristics (such as timing and intensity), and the prior state of the organism (such as arousal level) (Ellsworth & Scherer, 2003). In a second sub-check, the organism evaluates whether a stimulus event is likely to result in pleasure and approach behaviour or pain, withdrawal and avoidance behaviour (intrinsic pleasantness evaluation) (Ellsworth & Scherer, 2003). Even though the concept of pleasure is as old as the philosophical inquiry into human nature, and even though concepts of pleasurable rewards and reinforcement are the cornerstones of many influential psychological theories, we are still far from understanding which, and how, features of stimuli correlate with liking, pleasure or preference, on the one hand, or dislike, aversion or distress, on the other hand (Ellsworth & Scherer, 2003).

It is, at this point, important to highlight that the most basic sub-evaluations are precisely, those related to the novelty and intrinsic pleasantness (i.e., hedonic value) of stimuli. Both are often coded at a very low level of processing, often in a highly automatic fashion. Some theorists object to the use of terms such as ‘evaluation’ or ‘appraisal’ for this kind of low level information processing, insisting that these terms imply some higher “properly cognitive” operation [for current and historical aspects of this ongoing debate see Scherer (2001) and Schorr (2001)]. Because these dimensions are evolutionarily

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2 In the appraisal models framework, the term emotion is reserved for short periods of time during which functionally defined organismic subsystems are coupled or synchronized to produce an adaptive reaction to an event that is considered central to the individual's well-being. These organismic subsystems are the cognitive system, the autonomic system, the motor system, the motivational system and the monitor system (Delplanque et al., 2009).
important and fundamental to the experience of emotion, and because they can be processed on very low levels of cognitive functioning (Ellsworth & Scherer, 2003), as those of individuals with PIMD, they are considered in this thesis.

2.2.1 Heart rate and electrodermal activity changes as indicators of appraisals

While appraisal effects in the motor system are commonly expressed in the form of facial expressions and body movements, effects in the autonomic nervous system are mostly evidenced by changes in heart rate and electrodermal activity. According to several studies, heart rate is particularly sensitive to the novelty of stimuli, decelerating in response to a novel, low to moderate intensity stimulus (orienting response) and accelerating in response to an abrupt, highly intense, novel stimulus (startle reflex) (e.g., Turpin & Siddle, 1983; Turpin, Schaefer, & Boucsein, 1999; Vila et al., 2007). Also, studies suggest that heart rate deceleration seems to indicate pleasantness while acceleration seems to be a relevant physiological indicator of pain or discomfort (e.g., Bensafi et al., 2002; Anttonen & Surakka, 2005). In this regard, Delplanque et al. (2009) recently found evidence for temporal priority of stimulus novelty processing over pleasantness processing on cardiac activity, with novelty processing being observed about 2-4 seconds after stimulus onset, and pleasantness evaluation being observed about 5-10 seconds after stimulus presentation.

Electrodermal activity reflects sympathetic cholinergic function that induces changes in the skin’s resistance to electrical conduction. It can be spontaneously or reflexively evoked by a variety of internal or externally applied arousal stimuli (Vetrugno, Liguori, Cortelli, & Montagna, 2003; Cacciopo, Tassinary, & Bernston, 2007), and has been established over decades of research to be one of the most popular and convenient measures of the autonomic nervous system arousal (e.g., Ishchenko & Shev’ev, 1989; Blain et al. 2008; Lane, Reynolds, & Thacker, 2010). Electrodermal activity includes two distinct variables. The first one is the skin conductance level, the slow, tonic changes measured across long duration stimulus. The second variable consists of the electrodermal responses related to the presentation of a novel, unexpected, significant, or aversive stimulus; these are quick, phasic increases in electrodermal activity (of more than 0.01 μS$^3$) imposed on shifts in tonic level of conductivity and occurring in a 1-5 seconds latency window following stimulus onset (Cacciopo et al., 2007). With the exception of responses elicited by aversive stimuli, these responses are generally

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$^3$ Minimum values referred in the literature varied between 0.01 and 0.05 μS.
considered components of the orienting response produced during the novelty check (Cacciopo et al., 2007).

2.3 Dogs as potential sensory stimuli for individuals with PIMD

Over the last decades, interest has arisen almost worldwide in interventions that intentionally include or incorporate animals as part of therapeutic or recreational activities aiming, for instance, at providing sensory stimulation (e.g., Redefer & Goodman, 1989; Barak, Savorai, Mavashev, & Beni, 2001; Brown, 2005). Levinson (1969) began to actively implement these so-called animal-assisted interventions in the 1960s, but there is earlier evidence of animals, and particularly dogs, being used for therapeutic purposes (Serpell, 2006). The first recorded setting in which dogs were used therapeutically was in the mental hospital York Retreat in England, around 1850 (see Netting, Wilson, & New, 1987). There are also reports of dogs being used in a home for epileptics in Germany in the 1800s and in a World War II convalescent hospital (Netting et al., 1987).

The current acceptance of dog-assisted interventions in a number of institutions, such as hospitals, residential care facilities, and schools, is mostly based on a theoretical link between contact with a dog and human mind-body health [for a discussion on the variety of possible mechanisms of action that have been proposed see Kruger and Serpell (2006)], which has been gaining some experimental support. A number of studies have already associated these interventions with a significant reduction in cardiopulmonary pressures, neurohormone levels, anxiety and stress in various populations (e.g., DeMello, 1999; Barker, Pandurangi, & Best, 2003; Parslow & Jorm 2003; Friedmann, Thomas, Cook, Tsai, & Picot, 2007; Cole, Gawliński, Steers, & Kotlerman, 2007; Morrison, 2007). Reductions in pain perception, namely in hospitalized children, are also reported in Barker et al. (2003), with findings suggesting that dog assisted interventions may be a useful adjunct to traditional pain management (e.g., Velde, Cipriani, & Fisher, 2005; Sobo, Eng, & Kassity-Krich, 2006). Other experimental studies have shown that dog-assisted interventions can have positive effects in social interactions by promoting feelings of happiness, positive social behaviours and communication (e.g., Martin & Farnum 2002; Kovacs, Bulucz, Kis, & Simon, 2006), and decreasing aggression, agitation, self-absorption and stereotypical behaviours (e.g., Silva, Correia, Lima, Magalhães, & de Sousa, 2011; Churchill, Safaoui, McCabe, & Baun, 1999; Redefer & Goodman, 1989). Improvements in self-esteem and self-determination (e.g., Walsh & Mertin 1994; Chu, Liu, Sun, & Lin, 2009), and alleviation of depression and feelings of loneliness (e.g., Walsh & Mertin, 1994; Banks & Banks, 2002; Barker et al., 2003). Curiously, despite such a considerable body of evidence suggesting that dogs may be a source of emotional well-
being for individuals with a variety of characteristics and diagnoses, to date only one study has focused on the potential of dog-assisted interventions for individuals with PIMD.

Heimlich (2001) conducted an investigation aimed at assessing the effectiveness of a dog-assisted therapy program for 14 children with PIMD residing in a long-term care facility. Children were evaluated on four variables including attention span, physical movement, communication and compliance. Three raters observed the children on each variable for a three-week period before the beginning of the therapy sessions to establish the baselines. One animal and the same handler delivered the structured therapy program over eight sessions to each child. Although the obtained data showed a positive trend with the effect of animal-assisted therapy, due to a number of confounding factors (e.g., number of trials that ended up being smaller than originally intended due to direct negative impact of the therapy sessions on the animal’s health) it was not possible to make generalizations regarding the efficacy of the studied intervention.

The lack of research on animal-assisted interventions, and more specifically on the effects of dogs, per se, on the behaviour and physiology of individuals with PIMD, becomes even more curious as one considers Redefer and Goodman’s view (Redefer & Goodman, 1989) that dogs are a “powerful multisensory stimulus” - strong clear sounds, vivid visual impression, special smell, and innovation to touch - from which therapists can beneficiate to combat the low arousal levels of children with developmental disabilities, while promoting positive emotions (Redefer & Goodman, 1989). These animals are also “demanding- likely to follow, lick, and bark at the rejecting child, and their simple, repetitive nonverbal, actions, are easy to decode” thus facilitating interaction and engagement. Following this, one may entertain the hypothesis that dogs, as sensory stimuli, could have positive effects also on individuals with PIMD.

3. Aims of this thesis

The aims of the present thesis were twofold, both with potential implications for the emotional well-being of individuals with PIMD:

i) explore the combined use of behavioural and physiological measurements for an objective assessment of the sensory responsiveness of children with PIMD and of their potential appraisals of different stimuli;

ii) analyse the responsiveness of children with PIMD to a certified dog as to set a preliminary basis for future research to infer about the possibility of using such multisensory live stimuli during interventions.
In regard to the first objective two experimental studies were conducted: one pilot study aimed at accurately describing the behavioural and physiological reactions of one child with PIMD to a set of stimuli commonly used, in Portugal, during sensory interventions (Chapter 2), and one follow up study, using a similar, although somewhat modified, experimental protocol, aimed at extending findings to three additional children (Chapter 3).

In regard to the second objective, a study was conducted with the aim of rigorously assessing, for the very first time, the behavioural and physiological reactions of two children with PIMD to a certified dog (Chapter 4). As to better evidence the potential benefits of the animal, the responsiveness of the children to a familiar caregiver and to an artificial multisensory stimulus, specifically designed to be utilized in sensory interventions, were also assessed. Based on the results obtained in the experimental study presented in Chapter 4, a theoretical consideration was undertaken to highlight the need for future research to focus on the potential use of dogs as effective stimuli to be used, also, in functional communication training programs for individuals with PIMD (Chapter 5).

A total of 5 children with PIMD, aged between 3 and 6 years old, participated in the studies included in the present thesis. With the exception of one boy who participated in two studies (Chapter 3 and Chapter 4), all other children were only involved in one study. Permissions for the participants’ inclusion in the studies were obtained through informed consent provided by the caregivers (Appendix A). All experimental protocols were approved by the Ethics Committee of the O’Porto University (Annexe A).

4. References


Can you know me better? An exploratory study combining behavioural and physiological measurements for an objective assessment of sensory responsiveness in a child with profound intellectual and multiple disabilities

Lima, M., Silva, K., Magalhães, A., Amaral, I. & de Sousa, L.

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ABSTRACT

Sensory assessment of individuals with profound intellectual and multiple disabilities (PIMD) can be difficult for several reasons, including their idiosyncratic reactions to stimuli. This case report presents a combination of behavioural and physiological measurements aimed at providing an objective assessment of the sensory responsiveness of a child with PIMD.

The participant was presented with a set of nineteen stimuli commonly used during sensory interventions. Responsiveness to these stimuli was assessed in terms of motor movements, heart rate and electrodermal reactions.

Although the child only exhibited consistent motor reactions to three of the items, he showed consistent physiological reactions to a total of ten stimuli.

Obtained data, although preliminary, suggests that combining behavioural and physiological measurements may constitute a useful resource for assessing the actual responsiveness of individuals with PIMD. With such a resource, people close to these individuals could fine tune their interventions and guarantee their emotional well-being.
2.1. INTRODUCTION

Recently, individuals with profound intellectual and multiple disabilities (PIMD) have been the focus of many investigations (e.g., Lancioni et al. 2009; Van Der Putten et al. 2009; Vlaskamp & Van Der Putten 2009). According to Nakken and Vlaskamp (2007), these individuals have two key defining characteristics: (a) profound intellectual disabilities to such a degree that no existing standardized tests are applicable for a valid estimation of their level of intellectual capacity, and (b) profound neuromotor dysfunctions. Additionally, they show little or no apparent understanding of verbal language, no apparent symbolic interactions with objects, and nearly no ability for self-support being heavily dependent on personal assistance for everyday tasks (Goldbart 1997). These individuals have profound sensory limitations, including visual impairments (Van Splunder et al. 2003), auditory deficits (Evenhuis et al. 2001) and dysfunctions of taste, smell and touch (Bromley 2000). Also they may exhibit a range of medical conditions, including epilepsy (Kelly et al. 2004).

Studies suggest that any activity providing sensory inputs may contribute to adaptive changes in the brain, including a strengthening of existing synapses, the formation of new synapses, and recruitment of cortical tissue into the activated cortex that was previously not recruited (e.g., Amunts et al. 1997). Accordingly, many authors defend the use of sensory stimulation as an intervention designed to reduce the sensory deprivation and improve the motor skills and the cognitive functions of individuals with PIMD (e.g., Hotz et al. 2006). However, despite the heterogeneity of these people, and the need to have specific knowledge about which stimuli cause which reaction in each individual, the majority of sensory interventions are adapted to a whole group of people and not to individuals (Brodin & Renblad 2000).

Assessment of individuals with PIMD can be extremely difficult, particularly when they do not perform conspicuous reactions to sensory stimuli and look relatively calm even when experiencing significant arousal (Vlaskamp et al. 2007). To this extent, some researchers stress the need to consider physiological parameters, such as electrodermal reactions (EDR) and heart rate (HR), to examine the reactivity of the autonomic nervous system (ANS) to environmental input through different sensory modalities (e.g., Vlaskamp & Cuppen-Fonteine 2007).

Electrodermal reactions are known to be influenced primarily by sympathetic elicitation of sweat secretion in the presence of startling or threatening stimuli, aggressive or defensive feelings, and during emotional events (McIntosh et al. 1999). Also HR acceleration upon confrontation with a stimulus is commonly associated with sympathetic activation as an adaptive strategy to escape the potential dangers of threatening stimuli...
(Goodwin et al. 2006). Deceleration after apprehending a stimulus, in turn, is generally considered a component of a relaxation response (Chlan 1998).

Although Vos and colleagues (2010) have already identified some physiological correlates of negative and positive emotions in adults with PIMD, no further attempts have been made to use physiological measurements to assess specific reactions to sensory stimuli, namely those that are frequently used in clinical interventions. The present case report aimed at assessing the behavioural and physiological reactions of a child with PIMD to a set of stimuli ‘believed’ by clinicians to be appropriate for sensory interventions. This exploratory study is in the interest of people with PIMD in that it presents descriptive data that highlights the importance of considering, and combining, different methodological approaches to investigate the actual sensory responsiveness of these individuals before planning interventions.

2.2. METHODS

2.2.1. Participant

The participant in this study was a three-year-old Caucasian boy diagnosed with profound intellectual and multiple disabilities since the age of 3 months. He presented severe motor limitations together with sensory impairments and lack of speech. He showed serious visual and auditory impairments and exhibited generalized hypotonia associated with artrogriposis. He was tracheotomized and connected to a continuous positive airway pressure ventilator. Feeding was performed via nasogastric tube. He was kept in bed continuously and received physiotherapy on a daily routine. The participant presented a 0 to 3-month performance level in the Callier-azusa scale (Stillman 1978), as assessed by a physiotherapist with specialized training in the administration of this instrument. Noteworthy is the fact that he was not diagnosed with epilepsy; otherwise the presentation of stimuli could not have been performed (Takenoushi et al. 2010).

2.2.2. Sensory stimuli

Prior to the beginning of the study, exploratory interviews with professionals from various early intervention service centres, in Portugal, were conducted to identify the stimuli that are most commonly used during sensory interventions. Among these, a group of items was selected for the present study: 18 stimuli from 5 different sensory modalities (visual, olfactory, gustatory, auditory and tactile) and 1 stimulus providing a combination of simultaneous auditory, tactile and visual stimulation (see Table 1). All items have been
used in previous experimental studies with different populations [e.g., individuals with profound intellectual disabilities (Schmidt 1991; LaRosa 2007) and individuals with dementia (Bakshi 2004)], and represented no risk to the participant.

2.2.3. Setting

The participant was exposed to each of the referred stimuli at home, in a private room where distractions and interruptions were avoided. The temperature of the room remained within a narrow range for the comfort of the child. No other person but the researcher and two assistants were present in the room throughout the experimental sessions. The two assistants were previously trained to be familiarized with all the experimental procedures underlying the different phases of the study.

2.2.4. Experimental procedures

Stimuli were randomly presented for 5 seconds, at constant inter-stimuli intervals of 30 seconds, and care was taken to perform presentation as standardized as possible across experimental sessions. Despite the random presentation of the stimuli, attention was paid in order not to present gustatory stimuli consecutively. Items were presented only once during each session and were kept out of the participant’s sight both before and after presentation. A total of three experimental sessions were conducted (on non-consecutive days and always at the same time of the day to control for circadian rhythms in the ANS responses). A preliminary session was conducted with the aim of familiarizing the researcher and the assistants with both the participant and the setting. This session also provided an opportunity for the child to get used to the experimental procedure.

At the beginning of each session, the Carolina Record of Individual Behaviour (CRIB; Simeonsson et al. 1988) was administered in order to assess the child’s initial alertness state. The CRIB includes nine levels of arousal, from deep sleep to marked uncontrollable agitation. The study only proceeded if the child was in a quiet or active awake state (levels 5 and 6, respectively).

The participant was recumbent with the head of his bed elevated approximately 45° throughout the presentation of all stimuli and was never specifically directed to attend to the items nor was required to complete a task.
**Table 1:** List of the sensory stimuli presented to the participant.

<table>
<thead>
<tr>
<th>Stimuli</th>
<th>Sensory modalities</th>
<th>Modes of presentation of the stimuli</th>
<th>References</th>
</tr>
</thead>
<tbody>
<tr>
<td>Tambourine</td>
<td>Audition</td>
<td>The researcher taps a tambourine, about 2 meters from the participant and out of his sight, for 5 seconds.</td>
<td>Murphy et al. 2004</td>
</tr>
<tr>
<td>Bell</td>
<td>Audition</td>
<td>The researcher rings a bell, about 2 meters from the participant and out of his sight, for 5 seconds.</td>
<td>Schoen &amp; Miller 2008</td>
</tr>
<tr>
<td>Meditation single chime</td>
<td>Audition</td>
<td>The researcher rings a chime, about 2 meters from the participant and out of his sight, for 5 seconds.</td>
<td>Lancioni 2008</td>
</tr>
<tr>
<td>Strobe red light</td>
<td>Vision</td>
<td>The research positions a red strobe light, set to 10 flashes per second, slightly below the eye level of the participant, for 5 seconds.</td>
<td>Schoen &amp; Miller 2008</td>
</tr>
<tr>
<td>White light</td>
<td>Vision</td>
<td>The researcher positions a white LED flashlight pen, slightly below the eye level of the participant and turns it on and off, for 5 seconds.</td>
<td>Goodall &amp; Corbett 1982</td>
</tr>
<tr>
<td>Colourful puppet</td>
<td>Vision</td>
<td>The researcher positions a colourful puppet slightly below the eye level of the participant, for 5 seconds.</td>
<td>LaRosa 2007</td>
</tr>
<tr>
<td>Cold thermal bag</td>
<td>Touch</td>
<td>The researcher lays a cold thermal bag on the lateral part of participant’s right leg, below the knee, for 5 seconds.</td>
<td>Schmidt 1991</td>
</tr>
<tr>
<td>Hot thermal bag</td>
<td>Touch</td>
<td>The researcher lays a hot thermal bag on the lateral part of participant’s right leg, below the knee, for 5 seconds.</td>
<td>Schmidt 1991</td>
</tr>
<tr>
<td>Ear syringe</td>
<td>Touch</td>
<td>The researcher uses an ear syringe to apply puffs of air to the forehead of the participant, for 5 seconds.</td>
<td>Schmidt 1991</td>
</tr>
<tr>
<td>Soft surface</td>
<td>Touch</td>
<td>The researcher rubs the lateral part of participant’s right leg, below the knee, with a soft surface for 5 seconds.</td>
<td>Bakshi 2004</td>
</tr>
<tr>
<td>Rough surface</td>
<td>Touch</td>
<td>The researcher rubs the lateral part of participant’s right leg, below the knee, with a rough surface for 5 seconds.</td>
<td>Bakshi 2004</td>
</tr>
<tr>
<td>Smooth surface</td>
<td>Touch</td>
<td>The researcher rubs the lateral part of participant’s right leg, below the knee, with a smooth surface for 5 seconds.</td>
<td>Bakshi 2004</td>
</tr>
<tr>
<td>Vinegar essence</td>
<td>Olfaction</td>
<td>The researcher place a saturated swab with a vinegar essence about 3 cm from the participant’s nose, centered between nose and lips, and then moves it in approximately 3 cm path from the left to the right, for 5 seconds.</td>
<td>Schoen &amp; Miller 2008</td>
</tr>
<tr>
<td>Orange essence</td>
<td>Olfaction</td>
<td>The researcher place a saturated swab with an orange essence about 3 cm from the participant’s nose, centered between nose and lips, and then moves it in approximately 3 cm path from the left to the right, for 5 seconds.</td>
<td>Schoen &amp; Miller 2008</td>
</tr>
<tr>
<td>Vanilla essence</td>
<td>Olfaction</td>
<td>The researcher place a saturated swab with a vanilla essence about 3 cm from the participant’s nose, centered between nose and lips, and then moves it in approximately 3 cm path from the left to the right, for 5 seconds.</td>
<td>Schmidt 1991</td>
</tr>
<tr>
<td>Lemon solution</td>
<td>Gustation</td>
<td>The researcher applies 2 large drops of a lemon solution to the participant’s tongue, using a 20 ml sterile disposable pipette.</td>
<td>LaRosa 2007</td>
</tr>
<tr>
<td>Sugar solution</td>
<td>Gustation</td>
<td>The researcher applies 2 large drops of a sugar solution to the participant’s tongue, using a 20 ml sterile disposable pipette.</td>
<td>Schmidt 1991</td>
</tr>
<tr>
<td>Salt solution</td>
<td>Gustation</td>
<td>The researcher applies 2 large drops of a salt solution to the participant’s tongue, using a 20 ml sterile disposable pipette.</td>
<td>Schmidt 1991</td>
</tr>
<tr>
<td>Multi-Sensory Center device</td>
<td>Audition, Vision and Touch</td>
<td>The researcher places the device in contact with the sole of the participant’s right foot, for 5 seconds.</td>
<td></td>
</tr>
</tbody>
</table>

*Include studies in which the same stimuli were used; **All swabs were moistened prior to presentation.*
2.2.5. Data collection and analysis

2.2.5.1. Motor reactions

Sessions were videotaped for further description of the participant’s motor movements. A coding scheme using Table 2 was set up in the Observer XT Software version 7.0 (Noldus Information Technologies, Wageningen, The Netherlands) to determine the frequency and duration of each motor movement exhibited by the participant immediately before (5 seconds), during (5 seconds), and immediately after the presentation of each stimulus (20 seconds). Note that this participant was only able to produce two types of motor movements (limb and eyeball movements), both with a reduced amplitude. These movements were very subtle and not easy to detect.

Inter-observers reliability, using Pearson $r$ correlation, was assessed between the researcher and an observer familiar with children with PIMD, who independently scored all videotapes. The values obtained for $r$ were above 0.9 for the frequencies and durations of all the motor movements recorded throughout the study.

Motor reactions were considered as numerical changes from baseline in the frequencies and/or durations of the motor movements exhibited by the participant during stimuli presentation (during - baseline differences) and/or after stimuli presentation (after - baseline differences). Consistent reactions were considered as changes from baseline occurring in all experimental sessions (e.g., an increase from baseline in the duration of a specific movement, occurring in all three experimental sessions).

For each consistent reaction, Cohen’s d and effect size r were calculated to determine the magnitude of the difference between the mean frequencies and durations recorded during baseline and those observed during and/or after stimulus presentation.

Given the different periods of time considered in the protocol (5 seconds during baseline and stimuli presentation versus 20 seconds following stimuli presentation), the frequencies and durations of the motor movements exhibited by the participant after stimuli presentation were corrected accordingly.

Table 2: Motor movements exhibited by the participant and corresponding descriptions.

<table>
<thead>
<tr>
<th>Motor movements</th>
</tr>
</thead>
<tbody>
<tr>
<td>Flexion and extension movements, of reduced amplitude, of the limbs (arms, fingers and foot)</td>
</tr>
<tr>
<td>Eyeball movements</td>
</tr>
</tbody>
</table>
2.2.5.2. Physiological reactions

Electrodermal activity and HR were continuously recorded throughout the study, with the non-invasive ambulatory Biopac MP100 Data Acquisition System (Biopac Systems, Santa Barbara, California, USA).

The general method of collecting these physiological measures followed the procedures recommended by Cacioppo and colleagues (2007). Two pregelled disposable electrodes, placed on the child’s wrists, were used to record HR. In turn, electrodermal activity was collected through two unpolarizable electrodes (containing isotonic recording gel) placed on the volar surfaces of the distal phalanges of the middle and thumb fingers of the participant’s left hand (previously washed with soap and water); one ground electrode was placed on his left ankle. All electrodes were secured with sticky collars and then wrapped with Coban to remain in place throughout data collection. It is important to stress that the participant was given a 5-minute adaptation period prior to data collection as to allow him to get used to the electrodes.

All equipment was first extensively tested in a research laboratory so that no technical problems were expected to arise during the application of the protocol. Also the researcher and assistants received intensive training as to ensure adequate procedures guaranteeing the comfort of the participant at all stages.

Prior to analysis, physiological data were visually inspected in the Acqknowledge 3.9.1 Software (Biopac Systems, Santa Barbara, California, USA) and recording artefacts (e.g., large single increases or decreases in HR due to coughing) were identified and discarded. Also, event markers signalling the onset of each stimulus were placed so that physiological reactions could be analyzed.

Electrodermal reactions were defined as the largest peak (above 0.02µs; Cacioppo et al. 2007) occurring within 1 and 5 seconds after the onset of each stimulus (note that there is an approximate 1-second lag between the presentation of the stimulus and the resultant EDR peak, McIntosh et al. 1999). A particular sensory item was considered to elicit consistent electrodermal responding when EDRs to that item were recorded in all three sessions. In those cases, the mean values of the amplitude of the peaks were calculated.

Heart rate reactions were considered as numerical changes from baseline in the mean values of HR recorded during and/or following the presentation of the stimuli. Consistent HR reactions were considered as changes from baseline occurring in all experimental sessions (e.g., increases from baseline in response to a specific stimulus, occurring in all experimental sessions). For each consistent reaction, Cohen’s d and effect size r were calculated to determine the magnitude of the difference between the mean
values of HR recorded during baseline and those registered during and/or after stimulus presentation.

2.3. RESULTS

2.3.1. Motor reactions

Three out of the nineteen sensory items presented to the participant elicited consistent motor reactions. An increase from baseline in the frequency and duration of eyeball movements was observed, in all experimental sessions, during the presentation of the vinegar essence (Table 3; frequency: Cohen’s $d=2.57$, effect size $r=0.79$; duration: Cohen’s $d=1.42$, effect size $r=0.58$). Contrastingly, a decrease in the frequency and duration of eyeball movements was consistently observed following the presentation of the multisensory item (Table 3; frequency: Cohen’s $d=1.50$, effect size $r=0.60$; duration: Cohen’s $d=2.43$, effect size $r=0.77$). Finally, an increase in the frequency and duration of the limb movements was observed, in all experimental sessions, following the presentation of the white light (Table 3; frequency: Cohen’s $d=1.52$, effect size $r=0.60$; duration: Cohen’s $d=1.96$, effect size $r=0.70$).

2.3.2. Physiological reactions

The following items induced EDRs in all experimental sessions: the vinegar essence, the white light, the bell, the lemon solution, the sugar solution and the multisensory device. All reactions occurred during the presentation of the items (see Table 3 for mean amplitudes).

In respect to HR reactions, consistent increases were observed during and following the presentation of the vinegar essence (Table 3; during: Cohen’s $d=1.73$, effect size $r=0.65$; following: Cohen’s $d=1.94$, effect size $r=0.70$) and of the bell (Table 3; during: Cohen’s $d=8.48$, effect size $r=0.97$; following: Cohen’s $d=7.67$, effect size $r=0.97$). A consistent increase in HR was also observed during the presentation of the white light (Table 3; Cohen’s $d=1.76$, effect size $r=0.66$). Consistent decreases, in turn, were observed following the orange essence (Table 3; Cohen’s $d=1.93$, effect size $r=0.69$), during and following the tambourine (Table 3; during: Cohen’s $d=0.80$, effect size $r=0.37$; following: Cohen’s $d=1.01$, effect size $r=0.45$), during and following the salt solution (Table 3; during: Cohen’s $d=1.27$, effect size $r=0.54$; following: Cohen’s $d=1.29$, effect size $r=0.54$), during the hot bag (Table 3; Cohen’s $d=1.26$, effect size $r=0.53$) and during and
following the multisensory device (Table 3; during Cohen’s $d=2.16$, effect size $r=0.73$; following: Cohen’s $d=1.50$, effect size $r=0.60$).

**Table 3**: Consistent motor and physiological reactions exhibited by the participant during and following each stimulus presentation. The changes presented refer to mean differences from baseline.

<table>
<thead>
<tr>
<th>Sensory modality</th>
<th>Stimuli</th>
<th>During stimuli presentation</th>
<th>Following stimuli presentation</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Olfaction</strong></td>
<td>Vinegar</td>
<td><strong>Eyeball movements</strong> <em>(mean change in frequency=1.33, mean change in duration=2.00)</em></td>
<td><strong>HR</strong> <em>(mean change=6.2)</em></td>
</tr>
<tr>
<td></td>
<td></td>
<td><strong>HR</strong> <em>(mean change=5.89)</em></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td><strong>EDR</strong> <em>(mean amplitude=0.022)</em></td>
<td></td>
</tr>
<tr>
<td>Orange</td>
<td></td>
<td></td>
<td><strong>HR</strong> <em>(mean change=-5.3)</em></td>
</tr>
<tr>
<td>Vanilla</td>
<td></td>
<td></td>
<td><strong>N. C. R.</strong></td>
</tr>
<tr>
<td>Vision</td>
<td>Puppet</td>
<td><strong>N. C. R.</strong></td>
<td><strong>N. C. R.</strong></td>
</tr>
<tr>
<td>White light</td>
<td></td>
<td><strong>HR</strong> <em>(mean change=6.2)</em></td>
<td><strong>Limb movements</strong> <em>(mean change in frequency=1.13; mean change in duration=1.67)</em></td>
</tr>
<tr>
<td></td>
<td></td>
<td><strong>EDR</strong> <em>(mean amplitude=0.030)</em></td>
<td><strong>N. C. R.</strong></td>
</tr>
<tr>
<td>Red light</td>
<td></td>
<td><strong>N. C. R.</strong></td>
<td><strong>N. C. R.</strong></td>
</tr>
<tr>
<td>Audition</td>
<td>Chime</td>
<td><strong>N. C. R.</strong></td>
<td><strong>N. C. R.</strong></td>
</tr>
<tr>
<td>Tambourine</td>
<td></td>
<td><strong>HR</strong> <em>(mean change=-5.9)</em></td>
<td><strong>HR</strong> <em>(mean change=6.8)</em></td>
</tr>
<tr>
<td>Bell</td>
<td></td>
<td><strong>HR</strong> <em>(mean change=23.6)</em></td>
<td><strong>HR</strong> <em>(mean change=22.5)</em></td>
</tr>
<tr>
<td></td>
<td></td>
<td><strong>EDR</strong> <em>(mean amplitude=0.030)</em></td>
<td></td>
</tr>
<tr>
<td>Gustation</td>
<td>Lemon</td>
<td><strong>EDR</strong> <em>(mean amplitude=0.024)</em></td>
<td><strong>N. C. R.</strong></td>
</tr>
<tr>
<td></td>
<td>Sugar</td>
<td><strong>EDR</strong> <em>(mean amplitude=0.026)</em></td>
<td><strong>N. C. R.</strong></td>
</tr>
<tr>
<td>Salt</td>
<td></td>
<td><strong>HR</strong> <em>(mean change=-6.4)</em></td>
<td><strong>HR</strong> <em>(mean change=6.8)</em></td>
</tr>
<tr>
<td>Touch</td>
<td>Ear syringe</td>
<td><strong>N. C. R.</strong></td>
<td><strong>N. C. R.</strong></td>
</tr>
<tr>
<td></td>
<td>Hot bag</td>
<td><strong>HR</strong> <em>(mean change=-6.0)</em></td>
<td><strong>N. C. R.</strong></td>
</tr>
<tr>
<td></td>
<td>Cold bag</td>
<td><strong>N. C. R.</strong></td>
<td><strong>N. C. R.</strong></td>
</tr>
<tr>
<td></td>
<td>Rough surface</td>
<td><strong>N. C. R.</strong></td>
<td><strong>N. C. R.</strong></td>
</tr>
<tr>
<td></td>
<td>Smooth surface</td>
<td><strong>N. C. R.</strong></td>
<td><strong>N. C. R.</strong></td>
</tr>
<tr>
<td></td>
<td>Soft surface</td>
<td><strong>N. C. R.</strong></td>
<td><strong>N. C. R.</strong></td>
</tr>
<tr>
<td>Vision, audition and touch</td>
<td>Multi-sensory Center device</td>
<td><strong>HR</strong> <em>(mean change=-9.4)</em> <strong>EDR</strong> <em>(mean amplitude=0.024)</em></td>
<td><strong>Eyeball movements</strong> <em>(mean change in frequency=1.58; mean change in duration=2.65)</em></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td><strong>HR</strong> <em>(mean change=-6.8)</em></td>
</tr>
</tbody>
</table>

**Notes**: The mean changes in durations are shown in seconds and the mean changes in HR are shown in beats per minute. The mean amplitudes of EDR are shown in microsiemens. The symbol ‘N.C.R.’ stands for non consistent reactions, either motor or physiological.
2.4. DISCUSSION

Many people with PIMD show very little variation in observable behaviour and seem largely unresponsive to a range of sensory stimuli. In this regard, the importance of the present study lies in two main points: i) the suggestion that, even children with PIMD exhibiting a very limited behavioural repertoire can show consistent reactions to sensory stimulation, and ii) the suggestion that complementing behavioural with physiological parameters may evidence internal experiences (arguably comfort and discomfort) that would otherwise be considered non-existent.

The participant in the present study showed consistent increases/decreases in motor activity in response to three of the presented items. Although the motor reactions observed (movements of the limbs and movements of the eyeballs) were reported by the participant’s parents when asked about his sensory responsiveness, they were not confirmed by the view of clinicians (personal communications obtained prior to the beginning of the experimental study). Little agreement between parents and clinicians on questions regarding the reactions of children with PIMD to sensory stimulation is common being generally attributed to the idiosyncratic responses of this population (Maes et al. 2007).

Interestingly, the consistent increases in motor activity observed in the present study were accompanied by consistent increases in HR, while consistent decreases in motor activity were accompanied by consistent decreases in HR. It is known that unpleasant evaluations of sensory stimuli may produce increases in HR, as well as in motor activity, to reduce intake and processing of stimulation, or to reject or expel noxious matter (Cacioppo et al. 2007; Delplanque et al. 2009). Decreases in HR and in motor activity, in turn, are commonly interpreted as a relaxation response (e.g., Chlan 1998).

The participant in this study did not exhibit evident discomfort or defensive reactions to the presented stimuli. Nevertheless, when exposed, for instance, to the vinegar essence (a potentially aversive stimulus), he showed a consistent increase in motor activity, which, by occurring simultaneously with acceleration in HR, suggests some degree of discomfort. The consistent decrease of motor activity and HR observed in response to the multisensory device suggests a more positive evaluation of this sensory item. Similar results in studies with other populations have promoted the use of multisensory items in therapeutic interventions with the aim of inducing relaxation and reducing challenging or stereotyped behaviours (e.g., Hotz et al. 2006).

Seven out of the nineteen stimuli presented to the participant elicited consistent physiological reactions (EDR or HR reactions) but no motor ones. Such results seem to suggest that the participant may be a lot more responsive than could be thought from
considering observable behaviour alone. Attending to the actual responsiveness of children with PIMD is highly important in that it may allow proxy people to respond in an appropriate fashion, for example by removing highly arousing and potentially harmful/stressful stimuli (likely associated with consistent EDRs and increases in HR), or presenting pleasant items (likely associated with consistent EDRs and decreases in HR) (e.g., Rosenstein & Oster 1988; Maes et al. 2007). In this extent improved sensitivity and responsivity to both the motor and the physiological reactions of individuals with PIMD may not only have serious implications for their emotional well-being but it may also lead to a growing feeling of competency in caregivers.

Because presented data only refer to one participant, it may be questionable whether such a higher number of consistent physiological reactions when compared to motor ones can be generalized to a larger number of individuals with PIMD. Additional single-subject studies are needed to make stronger the claim that these individuals may be much more responsive than it is commonly assumed. The level of understanding provided by such studies will certainly promote person-centred, evidence-based interventions that recognize the importance of integrating behavioural and physiological assessments of sensory responsiveness when designing individualized profiles for individuals with PIMD (see Adams & Oliver 2011).

2.5. ETHICAL CONSIDERATIONS

The experimental protocol was approved by the ethics committee of the O'Porto University (Portugal). Previous to study participation, parents received written and spoken information about the aims and content of the investigation. Also they were informed about their rights in accepting the participation of their child and their right to later withdraw at any time with no adverse consequences to the participant (in such a case, all collected data would have to be destroyed). All procedures regarding the participant's name and confidential information are in compliance with the Helsinki Declaration. The findings from this research should be disseminated in a form that is accurate and clinically useful to the physicians, health care providers, participant, family and society at large.

2.6. ACKNOWLEDGMENTS

We would like to thank the participant as well as his family and his physiotherapist, from the Early Intervention Service at the ‘CERCI Gaia’, who so generously volunteered their time to participate in this study. Also we are thankful to all the professionals from various
Early Intervention Centers in Portugal who helped us with stimuli selection. Finally we thank Mariana Filipe for videotaping all sessions. Fundação para a Ciência e a Tecnologia funded the participation of Mariely Lima (FCT-SFRH/BD/44748/2008), Karine Silva (FCT-SFRH/BPD/37017/2007), and Ana Magalhães (FCT-SFRH/BPD/19200/2004).

2.7. REFERENCES


Chapel Hill: Frank Porter Graham Child Development Center, University of North Carolina at Chapel Hill.


Beyond behavioural observations: a deeper view through the sensory reactions of children with profound intellectual and multiple disabilities

Lima, M., Silva, K., Magalhães, A., Amaral, I. & de Sousa, L.
ABSTRACT

The present study was aimed at assessing the behavioural and physiological responsiveness of three children with profound intellectual and multiple disabilities (PIMD) to a set of sensory stimuli.

Eighteen items, ‘believed’ by clinicians to be appropriate for routine interventions, were presented to the participants and responsiveness was assessed in terms of i) consistent behavioural and electrodermal reactions and ii) biphasic changes in heart rate.

Results were twofold. First, all participants showed frequent consistent physiological reactions but rare consistent behavioural ones. Second, all participants showed biphasic changes in heart rate, pointing to the possibility of different appraisals (novelty and pleasantness) of most of the presented items.

Data here presented may have implications for the development and the emotional well-being of individuals with PIMD in that it suggests that reactions to, and potential appraisals of, sensory stimuli may occur despite the lack of consistent observable behaviours.
3.1. INTRODUCTION

Recent studies have been focusing on the factors that contribute to the emotional well-being of people with profound intellectual and multiple disabilities (PIMD) (e.g., Vos et al. 2010). People with PIMD have profound intellectual disabilities (Vlaskamp et al. 2007), profound neuromotor dysfunctions (Petry et al. 2007), sensory impairments (Evenhuis et al. 2001) and medical problems (Zijlstra & Vlaskamp 2005). Their communicative abilities are limited, being mostly situated at a pre- or proto-symbolic level characterized by bodily and idiosyncratic expressions (Grove et al. 1999). Overall, people with PIMD constitute a vulnerable group with heavy dependence on personal assistance for physical care, education, recreation and sensory stimulation (Nakken & Vlaskamp 2007).

Several authors highlighted the potential of individually tailored sensory stimulation to reduce the sensory deprivation and improve the motor skills and the cognitive functions of people with brain injuries (e.g., Hotz et al. 2006). Recent studies suggest that sensory interventions may improve the ability of the nervous system to process sensory information, having a positive impact on the individual's ability to participate in daily-life activities (e.g., Lane & Schaaf 2010). The severity and the complexity of disabilities impede people with PIMD to respond to sensory stimuli in a consistent, easy to decode, way (Lancioni et al. 2005). While some individuals may exhibit reflexive movements, and occasionally display spontaneous behaviours (e.g., smiling or crying), they rarely show sustained and reproducible responses to items presented to them during sensory interventions, leaving caregivers 'in the dark' about the hedonic value of different stimuli (see Hogg et al. 2001). To overcome this problem, researchers have proposed supplementing behavioural observations with measurements of physiological parameters, such as electrodermal activity (EDA) and heart rate (HR) (e.g., Lancioni et al. 2005).

Recent studies suggest that physiological reactions to a sensory stimulus may result from sequential appraisals of its relevance, in terms of novelty and pleasantness (e.g., Ellsworth & Scherer 2003; Sander et al. 2005). In a first check, an individual evaluates the novelty of a stimulus in the ongoing stream of information. While the detection of a novel, low- to moderate-intensity, stimulus produces an orienting response with associated changes in the support system (e.g., deceleration in HR), the detection of an abrupt, highly intense, novel stimulus elicits a startle reaction (e.g., acceleration in HR). In a second check, an individual evaluates whether a stimulus is likely to result in pleasure or pain, modifying the changes that have already been produced in the preliminary check. While an unpleasant evaluation produces a defensive reaction (e.g., HR acceleration), turning the body away from the unpleasant item, a pleasant evaluation results in a
sustained orienting response (e.g., HR deceleration), with motor behaviour that turns the body towards the stimulus.

The present study aimed at exploring whether combining behavioural and physiological assessments can evidence sensory reactions in individuals with PIMD that would not be visible with a behavioural approach alone. Three children with PIMD were presented with a number of sensory stimuli and their individual reactions were assessed in terms of observable behaviour, EDA and biphasic changes in HR. Individual differences in the observed reactions suggest the need for tailor-made stimulation programs.

3.2. METHODS

3.2.1 Participants

Participant 1 was a five-year-old Caucasian girl diagnosed with PIMD due to a perinatal encephalopathy. She presented spastic tetraparesia and profound intellectual disability. She showed auditory impairments and minimal or functional residual vision. This participant showed no conventional communication skills and was completely dependent on caregivers for fulfilment of basic needs.

Participant 2 was a five-year-old Caucasian boy diagnosed with PIMD due to a perinatal encephalopathy. He presented severe hypotonia and showed a profound level of intellectual disability and visual impairments. This participant showed no conventional communication skills and was completely dependent on caregivers for fulfilment of basic needs.

Participant 3 was a six-year-old Caucasian boy diagnosed with PIMD due to a mitochondrial cytopathy. He presented tetraparesia associated with severe hypotonia and profound intellectual disabilities. The degree of his sensory impairments was unknown and he did not show recognizable communication means or self-help skills.

No participants were diagnosed with epilepsy; otherwise the presentation of stimuli could not have been performed (Takenouchi et al. 2010).

3.2.2 Stimuli used in the experimental study

A group of 18 items from 5 different sensory modalities (visual, olfactory, gustatory, auditory and tactile) were used in the present study (see Table 1). Items were selected based on interviews with professionals working in early intervention who identified stimuli commonly used during sensory interventions. All items have been used in previous experimental studies (e.g., LaRosa 2007) and represented no risk to the participants.
Table 1: List of the stimuli presented to the participants.

<table>
<thead>
<tr>
<th>Stimuli</th>
<th>Sensory modalities</th>
<th>Modes of presentation of the stimuli</th>
<th>References</th>
</tr>
</thead>
<tbody>
<tr>
<td>Tambourine</td>
<td>Audition</td>
<td>The researcher taps a tambourine, about 2 meters from the participant and out of his sight, for 5 seconds.</td>
<td>Murphy et al. 2004</td>
</tr>
<tr>
<td>Bell</td>
<td>Audition</td>
<td>The researcher rings a bell, about 2 meters from the participant and out of his sight, for 5 seconds.</td>
<td>Schoen &amp; Miller 2008</td>
</tr>
<tr>
<td>Meditation</td>
<td>Audition</td>
<td>The researcher rings a chime, about 2 meters from the participant and out of his sight, for 5 seconds.</td>
<td>Lancionni 2008</td>
</tr>
<tr>
<td>single chime</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Strobe red light</td>
<td>Vision</td>
<td>The researcher positions a red strobe light, set to 10 flashes per second, slightly below the eye level of the participant, for 5 seconds.</td>
<td>Schoen &amp; Miller 2008</td>
</tr>
<tr>
<td>White light</td>
<td>Vision</td>
<td>The researcher positions a white LED flashlight pen, slightly below the eye level of the participant and turns it on and off, for 5 seconds.</td>
<td>Goodall &amp; Corbett 1982</td>
</tr>
<tr>
<td>Colourful puppet</td>
<td>Vision</td>
<td>The researcher positions a colourful puppet slightly below the eye level of the participant, for 5 seconds.</td>
<td>LaRosa 2007</td>
</tr>
<tr>
<td>Cold thermal bag</td>
<td>Touch</td>
<td>The researcher lays a cold thermal bag on the lateral part of participant’s right leg, below the knee, for 5 seconds.</td>
<td>Schmidt 1991</td>
</tr>
<tr>
<td>Hot thermal bag</td>
<td>Touch</td>
<td>The researcher lays a hot thermal bag on the lateral part of participant’s right leg, below the knee, for 5 seconds.</td>
<td>Schmidt 1991</td>
</tr>
<tr>
<td>Ear syringe</td>
<td>Touch</td>
<td>The researcher uses an ear syringe to apply puffs of air to the forehead of the participant, for 5 seconds.</td>
<td>Schmidt 1991</td>
</tr>
<tr>
<td>Soft surface</td>
<td>Touch</td>
<td>The researcher rubs the lateral part of participant’s right leg, below the knee, with a soft surface for 5 seconds.</td>
<td>Bakshi 2004</td>
</tr>
<tr>
<td>Rough surface</td>
<td>Touch</td>
<td>The researcher rubs the lateral part of participant’s right leg, below the knee, with a rough surface for 5 seconds.</td>
<td>Bakshi 2004</td>
</tr>
<tr>
<td>Smooth surface</td>
<td>Touch</td>
<td>The researcher rubs the lateral part of participant’s right leg, below the knee, with a smooth surface for 5 seconds.</td>
<td>Bakshi 2004</td>
</tr>
<tr>
<td>Vinegar essence**</td>
<td>Olfaction</td>
<td>The researcher place a saturated swab with a vinegar essence about 3 cm from the participant’s nose, centered between nose and lips, and then moves it in approximately 3 cm path from the left to the right, for 5 seconds.</td>
<td>Schoen &amp; Miller 2008</td>
</tr>
<tr>
<td>Orange essence**</td>
<td>Olfaction</td>
<td>The researcher place a saturated swab with an orange essence about 3 cm from the participant’s nose, centered between nose and lips, and then moves it in approximately 3 cm path from the left to the right, for 5 seconds.</td>
<td>Schoen &amp; Miller 2008</td>
</tr>
<tr>
<td>Vanilla essence**</td>
<td>Olfaction</td>
<td>The researcher place a saturated swab with a vanilla essence about 3 cm from the participant’s nose, centered between nose and lips, and then moves it in approximately 3 cm path from the left to the right, for 5 seconds.</td>
<td>Schmidt 1991</td>
</tr>
<tr>
<td>Lemon solution</td>
<td>Gustation</td>
<td>The researcher applies 2 large drops of a lemon solution to the participant’s tongue, using a 20 ml sterile disposable pipette.</td>
<td>LaRosa 2007</td>
</tr>
<tr>
<td>Sugar solution</td>
<td>Gustation</td>
<td>The researcher applies 2 large drops of a sugar solution to the participant’s tongue, using a 20 ml sterile disposable pipette.</td>
<td>Schmidt 1991</td>
</tr>
<tr>
<td>Salt solution</td>
<td>Gustation</td>
<td>The researcher applies 2 large drops of a salt solution to the participant’s tongue, using a 20 ml sterile disposable pipette.</td>
<td>Schmidt 1991</td>
</tr>
</tbody>
</table>

*Include studies in which the same stimuli were used; **All swabs were moistened prior to presentation.
3.2.3 Setting

Each participant was exposed to the stimuli, at home, in a private room where distractions and interruptions were avoided. No other person but the researcher and two assistants were present throughout testing. Assistants were previously trained to be familiarized with all the experimental procedures.

3.2.4 Experimental procedures

Stimuli were randomly presented for 5 seconds, at constant inter-stimuli intervals of 30 seconds, and care was taken to perform the presentation as standardized as possible across sessions. Despite the random presentation of the stimuli, attention was paid in order not to present gustatory stimuli consecutively. Stimuli were presented only once during each session and were kept out of the participants' sight both before and after presentation.

A total of five experimental sessions were conducted, on non-consecutive days and always at the same time of the day. A preliminary session was conducted to familiarize all participants with the experimental procedure.

At the beginning of each session, the Carolina Record of Individual Behaviour (CRIB; Simeonsson et al. 1988) was administered in order to assess each child's initial alertness state. The study only proceeded if the children were in a quiet or active awake state.

The participants were never specifically directed to attend to the items nor were they required to complete a task.

3.2.5 Data collection and analysis

3.2.5.1 Behavioural reactions

Experimental sessions were videotaped using a SONY HDR FX1 camera. Coding schemes using Table 2 were set up in the Observer XT Software version 7.0 (Noldus Information Technologies, Wageningen, The Netherlands) to determine the frequencies and durations of all the behaviours exhibited by each participant during a 5-second baseline period (immediately before the presentation of each stimulus) and within 10 seconds immediately after the onset of each stimulus.

Inter-observers reliability, using Pearson $r$ correlation, was assessed between the researcher and an independent observer who was familiar with children with PIMD and
who scored all videotapes. The values obtained for $r$ were above 0.9 for the frequencies and durations of all the behaviours recorded throughout the study.

Behavioural reactions to a particular stimulus were considered as numerical changes from baseline in the frequencies and/or durations of the behaviours exhibited by each participant during the 5-second period following stimulus onset (period 1) and/or the subsequent 5-second period (period 2). For each participant, consistent reactions were considered as changes from baseline occurring in all experimental sessions (e.g., a consistent increase in the frequency of a particular behaviour during period 1, occurring in all experimental sessions).

Table 2: Behaviours exhibited by the participants and corresponding descriptions.

<table>
<thead>
<tr>
<th>Behaviour</th>
<th>Description</th>
<th>Reference</th>
</tr>
</thead>
<tbody>
<tr>
<td>Disorganized</td>
<td>Continuous and disorganized movements of the four limbs</td>
<td>Becker et al. 1999</td>
</tr>
<tr>
<td>behaviour&lt;sup&gt;a&lt;/sup&gt;</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Startle</td>
<td>Grimacing (see below) and blinking followed by flexion of the four limbs</td>
<td>Huntsman et al. 2008</td>
</tr>
<tr>
<td>Tongue protrusion</td>
<td>Visible extension of the tongue outside of the mouth</td>
<td>Ricardo et al. 2005</td>
</tr>
<tr>
<td>Smiling</td>
<td>At least one corner of the lips rounded upward, with mouth opened or closed</td>
<td>Marsh et al. 2010</td>
</tr>
<tr>
<td>Frowning</td>
<td>Medial end of the eyebrows pulled together</td>
<td>Hwang et al. 2004</td>
</tr>
<tr>
<td>Eyes open wide</td>
<td>Sclera showing above the iris</td>
<td>Rothbart 1988</td>
</tr>
<tr>
<td>Grimacing</td>
<td>Brow bulge, eye squeeze, and deepening of the nasolabial furrow</td>
<td>Nishitani et al. 2009</td>
</tr>
<tr>
<td>Eye closure</td>
<td>Lids closed for more than 3 consecutive seconds</td>
<td>Koegel et al. 1987</td>
</tr>
<tr>
<td>Eye gaze</td>
<td>A sustained look at the stimulus for more than 3 consecutive seconds</td>
<td>Smith et al. 2010</td>
</tr>
<tr>
<td>Moan&lt;sup&gt;b&lt;/sup&gt;</td>
<td>A low, sustained, mournful cry lasting for more than 3 consecutive seconds (without cry)</td>
<td>Hadden &amp; von Baeyer 2002</td>
</tr>
</tbody>
</table>

<sup>a</sup>Only recorded for participant 3. <sup>b</sup>Only recorded for participant 1.

3.2.5.2 Physiological reactions

In each experimental session, EDA and HR were simultaneously and continuously recorded, throughout the presentation of all stimuli, with the non-invasive ambulatory Biopac MP100 Data Acquisition System (Biopac Systems, Santa Barbara, California, USA).

Collection of physiological parameters followed the procedures recommended by Cacioppo and colleagues (2007). Note here that all participants were given a 5-minute adaptation period prior to data collection as to allow them to get used to the equipment.
The hardware was first extensively tested in a research laboratory so that no technical problems were expected to arise during the application of the protocol. Also, the researcher and the assistants received intensive training to ensure adequate procedures guaranteeing the comfort of the participants at all stages.

Prior to analysis, physiological data were visually inspected in the Acqknowledge 3.9.1 Software (Biopac Systems, Santa Barbara, California, USA) and recording artefacts (e.g., large single increases or decreases in HR due to disorganized movements or moans and deep inspirations) were identified and discarded. Also, event markers signalling the onset of each stimulus were placed so that physiological reactions could be analyzed.

For each participant, electrodermal reactions (EDR) to a stimulus were defined, according to Cacioppo and colleagues (2007), as the largest peak (greater than 0.02µs) occurring within 1 and 5 seconds after the stimulus onset (note that there is an approximate 1-second lag between the presentation of the stimulus and the resultant EDR peak, McIntosh et al. 1999). For all reactions, the amplitude of the peak was measured according to Lane and colleagues (2010), from the point at which the skin conductance increases sharply to the point at which the conductance begins to fall. The latency of all reactions was also recorded for each participant. A particular stimulus was considered to elicit consistent electrodermal responding when EDRs to that stimulus were recorded in all 5 sessions. In those cases, the mean values of the latency and the amplitude of the peaks were calculated.

Following previous studies (e.g., Delplanque et al. 2009), biphasic changes in HR were considered to occur when i) the mean values of HR recorded within 5 seconds post-stimulus onset (period 1) differed significantly from those recorded during the baseline period (Wilcoxon matched pairs tests) and ii) the mean values of HR recorded within 5 seconds post-stimulus onset (period 1) differed significantly from those recorded in the 5-10 seconds window period (period 2) following stimulus onset (Wilcoxon matched pairs tests).

All statistical analyses were conducted using STATISTICA 7.0 (StatSoft, Tulsa, Oklahoma, USA) and a significance level of $p < 0.05$ was used.

3.3. RESULTS

Participant 1

Only two of the presented stimuli elicited consistent behavioural reactions in participant 1. In all experimental sessions, the sound of the bell elicited a startle, in the 5 second-period following its onset (period 1). In addition, the sour solution consistently elicited grimacing
and eye closure, in the 5-10 seconds window period following the onset of its presentation (period 2). The mean differences (± SD) from baseline in the frequencies and durations of these consistent reactions are shown in Table 3. None of these behaviours were observed during the baseline period.

Seven out of the eighteen presented items induced an EDR in all experimental sessions [see Table 3 for mean (± SD) amplitudes and latencies]. Finally, nine items induced biphasic changes in HR (Wilcoxon matched pairs tests: for all tests Z=2.023 and p=0.043). Table 3 shows the mean (± SD) values of the significant changes recorded. A total of seven items elicited both consistent EDRs and biphasic changes in HR.

Table 3: Consistent reactions exhibited by participant 1 including i) behavioural reactions [mean change in frequency (± SD); mean change in duration (± SD), ii) EDRs [mean amplitude (± SD) and mean latency (± SD)], and iii) biphasic changes in HR [mean change (± SD)]. All changes refer to mean differences from baseline.

<table>
<thead>
<tr>
<th>Stimulus</th>
<th>Behavioural reactions</th>
<th>EDR</th>
<th>HR</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Period 1</td>
<td>Period 2</td>
<td>Amplitude</td>
</tr>
<tr>
<td>Tambourine</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Bell</td>
<td>-</td>
<td>Startle 1</td>
<td>0.05 (±0.02)</td>
</tr>
<tr>
<td>Chime</td>
<td>-</td>
<td>-</td>
<td>0.09 (±0.01)</td>
</tr>
<tr>
<td>Strobe red light</td>
<td>-</td>
<td>-</td>
<td>0.03 (±0.01)</td>
</tr>
<tr>
<td>White light</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Puppet</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Cold thermal bag</td>
<td>-</td>
<td>-</td>
<td>0.03 (±0.01)</td>
</tr>
<tr>
<td>Hot thermal bag</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Ear syringe</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Soft surface</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Rough surface</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Smooth surface</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Vinegar essence</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Orange essence</td>
<td>-</td>
<td>-</td>
<td>0.03 (±0.01)</td>
</tr>
<tr>
<td>Vanilla essence</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Sour solution</td>
<td>-</td>
<td>Grimacing 4</td>
<td>4.3 (±1.8)</td>
</tr>
<tr>
<td>Sweet solution</td>
<td>-</td>
<td>Eye closure 2</td>
<td>0.03 (±0.01)</td>
</tr>
<tr>
<td>Salty solution</td>
<td>-</td>
<td>-</td>
<td>0.06 (±0.02)</td>
</tr>
</tbody>
</table>

* Only frequency was recorded.

Notes: Changes in duration are shown in seconds and changes in HR in beats per minute. The mean amplitudes of EDR are shown in microsiemens and the mean latencies in seconds. In cells with missing values (-) no consistent reactions were observed.
Participant 2

Six of the presented items elicited consistent behavioural reactions in participant 2. As in participant 1, the sound of the bell elicited a startle, in the 5 second-period following its onset (period 1). No other consistent behavioural reactions were observed during this period. In the 5-10 seconds window period following stimulus onset (period 2), participant 2 smiled in response to the cold bag, closed his eyes when presented with the ear syringe, and showed grimacing, frowning and eye closure behaviour at all gustatory stimuli (sour, sweet and salty solutions). The mean differences (± SD) from baseline in the frequencies and durations of these consistent reactions are shown in Table 4. Again, none of the referred behaviours were observed during the baseline period.

Ten out of the eighteen presented items induced an EDR in all experimental sessions [see Table 4 for mean (± SD) amplitudes and latencies]. Finally, thirteen items induced biphasic changes in HR (Wilcoxon matched pairs tests: for all tests $Z=2.023$ and $p=0.043$). Table 4 shows the mean (± SD) values of the significant changes recorded. A total of six items elicited both consistent EDRs and biphasic changes in HR.

Table 4: Consistent reactions exhibited by participant 2 including i) behavioural reactions [mean change in frequency (± SD); mean change in duration (± SD)], ii) EDRs [mean amplitude (± SD) and mean latency (± SD)], and iii) biphasic changes in HR [mean change (± SD)]. All changes refer to mean differences from baseline (see next page).
<table>
<thead>
<tr>
<th>Stimulus</th>
<th>Behavioural reactions</th>
<th>Period 1</th>
<th>Period 2</th>
<th>EDR</th>
<th>HR</th>
</tr>
</thead>
<tbody>
<tr>
<td>Tambourine</td>
<td>Startle</td>
<td>-</td>
<td>-</td>
<td>-1.13 (±0.14)</td>
<td>5.20 (±1.31)</td>
</tr>
<tr>
<td>Bell</td>
<td>-</td>
<td>0.16 (±0.01)</td>
<td>4.17 (±1.98)</td>
<td>5.89 (±2.84)</td>
<td>-7.93 (±4.23)</td>
</tr>
<tr>
<td>Chime</td>
<td>-</td>
<td>0.06 (±0.01)</td>
<td>3.19 (±1.51)</td>
<td>5.76 (±1.83)</td>
<td>7.07 (±4.04)</td>
</tr>
<tr>
<td>Strobe red light</td>
<td>-</td>
<td>0.12 (±0.04)</td>
<td>4.32 (±2.47)</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>White light</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-11.43 (±5.32)</td>
<td>13.24 (±4.39)</td>
</tr>
<tr>
<td>Puppet</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-3.56 (±2.04)</td>
<td>-11.22 (±9.56)</td>
</tr>
<tr>
<td>Cold thermal bag</td>
<td>-</td>
<td>1 (±0.0); 3.2 (±0.5)</td>
<td>-</td>
<td>-3.39 (±1.58)</td>
<td>-3.71 (±2.60)</td>
</tr>
<tr>
<td>Hot thermal bag</td>
<td>-</td>
<td>0.18 (±0.01)</td>
<td>4.88 (±2.33)</td>
<td>-6.14 (±0.85)</td>
<td>-2.31 (±1.59)</td>
</tr>
<tr>
<td>Ear syringe</td>
<td>-</td>
<td>4 (±1.3); 4.0 (±1.0)</td>
<td>-</td>
<td>-</td>
<td></td>
</tr>
<tr>
<td>Soft surface</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-6.31 (±3.41)</td>
<td>12.47 (±6.35)</td>
</tr>
<tr>
<td>Rough surface</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-13.02 (±7.78)</td>
<td>9.45 (±6.22)</td>
</tr>
<tr>
<td>Smooth surface</td>
<td>-</td>
<td>0.07 (±0.04)</td>
<td>4.25 (±0.79)</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Vinegar essence</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>2.40 (±1.46)</td>
<td>-8.79 (±4.40)</td>
</tr>
<tr>
<td>Orange essence</td>
<td>-</td>
<td>0.17 (±0.10)</td>
<td>4.57 (±3.00)</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Vanilla essence</td>
<td>-</td>
<td>0.03 (±0.01)</td>
<td>2.08 (±0.85)</td>
<td>-4.13 (±1.69)</td>
<td>-6.99 (±1.35)</td>
</tr>
<tr>
<td>Sour solution</td>
<td>-</td>
<td>0.46 (±0.40)</td>
<td>3.47 (±0.66)</td>
<td>-8.00 (±0.94)</td>
<td>7.79 (±1.18)</td>
</tr>
<tr>
<td>Sweet solution</td>
<td>-</td>
<td>1.3 (±0.5); 2.0 (±1.0)</td>
<td>4.5 (±1.61)</td>
<td>-4.09 (±0.43)</td>
<td>7.32 (±1.25)</td>
</tr>
<tr>
<td>Salty solution</td>
<td>-</td>
<td>0.17 (±0.02)</td>
<td>3.77 (±0.99)</td>
<td>-</td>
<td>-</td>
</tr>
</tbody>
</table>

Notes: Changes in duration are shown in seconds and changes in HR in beats per minute. The mean amplitudes of EDR are shown in microsiemens and the mean latencies in seconds. In cells with missing values (-) no consistent reactions were observed.

*Only frequency was recorded.*
Participant 3

Six of the presented items elicited consistent behavioural reactions in participant 3. As in participant 1 and 2, the sound of the bell elicited a startle, in the 5 second-period following its onset (period 1). No other consistent behavioural reactions were observed during this period. In the 5-10 seconds window period following stimulus onset (period 2), participant 3 smiled in response to the sweet solution, and showed disorganized movements to most of the tactile stimuli (cold thermal bag, soft surface, rough surface and smooth surface). The mean differences (± SD) from baseline in the frequencies and durations of these consistent reactions are shown in Table 5. None of the referred behaviours were recorded during the baseline period.

This participant exhibited more consistent EDRs than consistent behavioural ones. Twelve of the presented items elicited consistent electrodermal responding [see Table 5 for mean (± SD) amplitudes and latencies]. Also a high number of biphasic changes in HR were recorded. Ten of the presented sensory stimuli elicited biphasic changes in HR (Wilcoxon matched pairs tests: for all tests Z=2.023 and p=0.043). Table 5 shows the mean (± SD) values of the significant changes recorded. A total of eight items elicited both consistent EDRs and biphasic changes in HR.

3.4. DISCUSSION

Many authors who have been studying individuals with PIMD defend that identifying consistent reactions to stimulation is important in that it may help caregivers to determine i) which stimuli should be provided as to potentially increase their engagement in activities, and ii) which stimuli should be removed as to potentially make their environment less aversive, threatening and/or unpleasant (e.g., Maes et al. 2007).

The participants in the present study showed few consistent behavioural reactions to the presented stimuli. Participant 1 only reacted consistently to two of the eighteen items and participants 2 and 3 showed consistent behaviours only to 6 stimuli. Curiously, despite the scarce number of consistent behavioural reactions, all participants reacted physiologically to most of the presented stimuli. Interestingly, they all showed consistent biphasic changes in HR (in response to at least one item from each sensory modality), pointing to the occurrence of different appraisals at different points in time [as postulated by Scherer (2001) and Delplanque and colleagues (2009)]. One may consider that the earliest changes in HR (period 1) may have occurred in response to novelty detection, whereas the later ones (period 2) could have been related to a pleasantness check.
Table 5: Consistent reactions exhibited by participant 3 including i) behavioural reactions [mean change in frequency (± SD); mean change in duration (± SD)], ii) EDRs [mean amplitude (± SD) and mean latency (± SD)], and iii) biphasic changes in HR [mean change (± SD)]. All changes refer to mean differences from baseline.

<table>
<thead>
<tr>
<th>Stimulus</th>
<th>Behavioural reactions</th>
<th>EDR Amplitude</th>
<th>EDR Latency</th>
<th>HR Period 1</th>
<th>HR Period 2</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Tambourine</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Bell</td>
<td>Startle</td>
<td>0.03 (±0.00)</td>
<td>2.16 (±0.65)</td>
<td>15.43 (±7.82)</td>
<td>-6.32 (±3.82)</td>
</tr>
<tr>
<td>Chime</td>
<td></td>
<td>0.03 (±0.00)</td>
<td>2.64 (±1.53)</td>
<td>17.38 (±5.68)</td>
<td>-6.89 (±3.59)</td>
</tr>
<tr>
<td>Strobe red light</td>
<td></td>
<td>0.03 (±0.00)</td>
<td>2.32 (±0.51)</td>
<td>-13.42 (±7.49)</td>
<td>-9.21 (±4.34)</td>
</tr>
<tr>
<td>White light</td>
<td></td>
<td>0.03 (±0.00)</td>
<td>3.10 (±0.85)</td>
<td>10.05 (±5.82)</td>
<td>-9.25 (±5.67)</td>
</tr>
<tr>
<td>Puppet</td>
<td>Disorganized movements</td>
<td>0.03 (±0.00)</td>
<td>1.49 (±0.28)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Cold thermal bag</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hot thermal bag</td>
<td></td>
<td>0.03 (±0.00)</td>
<td>3.10 (±0.85)</td>
<td>10.05 (±5.82)</td>
<td>-9.25 (±5.67)</td>
</tr>
<tr>
<td>Ear syringe</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Soft surface</td>
<td>Disorganized movements</td>
<td>0.03 (±0.01)</td>
<td>2.01 (±0.28)</td>
<td>13.22 (±4.15)</td>
<td>10.65 (±5.16)</td>
</tr>
<tr>
<td>Rough surface</td>
<td></td>
<td>0.03 (±0.01)</td>
<td>2.32 (±0.77)</td>
<td>11.59 (±4.34)</td>
<td>11.28 (±3.94)</td>
</tr>
<tr>
<td>Smooth surface</td>
<td>Disorganized movements</td>
<td>0.03 (±0.01)</td>
<td>2.32 (±0.77)</td>
<td>11.59 (±4.34)</td>
<td>11.28 (±3.94)</td>
</tr>
<tr>
<td>Vinegar essence</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Orange essence</td>
<td></td>
<td>0.03 (±0.01)</td>
<td>3.19 (±2.58)</td>
<td>-16.50 (±5.21)</td>
<td>5.80 (±2.16)</td>
</tr>
<tr>
<td>Vanilla essence</td>
<td></td>
<td>0.03 (±0.00)</td>
<td>2.24 (±0.66)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sour solution</td>
<td></td>
<td>0.06 (±0.06)</td>
<td>1.4 (±0.21)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sweet solution</td>
<td>Smiling</td>
<td>0.04 (±0.03)</td>
<td>2.42 (±1.90)</td>
<td>11.56 (±4.66)</td>
<td>-14.21 (±7.05)</td>
</tr>
<tr>
<td>Salty solution</td>
<td></td>
<td>0.06 (±0.04)</td>
<td>1.25 (±0.07)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

*Only frequency was recorded.*

Notes: Changes in duration are shown in seconds and changes in HR in beats per minute. The mean amplitudes of EDR are shown in microsiemens and the mean latencies in seconds. In cells with missing values (-) no consistent reactions were observed.
In the present study, novelty was defined as contextual, meaning that the participants were presented with a novel set of stimuli for the first time in the experimental setting (and not necessarily for the first time in their lives) (as in Delplanque et al. 2005). Among the stimuli that elicited biphasic changes in HR, some induced, as first effect, a significant acceleration during period 1, suggesting a startle reaction to a highly intense stimulus. The startle behaviour observed in all participants as a reaction to the sound of the bell - a potentially high intense stimulus - seems to corroborate this suggestion. Other stimuli, in contrast, elicited, as a first effect, a significant deceleration of HR, pointing to an orienting response to a low-to-moderate stimulus.

According to Bradley and colleagues (2009), cardiac decelerations are related to ‘sensory intake’, reflecting processes involving extracting information from the sensory array, while cardiac acceleration indexes ‘sensory rejection’. In attentional terms, researchers commonly regard these two concepts, orienting and startle, as equivalent to engagement and disengagement, respectively (e.g., Turpin et al. 1999). While orienting reactions resulting from the novelty check involve attention directed toward novel stimuli, startle ones represent disengagement from an ongoing activity (i.e., interrupt), due to the occurrence of an abrupt stimulus. Both reactions may be followed either by further engagement or disengagement, depending on the succeeding evaluation of the hedonic value of the eliciting stimulus (Turpin et al. 1999).

Although scarce, the consistent behavioural reactions exhibited by the participants in the present study suggest some association between emotional valence and, at least, changes in HR (positive emotions: HR deceleration; negative emotions: HR acceleration). The smiling behaviour exhibited by participants 2 and 3, which is typically considered as an indicator of positive emotions (Green & Reid 1996), occurred in both cases, along with a significant deceleration in HR during period 2. In contrast, the observed grimacing and frowning expressions, commonly regarded as indicators of negative emotions (Green & Reid 1996), were associated with a significant acceleration in HR during period 2.

The idea that individuals with PIMD may retain the ability to evaluate pleasantness despite the severity of their impairments is at the basis of studies focusing on the expressions of pleasure and displeasure by these individuals (e.g., Petry & Maes 2006). To date, however, this ability has mainly been attributed based on subjective interpretations of consistent behaviours allegedly connected to situations of pleasure and contentment or pain and discontentment. What in the absence of consistent behaviours? The results obtained in this study suggest that, even if not showing consistent behavioural reactions, individuals with PIMD may be experiencing significant changes in physiology that may translate into improved or reduced emotional well-being.
Although these are preliminary results, they do suggest that attending to physiological parameters may be of particular importance to go beyond the fairly restricted range of behavioural indices used in previous research (e.g., Lancioni et al. 2005). One may question however, how practical and affordable is collecting physiological data such as the one recorded in the present study (HR and EDR). Nevertheless, low cost, and easy-to-use, HR and electrodermal analyses systems have already been designed and implemented to support clinical and educational interventions for individuals with PIMD (e.g., Kobayashi et al. 2010). The value of the information obtained through this method, and the undeniable implications for the emotional well-being of people with PIMD, are such that they fully justify the potential ‘trouble’ of setting up the hardware and software.

3.5. ETHICAL CONSIDERATIONS

The experimental protocol was approved by the ethics committee of the O’Porto University (Portugal). Previous to study participation, parents received written and spoken information about the aims and content of the investigation. Also they were informed about their rights in accepting the participation of their child and their right to later withdraw at any time with no adverse consequences to the participant (in such a case, all collected data would have to be destroyed). All procedures regarding the participant’s name and confidential information are in compliance with the Helsinki Declaration. The findings from this research should be disseminated in a form that is accurate and clinically useful to the physicians, health care providers, participant, family and society at large.

3.6. ACKNOWLEDGEMENTS

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3.7. REFERENCES


CHAPTER 4

Behavioural and physiological responses of children with profound intellectual and multiple disabilities to a therapy dog: a pilot study

Lima, M., Silva, K., Magalhães, A., Amaral, I. & de Sousa, L.
ABSTRACT

Dog assisted interventions have been growing in popularity in many care units for children with profound intellectual and multiple disabilities (PIMD). Although a previous empirical investigation assessing the efficacy of a dog-assisted therapy program for children with PIMD reported a positive effect for all participants, no other studies have tried to evaluate, through rigorous scientific protocols, the pertinence of implementing dog-assisted interventions for these individuals.

The aim of the present study was to quantify the effects of a certified therapy dog on the behaviour and physiology (heart rate and skin conductance level) of two children with PIMD. As to better evidence the potential benefits of the animal to these two children, we also evaluated their behavioural and physiological responsiveness to a familiar caregiver and to an artificial multisensory stimulus.

Both participants appeared to respond positively to the presence of the dog in much the same way as they responded to the familiar caregiver. Contrastingly, children showed opposite responses to the artificial multisensory stimulus.

The present study provides quantitative indications that therapy dogs may promote emotional well-being in some children with PIMD, namely as pleasant, relaxing and attention getting stimuli with which these people could positively engage during planned interventions.
4.1. INTRODUCTION

Individuals with profound intellectual and multiple disabilities (PIMD) are characterized by severe or profound motor disabilities combined with profound intellectual impairments (Nakken & Vlaskamp 2007). In addition they commonly experience sensory impairments (Evenhuis et al. 2001) and suffer from various health problems, such as seizures (Colding & MacDonald 2009) and respiratory disorders (Wallis 2009). These individuals have little or no apparent understanding of verbal language (Vlaskamp & Van Der Putten 2009) and they communicate via idiosyncratic facial expressions, sounds, movements, body postures or muscle tension (Vos et al. 2010). Overall, they are highly dependent on personal assistance for everyday tasks so that caregivers need to constantly attend to their basic needs (e.g., eating, drinking, changing and positioning). In some cases, parents of a child with PIMD can devote up to 18 hours a day on essential caring tasks and therapeutic and educational activities (Tadema & Vlaskamp 2010).

As highlighted by many authors, any activity which increases alertness, engagement and enjoyment is very important for individuals who are dependent on others and whose disabilities are such that they frequently appear very passive (e.g., Hogg et al. 2001; Vlaskamp et al. 2003). In their daily routines, people with PIMD are offered different types of activities, like activities in a multisensory environment, activities outdoors and play activities (Vlaskamp & Nakken 1999). Recently, the use of therapy dogs, as facilitators of activities, has grown in popularity in many care units for individuals with PIMD, partly in response to published accounts of their socio-emotional benefits for people with other disabilities. Redefer and Goodman (1989) showed that dogs, by being a powerful multisensory stimulus – strong clear sound, vivid visual impression, a special smell, and art innovation to touch in a unique interactive style – can help combat the low sensory and affective arousal levels of autistic children. Similarly, Martin and Farnum (2002), as well as Silva and colleagues (2011), found evidence that interactions with dogs can increase socially appropriate behaviours and decrease self-absorption and stereotyped behaviours in individuals with autism. Also according to experimental studies, when utilized as adjuncts to activities for elderly people residing in hospitals and nursing homes, dogs constitute a relaxing focus of attention, promoting feelings of safety and happiness and providing a source of ‘contact comfort’ (e.g., Jorgenson 1997). Interestingly, some recent findings suggest that animal contact may also have important physiological effects. Morrisson (2007), for example, showed that interactions with dogs can have benefits in depression, anxiety, perceived quality of health, and loneliness in different client populations, as well as lead to improvements in blood pressure, HR, and salivary immunoglobulin A levels. Also, Tsai et al. (2010) found that the presence of a dog
can decrease physiological arousal, namely in hospitalized children, and therefore may be useful in helping them to cope better in a hospital setting.

In 2001, Heimlich developed a quantitative research design to assess the efficacy of a dog-assisted therapy program for children with multiple disabilities. Although obtained data pointed to a positive effect for all participants, no further studies have focused on the potential of therapy dogs to promote emotional well-being in individuals with PIMD. The present study aimed at assessing the behavioural and physiological responses of two children with PIMD to a therapy dog. As to better evidence the potential positive effects of the animal, we also assessed the responsiveness of the participants to two elements pertaining to their daily life: a familiar caregiver, with whom each participant is attached to, and an artificial multisensory stimulus designed specifically to be utilized in sensory interventions.

4.2. METHODS

4.2.1 Participants

Participant 1 was five-year-old Caucasian boy diagnosed with profound intellectual and multiple disabilities due to a perinatal hypoxic-ischemic encephalopathy. He presented with severe hypotonia thus spending most time in a wheelchair and showed visual impairments with diagnoses of cortical visual impairment. This participant lacked any conventional communication skills and was completely dependent on caregivers for fulfilment of all his basic needs. He attended kindergarten on a full-time basis and received direct support, twice a week, from a special education teacher.

Participant 2 was a six-year-old Caucasian boy diagnosed with profound intellectual and multiple disabilities due to a mitochondrial cytopathy (complex I). He presented with diplegia, showed stereotyped movements and tactile defensiveness, and spent most time in a wheelchair. He also showed auditory impairments, namely a mild to severe sensorineural hearing loss in his right ear. He did not possess recognizable communication means or self-help skills and showed a high level of dependency on personal assistance for everyday tasks. This participant also attended kindergarten on a full-time basis and received direct support, twice a week, from a special education teacher.

Noteworthy is the fact that participants were not diagnosed with epilepsy (Takenoushi et al. 2010), had no history of bad experiences with dogs, nor any medical conditions, such as asthma or allergies, that could be aggravated by the presence of an animal (Friesen 2010); otherwise the study could not have been performed.
4.2.2. Experimental conditions

An experimental design was planned to analyse and compare the behavioural and physiological responses of each participant to three different experimental conditions: i) presence of a therapy dog, ii) presence of a familiar caregiver and iii) presentation of an artificial multisensory stimulus (Multi-Sensory Center stimulus; 11" x 7", Enabling Devices) designed to provide a combination of auditory, tactile and visual stimulation.

The dog, a female adult Labrador retriever, was recruited, trained and certified by Ânimas, a Portuguese association, member of the Assistance Dogs International Inc. (ADI). All training programs use positive reinforcement (reward-based) approaches and follow the standards and ethics developed by the ADI Standards and Ethics Committee. Certification helped to ensure that the animal carried liability insurance, had the appropriate temperament for the present study and behaved in a predictable manner throughout all experimental sessions (e.g., unperturbed demeanour when confronted by unusual noises, movements and smells). The animal was examined by a veterinarian to ensure she was free from parasitic infections and was current on all the required vaccinations, thus posing no risk of zoonoses transmission to the participants.

4.2.3 Setting

Each participant was tested at home, in a private room where distractions and interruptions were avoided. The temperature of the room remained within a narrow range for the comfort of the child. The principal researcher and two assistants were present in the room during all experimental procedures. The two assistants were previously trained to be familiarized with all the experimental procedures underlying the different phases of the study.

4.2.4 Experimental procedures

Each participant was randomly presented with the therapy dog, the familiar caregiver and the artificial multisensory stimulus during independent experimental sessions, conducted on different and non-consecutive days, and always at the same time of the day. For each condition, 5 experimental sessions were conducted. Each experimental session consisted of 9 minutes of data collection beginning with a 3 min baseline period (period 1) followed by a 3 min period of the scheduled condition (period 2) and a subsequent 3 min post-condition rest period (period 3).
During sessions with the therapy dog, the animal was positioned in a chair, next to the participant, and was silently instructed by the principal researcher to vocalize, touch or lick the child as to provide sensory stimulation. It is here important to highlight that the researcher was qualified as a certified dog handler. She took specific training from Ânimas and passed an evaluation of knowledge about how to identify signs of stress from dogs and how to appropriately mediate the interaction of dogs with various client populations, namely children with PIMD. As handler, the researcher was obliged to be in full and direct control of the dog during the experimental sessions. She never interacted directly with the participant. At the end of each of these sessions the researcher disinfected the participant’s skin surface that was in contact with the dog with an alcohol-based hand rub.

The familiar caregiver sat next to the participant and provided him with sensory input, for instance by caressing his face, kissing and singing. As to avoid confounding effects, the principal researcher was always present in the room during sessions with the presence of the caregiver but never interacted with her or the participant.

Finally, during sessions involving the presentation of the artificial multisensory stimulus, the principal researcher placed the stimulus in close contact with the right foot of the participant.

In each experimental session, and prior to data collection, the Carolina Record of Individual Behaviour (CRIB; Simeonsson et al. 1988) was administered in order to assess each child’s initial alertness state. The CRIB includes nine levels of alertness, from deep sleep to marked uncontrollable agitation. The data collection only proceeded if the child was in a quiet or active awake state (levels 5 and 6, respectively). Also prior to data collection, each participant was given a 5 minute period to acclimatize to the physiological recording equipment.

Before the commencement of the study, a preliminary session was conducted with the aim of familiarizing each child with the experimental procedure, the therapy dog and the artificial multisensory stimulus. This session also provided an opportunity for the principal researcher and the assistants to familiarize with both the participant and the setting.

4.2.5 Data collection and analysis

4.2.5.1 Behavioural responses

All experimental sessions were videotaped using a SONY HDR FX1 camera. The operational speed of the camera was set at 30 frames per second. Coding schemes using
Table 1 were set up in the Observer XT Software version 7.0 (Noldus Information Technologies, Wageningen, The Netherlands) to determine the durations of the behaviours exhibited by each participant during i) the 3 min baseline period preceding each scheduled condition (period 1), ii) the 3 min period of each condition (period 2) and iii) the 3 min post-condition rest period (period 3).

Table 1: Behaviours exhibited by the participants and corresponding descriptions.

<table>
<thead>
<tr>
<th>Behavioural Category</th>
<th>Behaviour</th>
<th>Description</th>
<th>Reference</th>
</tr>
</thead>
<tbody>
<tr>
<td>Self-engagement</td>
<td>Stereotyped body rocking*</td>
<td>A continuous back-and-forth movement of the torso, repeated two or more times within a 10 seconds recording interval</td>
<td>Durand &amp; Carr 1987</td>
</tr>
<tr>
<td>Engagement/interest</td>
<td>Eye gaze</td>
<td>A sustained look at the therapy dog, the caregiver or the artificial multisensory stimulus, for more than 3 consecutive seconds</td>
<td>Watson et al. 2010</td>
</tr>
<tr>
<td>Facial expression indicative of happiness</td>
<td>Smiling</td>
<td>At least one corner of the lips rounded upward, with mouth open or closed</td>
<td>Green &amp; Reid 1996</td>
</tr>
<tr>
<td></td>
<td>Frowning</td>
<td>Medial end of the eyebrows pulled together</td>
<td>Green &amp; Reid 1996</td>
</tr>
<tr>
<td>Facial expressions or vocalizations indicative of unhappiness</td>
<td>Grimacing</td>
<td>Brow bulge, eye squeeze, and deepening of the nasolabial furrow</td>
<td>Nishitani et al. 2009</td>
</tr>
<tr>
<td></td>
<td>Moan</td>
<td>A low, sustained, mournful cry lasting for more than 3 consecutive seconds</td>
<td>Green &amp; Reid 1996</td>
</tr>
<tr>
<td>Avoidance</td>
<td>Eye closure</td>
<td>Lids closed for more than 3 consecutive seconds</td>
<td>Koegel et al. 1987</td>
</tr>
<tr>
<td></td>
<td>Head away</td>
<td>Head turned away from the stimulus</td>
<td>Hadden &amp; von Baeyer 2002</td>
</tr>
</tbody>
</table>

*Only exhibited by participant 2.

Inter-observers reliability, using Pearson $r$ correlation, was assessed between the researcher and an independent observer who scored all videotapes. This observer had previous experience with children with PIMD and data collection methods. The values obtained for $r$ were above 0.9 for the durations of all the behaviours recorded during data collection.

Significant changes from baseline in the durations of the behaviours exhibited by each participant were assessed using Wilcoxon matched pairs tests. When significant changes were observed for more than one experimental condition, differences between conditions were assessed (Friedman ANOVAs for differences between three conditions or Wilcoxon matched pairs tests for differences between two conditions). For each condition,
significant differences between baselines and rest periods were also assessed (Wilcoxon matched pairs tests). Note that the duration of eye gaze, defined here as a sustained look at the therapy dog, the familiar caregiver or the artificial multisensory stimulus, was only compared between conditions (Friedman ANOVA followed by Wilcoxon matched pairs tests).

4.2.5.2 Physiological responses

In each experimental session, electrodermal activity and HR were simultaneously and continuously recorded with the non-invasive ambulatory Biopac MP100 Data Acquisition System (Biopac Systems, Santa Barbara, California, USA).

The general method of collecting these physiological parameters followed the procedures recommended by Cacioppo and colleagues (2007). Two pregelled disposable electrodes, placed on the child’s wrists, were used to record HR. Electrodermal activity, in turn, was collected through two unpolarizable electrodes (containing isotonic recording gel) placed on the volar surfaces of the distal phalanges of the middle and thumb fingers of the participant’s left hand (previously washed with soap and water); one ground electrode was placed on his left ankle. All electrodes were secured with sticky collars and then wrapped with Coban to remain in place throughout the study. All equipment was first extensively tested in a research laboratory by staff from the company so that no technical problems were expected to arise during the application of the protocol. Also the researcher and assistants received intensive training as to ensure adequate procedures guaranteeing the comfort of the participant at all stages.

Prior to analysis, physiological data were visually inspected in the Acqknowledge 3.9.1 Software (Biopac Systems, Santa Barbara, California, USA) and recording artefacts (e.g., large single increases or decreases in HR due to coughing) were identified and discarded. Also, event markers signalling the onset of each stimulus were placed so that physiological responses could be analyzed.

Each of the three different periods of data collection was divided into 18 sequential 10-second blocks. For each block, within each period, the mean skin conductance level (SCL) and heart rate (HR) were scored. For each condition, changes from baseline in the observed scores (of SCL and HR) were assessed using Wilcoxon matched pairs tests. When significant changes were observed for more than one condition, differences between conditions were assessed (Friedman ANOVAs for differences between three conditions or Wilcoxon matched pairs tests for differences between two conditions). For each condition, significant differences between baseline and rest periods were also assessed (Wilcoxon matched pairs tests).
All statistical analyses were conducted using STATISTICA 7.0 (StatSoft, Tulsa, Oklahoma, USA) and a significance level was used ($p<0.05$). Normalization of the behavioural and physiological data relative to baseline further ensured that differences in responses were the result of the experimental condition and not a consequence of day-to-day changes in the health status (e.g. pain, medication and lack of sleep) of the participants.

4.3. RESULTS

Participant 1

Results of the Wilcoxon matched pairs tests showed that the presence of the therapy dog was associated with a significant decrease from baseline in the duration of moaning behaviour (mean duration during baseline=15.2 sec., mean duration during period 2=1.0 sec.; Figure 1A; $Z=2.023$, $p=0.043$). The artificial multisensory stimulus, in turn, was associated with a significant decrease from baseline in the durations of moaning (mean duration during baseline=20.2 sec., mean duration during period 2=0.8 sec.; Figure 1A), frowning (mean duration during baseline=20.4 sec., mean duration during period 2=0.0 sec.; Figure 1B) and grimacing behaviours (mean duration during baseline=10.4 sec., mean duration during period 2=0.0 sec.; Figure 1C; $Z=2.023$, $p=0.043$ for all three tests). Curiously, no significant changes from baseline in the durations of any of these indices of unhappiness were observed in the presence of the caregiver. When comparing the observed changes in the duration of moaning between sessions involving the dog and those involving the artificial multisensory stimulus no significant difference was observed.

Obtained results also showed that all conditions (therapy dog, caregiver and artificial multisensory stimulus) were associated with a significant increase from baseline in the duration of smiling behaviour (therapy dog: mean duration during baseline=0.0 sec., mean duration during period 2=39.8 sec.; caregiver: mean duration during baseline=0.0 sec., mean duration during period 2=11.8 sec.; artificial multisensory stimulus: mean duration during baseline=0.0 sec., mean duration during period 2=19.2 sec.; Figure 2A; $Z=2.023$, $p=0.043$ for all three tests). When comparing the observed changes between conditions, significant differences were found (Friedman ANOVA: $\chi^2=10.00$, $df=2$, $p=0.006$; mean change associated with the presence of the dog=39.8 sec., mean change associated with the presence of the caregiver=11.8 sec., mean change associated with the presentation of the artificial multisensory stimulus=19.2 sec.). Subsequent Wilcoxon matched pairs tests showed significant differences among all conditions ($Z=2.023$, $p=0.043$ for all tests).
Figure 1: Mean durations of (A) moaning, (B) frowning, and (C) grimacing behaviours exhibited by participant 1 during baseline and period 2 of each experimental condition. Significant differences between baseline and period 2 are marked with an asterisk (*). Error bars represent standard errors. (Figure 1C is presented on the next page).
Finally results showed that participant 1 looked longer at the dog and at the caregiver than at the artificial multisensory stimulus (mean time looking at the dog=30.6 sec., mean time looking at the caregiver=23.4 sec., mean time looking at the artificial multisensory stimulus=0.0 sec; Figure 2B; $\chi^2=8.40$, $df=2$, $p=0.015$; dog versus artificial multisensory stimulus: $Z=2.023$, $p=0.043$; dog versus caregiver: $Z=0.674$, $P=0.500$; caregiver versus artificial multisensory stimulus: $Z=2.023$, $p=0.043$).

No further significant changes from baseline in the durations of the recorded behaviours were found. Also, no significant differences were found between baseline and rest periods, for all conditions.
Figure 2: Mean durations of (A) smiling behaviour exhibited by participant 1 during baseline and period 2 of each experimental condition, and (B) eye gazing behaviour exhibited during period 2 of each experimental condition. Significant differences between baseline and period 2 are marked with an asterisk (*). Error bars represent standard errors.
In respect to physiological measurements, results of the Wilcoxon matched pairs tests showed a significant decrease from baseline in the mean values of HR recorded in the presence of the dog [mean HR during baseline=120.867 beats per minute (BPM), mean HR during period 2=115.526 BPM; Z=2.341, \( p=0.019 \)] and in the presence of the caregiver (mean HR during baseline=119.425 BPM, mean HR during period 2=117.418 BPM; Z=2.162, \( p=0.030 \)) (Figure 3A). The presentation of the artificial multisensory stimulus, in contrast, was associated with no significant change in HR. When comparing the observed changes between sessions involving the dog and those involving the caregiver no significant differences were found.

Results of the Wilcoxon matched pairs tests showed a significant decrease from baseline in the mean SCL observed in the presence of the dog (mean SCL during baseline=-0.182 microsiemens, mean SCL during period 2=-0.195 microsiemens; Z=2.646, \( p=0.008 \)) and during the presentation of the artificial multisensory stimulus (mean SCL during baseline=-0.189 microsiemens, mean SCL during period 2=-0.199 microsiemens; Z=2.239, \( p=0.025 \)) (Figure 3B). The presence of the caregiver, in contrast, was associated with no significant change in SCL. When comparing the observed changes between sessions involving the dog and those involving the artificial multisensory stimulus no significant differences were found.

No significant differences were found between baselines and rest periods, in terms of both HR and SCL, for all conditions.
Figure 3: Mean values of (A) HR and (B) SCL exhibited by participant 1 during baseline and period 2 of each experimental condition. Significant differences between baseline and period 2 are marked with an asterisk (*). Error bars represent standard errors.
Participant 2

Results of the Wilcoxon matched pairs tests showed a significant decrease from baseline in the duration of body rocking behaviour in the presence of the dog (mean duration during baseline=99.8 sec., mean duration during period 2=0.0 sec.; Figure 4A) and in the presence of the caregiver (mean duration during baseline=106.0 sec., mean duration during period 2=7.2 sec; Figure 4A; Z=2.023, p=0.043 for all tests). No significant changes in the duration of these behaviours were observed during the presentation of the artificial multisensory stimulus. When comparing the observed changes in the duration of body rocking behaviour between sessions involving the dog and those involving the mother, no significant differences were found. The presentation of the artificial multisensory stimulus was associated with a significant increase from baseline in the durations of both frowning (mean duration during baseline=0.0 sec., mean duration during period 2=51.8 sec.; Figure 4B) and moaning behaviours (mean duration during baseline=2.0 sec., mean duration during period 2=43.2 sec.; Figure 4C; Z=2.023, p=0.043 for all two tests). The presence of the caregiver and that of the dog were associated with no significant changes in the duration of these behaviours.

Obtained results also showed that the presence of the dog and that of the caregiver was associated with a significant increase in the duration of smiling (therapy dog: mean duration during baseline=0.0 sec., mean duration during period 2=9.8 sec.; Z=2.023, p=0.043; caregiver: mean duration during baseline=0.0 sec., mean duration during period 2=3.2 sec.; Z=2.023, p=0.043; Figure 5A). The artificial multisensory stimulus, in contrast, was associated with no significant change from baseline in the duration of smiling. When comparing the observed changes between sessions involving the dog and those involving the caregiver significant differences were found (mean change caused by the presence of the dog=9.8 sec., mean change caused by the presence of the caregiver=3.2 sec; Z=1.753, p=0.080).
Figure 4: Mean durations of (A) body rocking, (B) frowning, and (C) moaning behaviours exhibited by participant 2 during baseline and period 2 of each experimental condition. Significant differences between baseline and period 2 are marked with an asterisk (*). Error bars represent standard errors. (Figure 4C is presented on the next page).
Participant 2 looked longer at the dog than at the caregiver and the artificial multisensory stimulus (mean time looking at the dog=91.0 sec., mean time looking at the caregiver=19.4 sec., mean time looking at the artificial multisensory stimulus=10.0 sec.; \( \chi^2=8.40, df=2, p=0.015 \); dog versus artificial multisensory stimulus \( Z=2.023, p=0.043 \); dog versus caregiver: \( Z=2.023, p=0.043 \); caregiver versus artificial multisensory stimulus: \( Z=1.483, p=0.138 \); Figure 5B). Contrastingly, this participant avoided the artificial multisensory stimulus for longer periods of time, as shown by the durations of the behaviour ‘head away’ (therapy dog: mean duration=5 sec., caregiver: mean duration=14.2 sec., artificial multisensory stimulus: mean duration=46.4 sec; \( \chi^2=8.40, df=2, p=0.015 \); dog versus artificial multisensory stimulus: \( Z=2.023, p=0.043 \); dog versus caregiver: \( Z=1.753, p=0.080 \); caregiver versus artificial multisensory stimulus: \( Z=2.023, p=0.043 \); Figure 5C).
Figure 5: Mean durations of (A) smiling, (B) eye gazing, and (C) head away behaviours exhibited by participant 2 during baseline and period 2 of each experimental condition. Significant differences between baseline and period 2 are marked with an asterisk (*). Error bars represent standard errors. (Figure 5C is presented on next page).
Figure 5: (C) Mean durations of head away behaviours exhibited by participant 2 during baseline and period 2 of each experimental condition. Significant differences between baseline and period 2 are marked with an asterisk (*). Error bars represent standard errors.

No further significant changes from baseline in the durations of the recorded behaviours were found. Also, no significant differences were found between baselines and rest periods, for all conditions.

In respect to physiological measurements, results of the Wilcoxon matched pairs tests showed a significant decrease in HR in the presence of the dog (mean HR during baseline=91.342 BPM, mean HR during period 2=81.335; $Z=2.379$, $p=0.017$; Figure.6A). A significant increase in HR was found during the presentation of the artificial multisensory stimulus (mean HR during baseline=95.062 BPM, mean HR during period 2=114.477; $Z=3.416$, $p<0.001$; Figure 6A). When comparing the observed changes between sessions involving the dog and those involving the artificial multisensory stimulus, significant differences were found (mean change caused by the presence of the dog=-10.007 BPM, mean change caused by the presentation of the artificial multisensory stimulus=19.415 BPM; $Z=2.845$, $p=0.004$).

Results of the Wilcoxon matched pairs tests showed a significant increase from baseline in the mean SCL observed during the presentation of the artificial multisensory
stimulus (mean SCL during baseline=-0.190, mean SCL during period 2=-0.183; Figure 6B; Z=2.619, p=0.009).

Figure 6: Mean values of (A) HR and (B) SCL exhibited by participant 2 during baseline and period 2 of each experimental condition. Significant differences between baseline and period 2 are marked with an asterisk (*). Error bars represent standard errors.
Finally, no significant differences were found between baselines and rest periods, in terms of both HR and SCL, for all conditions.

4.4. DISCUSSION

The current study is the first to quantify the behavioural and physiological responses of children with PIMD to the presence of a certified therapy dog. Obtained results seem to suggest an apparent positive responsiveness to the animal for both participants. The significant increase in the duration of smiling behaviour exhibited by participant 1 and participant 2, together with the significant decrease in the durations of moaning, frowning and grimacing exhibited by participant 1, suggest a feeling of pleasure and contentment associated with the presence of the dog. The fact that, when compared to the caregiver and the artificial multisensory stimulus, the dog induced similar, or even more pronounced, changes in the durations of the referred behaviours (smiling, grimacing, frowning and moaning) strongly highlights its potential as a novel pleasant stimulus.

Although happiness is a rather complex construct that involves multiple components (Lancioni et al. 2005), the present results do, at least, point to a happier mood (in relation to baseline) and give weight to the idea that dogs could be considered as a potential stimulus to be included in leisure activities for individuals with PIMD. According to Lancioni and colleagues (2005), problems in finding highly pleasurable stimuli are at the basis of some of the failures occurring in structured stimulation sessions, which do not increase the indices of happiness of individuals with PIMD.

The sustained looks that both participants gave at the animal (similar in duration or significantly longer than those directed at the caregiver and at the artificial multisensory stimulus) evidence its value as an attention-getting stimulus. The significant decrease in HR experienced by the participants in the presence of the dog, suggests, in turn, an apparent calming effect that is of particular importance in that it may help individuals to engage with their immediate environment, including, for example, peers and adults (see Friesen 2010). A calming effect was particularly evident in the case of participant 2 who exhibited a significant decrease in the duration of stereotyped body rocking in the presence of the dog. Note here that this behavioural change may have serious clinical implications in that the presence of stereotyped behaviour in the repertoire of individuals with PIMD can interfere with their (already limited) learning opportunities and can become so intrusive that it affects their ability to attend and interact with the environment (Lancioni et al. 2009; Patterson et al. 2009). The fact that this participant seemed to relax in the presence of the dog is particularly interesting. Based on the tactile defensiveness of this child, one could be expecting a negative reaction to the touch of the animal (as for
example when the dog licked his face or feet) similar to that exhibited during the presentation of the artificial multisensory stimulus. Although no conclusive proof is available, such contrast in responsiveness could have been due to the so-called ‘contact comfort’ provided by the dog (see Jorgenson 1997).

What about maintenance in time of the observed changes, in both behaviour and physiology? Obtained results showed no significant differences between baseline and rest periods, thus showing a rapid ‘erosion’ of the changes induced by the presence of the dog (note here that the same situation occurred with the caregiver and the artificial multisensory stimulus). Although no conclusive assertions can be made without further investigations, one can at least entertain the hypothesis that the reduced time period of each experimental condition (3 minutes) may have hampered the retention of changes over time.

According to Vlaskamp and colleagues (2007), when planning activities for people with PIMD, detailed knowledge of each individual’s idiosyncratic responses to the elements involved is crucial. Stimuli that result in avoidance behaviours (as the multisensory did for participant 2) have to be removed from the individual’s environment to potentially make it less aversive or unpleasant. Contrastingly, items that induce more positive responses, such as the dog in the present study, could be used as reinforcing stimuli, for example in training programs, or provided to the person non-contingently during the day to help make his/her routine environment more pleasant (Green & Reid 1996). Practical implications of decision-making however, have always to be discussed in terms of the time cost for implementation of activity programs, their suitability within the person’s daily context and the economical cost of the materials required (Lancioni et al. 2005). Because dog-assisted interventions can be rapidly implemented and are, generally, completely run by volunteers (certified handlers and their trained dogs) they are cost-effective and, as highlighted by DeCourcey and colleagues (2010), provide the community with opportunities to serve and get involved in public health. Certified therapy dogs have the potential to work safely and comfortably within a domestic or institutionalized setting without representing any risk of infection or injury. One has, however, to keep in mind that such interventions may not be appropriate (or pleasant) for all individuals with PIMD (see DeCourcey 2010).

The sample size of this study is very limited and the reduced number of sessions recommends caution in interpreting the presented data and drawing conclusions or making extensive generalizations. Nevertheless, obtained results offer an important platform to support further investigations of the potential of dogs as pleasant stimuli, promoters of emotional well-being in individuals with PIMD.
4.5. ETHICAL CONSIDERATIONS

The experimental protocol was approved by the Ethics Committee of the O’Porto University (Portugal). For security, and to avoid risks from exposure to the certified therapy dog, only children who had no allergies to animals, no prior traumatic incidence with animals, and no symptomatic immunosuppression were eligible for inclusion in the present study. Previously to participation, parents received written and spoken information about the aims and content of the investigation. Also they were informed about their rights in accepting the participation of their child and their right to later withdraw at any time with no adverse consequences to the participants (in such a case, all collected data would have to be destroyed). All procedures regarding the participants’ name and confidential information are in compliance with Helsinki Declaration. The findings from this research should be disseminated in a form that is accurate and clinically useful to the physicians, health care providers, participants, families and to the society at large.

4.6. ACKNOWLEDGEMENTS

We would like to thank the participants as well as their families who so generously volunteered their time to participate in this study. Also we thank Mariana Filipe for videotaping all the sessions and the members of Ânimas, especially Sebastião Castro Lemos and Bruna. Fundação para a Ciência e a Tecnologia funded the participation of Mariely Lima (FCT-SFRH/BD/44748/2008), Karine Silva (FCT SFRH/BPD/37017/2007), and Ana Magalhães (FCT-SFRH/BPD/19200/2004).

4.7. REFERENCES


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Finding an ally: can dogs help therapists to promote communication in individuals with profound intellectual and multiple disabilities?

*Personal view*

Lima, M., Silva, K., Amaral, I., & de Sousa, L.

*Submitted to Journal of Alternative and Complementary Medicine*
Dog-assisted interventions have been practiced for many years and there is now an increasing body of research demonstrating their efficacy, namely to enhance communication skills in a number of populations [see studies thoroughly reviewed elsewhere; e.g., Nimer & Lundhal (2007)]. In face of this evidence, academics and clinicians involved in the research and implementation of effective communication programs for persons with profound intellectual and multiple disabilities (PIMD) cannot be but impressed with the dearth of empirical studies on these individuals. Aside from Heimlich (2001), investigators have not yet focused on the potential of dogs as allies to help promote communication in individuals with PIMD. In this personal view article we argue that it is crucial to prioritize research on dog-assisted interventions for these persons as to keep up with the advances in scientific knowledge that have already beneficiate the functional communication training of individuals with a variety of diagnoses.

Individuals with PIMD have little or no apparent understanding of verbal language and they primarily communicate in a non- or pre-symbolic way via an unconventional and idiosyncratic repertoire of behaviours, including body movements, facial expressions, vocalizations and changes in muscle tone (Hostyn et al. 2010). The fact that these communicative signals are person- and context bounded poses a real challenge to caregivers in assigning meaning to them. This often results in lack of consistent responses from caregivers, a lowering of their expectations and reduced efforts in engineering opportunities for communicative interactions. Many individuals with severe difficulties in producing readable (i.e., consistent and predictable) signals, therefore, are unrewarded for all their efforts and may fail to develop a full range of communicative functions (Forster & Iacono 2008). Also they are left with a feeling of helplessness and incompetence, with the potential that passivity, lack of interest, frustration, boredom and/or challenging behaviours will rapidly develop and hinder the acquisition of communicative skills. Accordingly, studies show that the time persons with PIMD spend in awake active-alert states (during which they are most likely to engage in joint attention and successful communication interactions) is very low during typical everyday life (e.g., Roberts et al. 2005). On contrast, these individuals spend long periods of time in state behaviours that impede them to interact and communicate (e.g., challenging behaviours and drowsy, daze and sleep behaviours).

How could dogs help therapists to promote communication in individuals with PIMD? We feel that several research efforts have to be made if we are to gain a complete understanding on the potential of dog-assisted interventions.

We propose that, in a preliminary stage, studies could be directed at assessing whether dogs – as stimuli providing multisensory stimulation in a unique interactive style -
can be pleasant, attention getting and relaxing stimuli with which these individuals can positively engage. Such attributes are important in that they could prime individuals for communication as already demonstrated in a number of studies in, for example, children with autism spectrum disorders (e.g., Martin & Farnum, 2002; Silva et al. 2011), people with dementia (e.g., Mano et al. 2003; Buettner et al. 2010), and individuals with aphasia (e.g., Macauley 2006; LaFrance et al. 2007). Also the so-called ‘contact comfort’ provided by the touch of a dog was found to have dearousing effects in individuals with different diagnoses who spend long periods of time in agitated states with stereotyped behaviours (note that, among other benefits, dearousing can enhance coordination of movements necessary for producing readable communicative signals) (e.g., De Courcey 2010; Tsai et al. 2010).

In a following stage, studies should be designed to determine whether dogs consistently represent preferences (in comparison for instance with inanimate objects or even peers and caregivers). Our research group recently conducted a first pilot study aimed at assessing and comparing the behavioural and physiological responses of two children with PIMD to a therapy dog, a familiar caregiver and an artificial multisensory stimulus; interestingly, results showed that both participants responded positively to the presence of the dog, in much the same way as they responded to the caregiver (Lima et al. submitted for publication). It is known that preferred stimuli are likely to function as reinforcers when applied contingently in skill training programs for profoundly handicapped persons (Logan et al. 2001). As such, we feel that it would also be interesting to evaluate the potential of dogs as reinforcers when teaching cause-effect skills (which are at the basis of the development of different communication functions) to individuals with PIMD. According to learning theory, a communicative sign that generates a pleasurable response will be reinforced and thus will be more likely to occur in the future. Unpleasant or anxiety-provoking responses, in contrast, may result in avoidance or withdrawal behaviour [see Kruger & Serpell (2006)]. In this extent, one can entertain the hypothesis that the unambiguous behaviours that trained dogs can exhibit as contingent responses to potential communicative signals (e.g., barking, jumping, licking, rolling) may be highly motivating and satisfying for, at least, certain individuals with PIMD. By consequence, dogs’ responses may help them to learn that some elements of the environment may be responsive and controllable. Also, individuals with PIMD may profit, from the multiple opportunities that naturally occur in the presence of the dog, to practice alternative modes of behaviour (contrasting for instance to the loud, erratic, aggressive or unpredictable manifestations that are common elements of their behavioural). Finally, another interesting question to be addressed by future empirical studies relates to the effects of the presence of dogs on the mood, attitude and sensitivity of the therapists.
during functional communication training programs. According to Kruger and Serpell (2006), positive effects may well improve the development and maintenance of a working alliance between the individual with PIMD and the therapist, throughout all stages of the intervention.

For the above proposed studies to be sound, we consider that there is a need to use rigorous scientific protocols including clear definitions of the goals of the research, proper instruments to measure and document specific outcome variables, gathering of sufficient sample sizes and the use of a control condition (as highlighted in Martin & Farnum, 2002). It is imperative that protocols are as detailed as possible as to allow for the replicability of the study. Also, it is important that researchers present extensive descriptions of the site population(s) from which their study samples are drawn so that practitioners and other researchers can determine the similarity with their own facilities (Wilson & Barker 2003).

It is our belief that future research will highlight important positive aspects of dog-assisted interventions for improving communication in individuals with PIMD. Indeed, clinically speaking, we share Kruger and Serpell’s view (2006) in that it may be hard to imagine a better combination of attributes: a tool that can have the potential to simultaneously alert, engage, prime and relax individuals with such profound levels of disabilities. As this article is definitely a personal view, we would like to conclude by alerting to the fact that assessing evidence that dog-assisted interventions may help promote communication in individuals with PIMD will not be sufficient to make the decision about their implementation. Will caregivers be interested in these interventions? What are the full costs per unit of expected outcomes? Will the intervention broadly cover the PIMD spectrum? Will any difference in individual characteristics impact the intervention effectiveness? As to transfer evidence to practice it will be fundamental to evaluate the applicability and generalizability of research findings by considering these, and other, questions related to political and social acceptability, resource availability (personal and financial), organizational expertise and capacity, magnitude of the ‘reach’ effectiveness of the intervention and target population characteristics.

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1. Overview of the results

This section aims to present a summary of the results obtained in the experimental studies described in Chapters 2, 3 and 4.

In Chapter 2, a pilot study was presented with the aim of describing the behavioural and physiological responsiveness of one child with PIMD to 19 sensory stimuli commonly used by clinicians during sensory interventions. Behavioural responsiveness was evaluated in terms of consistent motor reactions. Physiological responsiveness was assessed in terms of consistent electrodermal reactions and consistent changes from baseline in heart rate. Results of this study showed that the child only exhibited consistent observable motor reactions to three of the presented items. He did show, however, consistent physiological reactions to a considerably higher number of stimuli (N=10), suggesting that, even when they seem largely unresponsive in terms of observable behaviours, individuals with PIMD may react to stimulation.

The study presented in Chapter 3 aimed at extending the findings presented in Chapter 2 to three additional children with PIMD, using a similar, although slightly modified, experimental protocol. Obtained results corroborated those reported in Chapter 2 in that all participants exhibited only rare consistent behavioural reactions to the presented items, but showed a number of consistent physiological ones. Interestingly, it was possible to identify biphasic changes in heart rate in response to several items, pointing, according to several theoretical models, to the possibility of different appraisal processes, namely in respect to novelty and pleasantness of sensory stimuli (Ellsworth & Scherer, 2003; Anttonen & Surakka, 2005; Vila et al., 2007; Scherer, 2009; Delplanque et al., 2009).

The objective of the study presented in Chapter 4 was to analyse the responsiveness of two children with PIMD to a certified dog, using a combination of behavioural and physiological measurements, as in the previous Chapters. As to better evidence the potential benefits of dogs, responsiveness to a familiar caregiver and to a multisensory device were also assessed. Interestingly, both participants appeared to respond positively to the animal, in much the same way as they responded to the familiar caregiver. This study provided the first quantitative behavioural and physiological indications that one could consider the possibility of using dogs in interventions for people with PIMD, namely as pleasant, relaxing and attention getting stimuli with which these individuals could engage.
2. Behavioural and physiological responsiveness of individuals with PIMD (Chapters 2 and 3)

2.1 Methodological issues

A number of methodological issues are worth considering before thoroughly discussing the results presented in this thesis. As this was the first time that responsiveness of individuals with PIMD to stimulation was assessed by means of both behavioural and physiological measurements, there were no previous studies, serving as a starting point, to help optimize the experimental protocol. A pilot study (Chapter 2), therefore, had to be conducted. The experience gained from this first study and the theoretical support provided by both scientific literature and scientific discussions with researchers working in the field of psychophysiology were at the basis of the methodological modifications introduced in the study presented in Chapter 3.

In Chapter 2, the motor reactions of a child, with extremely severe neuromotor impairments and a very limited behavioural repertoire (see Table 2), were recorded during 5 seconds, and for 20 seconds following the presentation of each stimulus. The participants in the study presented in Chapter 3 had less motor impairments and could produce a higher number of behavioural reactions (see Table 2). As such, the period of data collection following stimulus presentation was shortened to 5 seconds, as to reduce the probability of recording behaviours that were not specific reactions to the presented items. A similar change was made regarding heart rate measurements. In the study presented in Chapter 2, heart rate was recorded during two consecutive periods: during stimulus presentation (5 seconds) and following the presentation of each stimulus (20 seconds). In the study presented in Chapter 3, and as for behavioural data, heart rate was recorded during (5 seconds) and following the presentation of each stimulus (only for 5 seconds). This decision was made with the aim of identifying biphasic changes in heart rate, pointing to different appraisal processes. Recent studies (e.g. Vila et al., 2007; Delplanque et al., 2009) have shown that appraisal processes and their efferent peripheral effects occur in sequential order. More specifically, studies have found evidence of temporal priority of stimulus novelty evaluation over stimulus pleasantness evaluation on cardiac activity, with the first being generally observed about 2-4 seconds after stimulus onset, while the latter commonly occurs about 5-10 seconds after stimulus onset (e.g., Delplanque et al., 2009).

Two other differences can be detected in the methodologies followed in the studies presented in Chapter 2 and 3, namely regarding the number of experimental sessions and the items presented to the participants. In the study described in Chapter 2, although 5
sessions were initially planned, only 3 were actually conducted because the participant had to be hospitalized for a long period of time. Also the Multi-Sensory Center device presented to this child was no longer used due to the apparent magnitude of discomfort that it induced, during a preliminary session, to one of the three children that participated in the study presented in Chapter 3. Ethical obligations, thus, were at the basis of the decision to remove this item from the list of stimuli to be presented to the participants in that study (Chapter 3).

Another methodological issue that is worth discussing relates to data analysis and data presentation. The reduced number of experimental sessions (N=3) conducted in the study described in Chapter 2 rendered the use of statistical tests meaningless, since no significant findings could ever be obtained. Given that the purpose of the pilot study was to explore the potential clinical implications of combining behavioural and physiological measurements to assess the sensory responsiveness of children with PIMD, the decision was made to present a detailed description of the consistent reactions of the participant to the presented items. Looking only for statistically significant results, in a study with only one participant, and with such a reduced number of sessions, could have lead to an underestimation of the actual responsiveness of the participant, and by consequence, of the true potential of the method employed for its assessment. Although larger (N=5), the number of experimental sessions conducted in the study presented in Chapter 3 also undermined the possibility of unveiling significant differences from baseline in the frequencies and durations of the behaviours recorded during and following stimulation. As such, the decision to focus on consistent behavioural reactions was maintained. The statistical analysis of heart rate variation, in contrast, could (and did) produce significant results.

Finally, it is important to mention that only a small number of subjects were able to participate in the studies presented here due to both the relatively small group of 3 to 6 year-old children with PIMD in Portugal, and to the need to exclude individuals with epilepsy as to avoid the elicitation of seizures (Takenouchi, Yap, Engel, &Perlman, 2010). Also, only single-case studies are presented because the variety of co-morbidities of the participants did not allow for data analyses to be conducted at a group level. Note that single-subject methods are particularly well suited to study individuals with PIMD, whose characteristics and life circumstances are exceedingly heterogeneous in nature. This heterogeneity creates substantial problems when researchers attempt to use group-based methodologies to address questions about the effectiveness of specific interventions. In fact, group-based studies designed to determine the effectiveness of interventions on individuals with PIMD are in the minority; there exists a much more substantial body of
research using primarily single-case design methods (e.g., Petry & Maes, 2006; Lancioni et al., 2010a,b).

2.2 Physiological responsiveness: consistency and possible interpretations

As highlighted in the Introduction section of this thesis (Chapter 1), it is extremely difficult to assess the responsiveness of individuals with PIMD and the hedonic value of the stimuli that are presented to them. According to several authors, this poses a huge problem in practice as it leaves caregivers “in the dark” about the most suitable stimuli to be included (and about those to be avoided) in interventions aimed at improving the emotional well-being of these people (e.g., Carnaby, 2007; Vlaskamp & Cuppen-Fonteine, 2007; Vlaskamp, Hiemstra, & Wiersma, 2007; Brinkman, 2009).

Data obtained in the studies presented in Chapter 2 and 3 highlight the fact that considering behavioural reactions alone may not be sufficient for an accurate assessment of the responsiveness of individuals with PIMD. Clearly, stimuli that may not induce any consistent behavioural reactions can elicit consistent physiological ones, which may point out to improved or reduced emotional well-being. However, when commenting on the importance of supplementing behavioural observations with additional measurement techniques, Vlaskamp et al. (2007) argue that the physiological reactions of individuals with PIMD may not be sufficiently consistent as to elucidate on the hedonic value of stimuli. An interesting aspect of the data reported in Chapter 2 and 3 is that they seem to suggest the opposite. Although participants have very different characteristics (see their detailed descriptions presented in the Methods section of Chapter 2 and 3), they all showed the same pattern of responsiveness: a number of physiological reactions that were consistent across all experimental sessions (i.e., either during and/or following stimulus presentation), but only rare behavioural ones.

When interpreting the obtained, consistent, physiological reactions in light of appraisal theories (Scherer, 1984, 1999, 2001, 2009; Ellsworth & Scherer, 2003; Delplanque et al., 2009), a number of different stimuli can be identified as relevant, arousing, and potentially pleasant or unpleasant for each participant. To aid the comprehension of the reader and to allow interpretation of the data presented in Chapters 2 and 3, Table 1 shows a synthetic recapitulation of the response patterning associated to the evaluation stimuli as novel and pleasant/or unpleasant [following Scherer (2009)].
Table 1: Response patterning associated to stimuli processed as novel and pleasant/unpleasant [following Turpin (1986) and Scherer (2009)].

<table>
<thead>
<tr>
<th>Evaluation</th>
<th>Stimulus features</th>
<th>Response patterning</th>
</tr>
</thead>
<tbody>
<tr>
<td>Novelty evaluation</td>
<td>Novel, low to moderate intensity, stimulus</td>
<td>Heart rate deceleration, skin conductance reactions, EEG alpha changes, modulation of the P3a in ERPs, digital vasoconstriction, cephalic vasodilatation, local muscle tonus changes, brows and lids up, frown, jaw drop, gaze directed, interruption of speech and action, raising head.</td>
</tr>
<tr>
<td></td>
<td>November, abrupt, high intensity, stimulus</td>
<td>Heart rate acceleration, skin conductance reactions, reflexive eye blink, digital vasoconstriction, cephalic vasoconstriction, local muscle tonus changes, interruption of speech and action.</td>
</tr>
<tr>
<td>Pleasantness evaluation</td>
<td>Pleasant stimulus</td>
<td>Heart rate deceleration, skin conductance reactions, inhalation, salivation, pupillary dilatation, lids up, open mouth and nostrils, lips part and corners pulled upwards, gaze directed, pharyngeal expansion, vocal tract shortened and relaxation of tract walls, centripetal hand and arm movements, expanding posture, approach locomotion.</td>
</tr>
<tr>
<td></td>
<td>Unpleasant stimulus</td>
<td>Heart rate acceleration, skin conductance reactions, decrease in salivation, pupillary constriction, slight muscle tonus increase, brow lowering, lid tightening, eye closing, nose wrinkling, lip corner depression, chin raise, lip press, nostril compression, tongue thrust, gaze aversion, faucal and pharyngeal constriction, vocal tract shortened and tensing of tract walls, centrifugal hand and arm movements, shrinking posture, avoidance locomotion.</td>
</tr>
</tbody>
</table>

Interestingly, when looking at the consistent, physiological reactions that occurred simultaneously with consistent behavioural ones, an interesting pattern was observed that is in agreement with the response patterning presented in Table 1. Behavioural reactions indicative of discomfort were always accompanied by heart rate accelerations, while those indicative of pleasantness were repeatedly accompanied by heart rate deceleration, thus suggesting that consistent physiological reactions of individuals with PIMD may have the potential to elucidate on the hedonic value of the stimuli that are presented to them.

Although, in the psychophysiological tradition, it already exists an extensive amount of research on the relationship between physiological parameters and emotional well-being (e.g., Hamm, Schupp, & Weike, 2003; Bradley & Lang, 2007; Larsen, Berntson, Poehlman, Ito, & Cacciopo, 2008), until now, and apart from the studies included in this thesis, there has been only one study investigating this connection in people with PIMD. With the aim of identifying the psychophysiological correlates of pleasantness and unpleasantness in people with PIMD, Vos, De Cock, Petry, Noortgate, and Maes, (2010) conducted a study in which they measured the behavioural and physiological reactions (including electrodermal reactions and changes in heart rate) of three individuals with
PIMD towards positive and negative stimuli situations (e.g., a positive stimulus presented during a group activity and a negative stimulus presented when the participant was eating). Curiously, and although the authors expected that heart rate would be higher during negative situations than positive ones, an inverse pattern was found. As a possible explanation for this unexpected result, Vos et al. (2010), pointed to an attention regulation process. That is, these authors claimed that, when confronted with an unpleasant situation, the participants in their study, who could not use fight or flight behaviour, could have tried to block the negative stimulus by giving it less attention, which resulted in less arousal and in heart rate deceleration. Given that the stimuli situations (both pleasant and unpleasant ones) used in their study were selected on the basis of staff's opinions, the authors also raised the hypothesis that lower heart rates were associated with so-called ‘unpleasant’ situations because of wrong interpretations on the part of the staff. That is, the situations selected might not have elicited the intended emotions. This hypothesis raised by Vos et al. (2010) clearly highlights the extreme difficulty that caregivers have in identifying the hedonic value of sensory stimuli to be presented to individuals with PIMD (that is, in identifying stimuli that may be pleasant and unpleasant for these people). Moreover, it stresses the need for considering non-subjective measures of their emotional well-being.

2.3 Clinical implications and applicability of physiological measurements

A major challenge for caregivers and investigators working with individuals with PIMD is to find an easy to use tool for an accurate assessment of the sensory responsiveness and emotional well-being of these individuals. According to Petry and Maes (2006), combining multiple strategies seems to be the most appropriate approach for examining internal states of individuals with limited communicative skills; an idea that is consistent with the notion of “methodological pluralism” advocated by Schalock et al. (2000), as well as Reid and Green (2002).

The apparent benefits of complementing behavioural observations with physiological measurements seem twofold. It allows caregivers to have a more accurate insight into i) the actual sensory responsiveness of individuals with PIMD (even if not showing consistent behavioural reactions to environmental stimuli, these individuals may be able to react physiologically in a consistent way) and ii) the potential appraisals (novelty and pleasantness) that people with PIMD seem able to make of the various stimuli that are presented to them.

Drafting a profile of the ways a person with PIMD reacts to and appears to appraise (in terms of novelty and pleasantness) different stimuli may be an important
aspect of the quality of support for this group, considering that typical responsiveness of a
given person is often not known at the moment of planning stimulation interventions
(Zijlstra, 2003). Information obtained from physiological assessments should, therefore, be
part of the support plan for each person with PIMD, as it will allow caregivers to assure
that what is presented to each individual with PIMD is positive, that is, personally relevant,
stimulating, attention-getting and not aversive nor unpleasant (Carnaby, 2007). Importantly, this can lead to a growing feeling of competency in caregivers.

Stimuli identified as pleasant can be used in a number of interventions, being
particularly important in those requiring the use of reinforcing items for training purposes.
The actual reinforcing effects of these stimuli have to be tested by making them contingent upon a specific desirable behaviour and recording whether the behaviour increases or decreases in frequency or duration (Logan et al., 2001). Even if positive stimuli end up by not having a reinforcing value in a training program, they can be provided to an individual with PIMD non-contingently, during the day, for basic sensory stimulation purposes and to help make his/her routine environment more pleasant. Note here that the social environment (e.g., caregivers establishing eye contact and providing positive comments) that accompanies stimuli presentation is of great importance in that the hedonic value of the presented items, and their alertness effects, can be highly strengthened when systematically combined with social interactions (Petry & Maes, 2007).

One may question how affordable and practical is collecting physiological data
such as the one recorded in the present thesis (heart rate and electrodermal activity). Indeed, the main criticisms pointed at this method relate to the fact that specific equipment may be expensive and difficult to use in practice, requiring specialized technicians (Vlaskamp et al., 2007). Low cost, and easy-to-use, heart rate and electrodermal analyses systems have, however, recently been designed and implemented, in several countries, to support clinical and educational interventions, namely for individuals with PIMD (e.g., Kobayashi, Nunokawa, & Ooe, 2010). Moreover, the value of the information that may be obtained through these methods, and the undeniable implications for the emotional well-being of people with PIMD, are such that it fully justifies the potential ‘trouble’ of setting up the hardware and software.

3. Dogs as potential pleasant and engaging stimuli (Chapters 4 and 5)

The study presented in Chapter 4 is innovative in that it reports, for the very first time in
children with PIMD, a combination of behavioural and physiological reactions to the
presence of a certified dog. Obtained results are very interesting in that they evidence a
positive response pattern, similar to that reported for a number of other populations (e.g.,
The presence of the dog was associated with a significant increase, from baseline, in the duration of smiling behaviour along with a significant reduction in the duration of facial expressions indicating some type of discomfort or in the duration of stereotyped body rocking. Also, the dog seemed to attract the attention of both participants, as shown by their direct gazes. Positive physiological effects were also associated with the dog, namely a reduction, from baseline, of heart rate during the presentation of the animal. A significant decrease in skin conductance level was also noted in one participant. Curiously, no differences from baseline, in both behaviour and physiology, were observed following the presentation of the dog. In another words, it seems that the positive behavioural and physiological changes elicited by the animal did not maintain over time - possibly because the reduced time of presentation undermined the retention of changes.

### 3.1 Theoretical models underlying the positive effects of dogs

How to explain the apparent positive effects of the dog on the two children with PIMD that participated in the study presented in Chapter 4? The field of dog-assisted interventions still lacks today a unified, widely accepted, and empirically supported theoretical framework for explaining how and why contact with dogs can have positive effects on human health (Kruger & Serpell, 2006). Nevertheless, a considerable variety of theories can be found in the literature, most of which focus on the supposedly unique intrinsic attributes of dogs.

The simple fact that dogs are a live stimulus may, according to the "biophilia hypothesis" proposed by Wilson (1984), help explaining why their presence, and interaction with them, can produce calming effects in humans (as those reported in Chapter 4). What this theory asserts is that human beings have a genetically based need and propensity to affiliate with other living organisms [for an extensive discussion on this hypothesis see Kahn (1997)]. Although a handful of animal species have come into close contact with humans through the process of domestication, dogs, as a highly social species (Savolainen, Zhang, Luo, Lundeberg, & Leitner, 2002; Hare & Tomasello, 2005), have established an unique ‘social symbiotic relationship’ with humans over the last 15,000-20,000 years (Hare et al., 2010; Miklosi, Topál, & Csányi, 2004). According to many authors (e.g., Odendaal, 2000; Odendaal & Meintjes, 2003) at the basis of this relationship is a two-way fulfilling of attentionis egens (i.e., a need for attention that has been clearly identified in well-developed social systems, as a universal emotional need), which has already been supported by physiological data. Studies analysing the
physiological correlates of dog-human physical and visual contact found a significant increase in both species in the concentrations of hormones indicative of positive affiliation interactions (e.g., Odendaal & Lehmann, 2000; Nagasawa, Kikusui, Onaka, & Ohta, 2009; Nagasawa, Moji, & Kikusui, 2009). In this regard, it has been argued that typical positive attention between human and human could be replaced by human-animal interaction, and that, when there is an additional need for attentionis egens, as in the case of individuals with physical and intellectual disabilities, dogs could assist in interventions as therapeutic agents (Odendaal, 2000; Odendaal & Lehmann, 2000).

Another intrinsic feature of dogs that can also contribute to understanding the potential value of incorporating these animals into therapeutic contexts relates to their apparent ability for emotional synchronization with humans (e.g., Paxton, 2000; Schleidt & Shalter, 2003; Topal et al. 2009; Silva & de Sousa, 2011). Although empirical evidence is yet very limited, studies conducted so far have found that dogs can show emotional synchronization by attending to various visual or acoustic social signals emitted by humans. For example, a study showed that during human-dog play, play signals emitted by one element (dog or human) seemed to have the potential to evoke play behaviour from the other, probably through synchronization of emotional states (Rooney, Bradshaw, & Robinson, 2001).

3.2. Possible routes to further explore the potential of dogs

Additional studies, with larger samples, are needed to further extend the findings presented here and to determine whether dogs can indeed, as suggested, be positive, attention getting stimuli with which individuals with PIMD can engage. In that case, one particularly interesting line of investigation could be to assess whether the intrinsic attributes of dogs can be empowered by interaction with other factors that are known to also affect the behavioural responsiveness of individuals with PIMD, namely personal attributes of caregivers and environmental attributes. Also, it could be interesting to study whether longer periods of presentation of the animal could yield different, and/or more consistent results over time. As a next step, research should focus on assessing the potential benefits of introducing dogs into planned animal assisted activities and therapy programs with very specific goals and purposes, namely to promote communication in individuals with PIMD (see the discussion presented in Chapter 5). Positive benefits of dog assisted interventions in the field of communication have already been reported in a number of studies with individuals with different diagnoses. For example, Walters and colleagues (2006) found that the presence of a dog can increase communication between a teacher and children with profound developmental disabilities. These authors analysed the
effects of the presence of a dog on the communication patterns between three children with developmental disabilities and their teacher at an elementary school. They found that all participants demonstrated an increase in overall verbal and non-verbal positive initiated behaviours toward both the teacher and the dog (e.g., smiling, nodding head, answering questions when asked, requesting help). As another example, Kovacs et al. (2006) analysed the effects of dog-assisted therapy in the communicative skills of severely disabled chronic schizophrenic patients, and found significant improvements in various domains of nonverbal communication. Similarly, LaFrance (2007) explored the impact of the presence of a dog on the overt social-verbal and social nonverbal communication skills of a sole participant with aphasia and found significant improvements related to both.

Independently of the type of intervention (activities or therapy programs) to be evaluated by future research, a number of commitments have to be made, regarding the safety of the participants and that of the animal. Certified dogs have to be at least one year old, up to date on all vaccinations and must have annual health and behavioural examinations. The certification process should include an evaluation of the dogs’ temperament and the quality of the interaction between dogs and handlers when exposed to a variety of stressful situations. Any displays of aggression, distraction, and nervousness immediately have to disqualify a dog for research/intervention purposes (Sockalingam, 2008). Note that, in Portugal, Ânimas is the only certified organization, member of the Assistance Dogs International Inc. (ADI), which is dedicated to improving human health and well-being by fostering human-animal interaction. It provides standards of excellence and training to therapy dog handlers and dogs, and guarantees a high level of certification. Also, it is important to note that, just as not every dog is a suitable partner for dog-assisted interventions, so too, not every individual with PIMD is an appropriate candidate for this type of intervention. For example, individuals with allergies or asthma, or those prone to seizures due to high levels of excitement must be excluded from these interventions/studies. Individuals who have had a bad experience with dogs or exhibit aggressive behaviour must, likewise, be excluded for the safety of both the animal and the person. Finally, consideration of the dogs’ health and welfare is another important ethical issue which must not be overlooked by future research. The animals need to be closely monitored for physical signs of discomfort, stress and fear, throughout all stages of future experimental studies.

4. Final considerations

Even though one has to be very cautious in interpreting the findings reported here, and although future research is crucial for conclusive inferences to be made, this thesis has
brought forth two new lines in the study of sensory stimulation of children with PIMD: i) the investigation of the physiological responsiveness to stimulation as a potential indicator of appraisal processes regarding the novelty and pleasantness of sensory stimuli and ii) the investigation of the potential effects of dogs on individuals with PIMD, as stimuli providing visual, auditory, tactile and olfactory stimulation in a unique interactive style.

In respect to the first line of investigation, the significance of the results presented here should be highlighted when considering the extreme difficulty to assess objectively the sensory responsiveness of individuals with PIMD as to find pleasant stimuli to be presented to these individuals. The line of investigation regarding the use of dogs as a potential source of emotional well-being for people with PIMD, in turn, not only extends existing literature, but it also sets a preliminary basis supporting the inclusion of dogs in interventions for these individuals. Importantly, the full potential of such dog-assisted practices, namely for promoting communication, has yet to be empirically exploited, but hopefully will be explored by future studies.

Finally, it seems crucial that academic institutions promote research in the fields explored here and that results are disseminated among health and educative professionals working with individuals with PIMD. Indeed, serving the public good, which is why publicly funded research is supported, is possible if research results are widely disseminated. Also, established conclusions in research studies could (and should) be part of the academic curriculums of health and social science students so that they will reach an effective training that encompasses the most recent developments in research on individuals with PIMD. In parallel, researchers should collaborate closely with public health and policy makers in order to enable them to better channel scarce public health resources into the interventions that are more likely to guarantee the emotional well-being of individuals with PIMD.

5. References


Vlaskamp, C., Hiemstra, S. J., & Wiersma, L. A. (2007). Becoming aware of what you know or need to know: gathering client and context characteristics in day services for


Eu, Mariely Gestosa Lima, Terapeuta da Fala, portadora da Cédula Profissional nº C -012048186 encontro-me a realizar o meu Projecto de Doutoramento no Instituto de Ciências Biomédicas Abel Salazar, na Universidade do Porto.

No presente documento venho solicitar o seu consentimento para que o/a __________________ possa participar neste projecto de investigação. O objectivo deste estudo é analisar as respostas fisiológicas e comportamentais, de crianças com multideficiência, a diferentes tipos de estimulação sensorial. Este estudo implicará a apresentação de diferentes estímulos, uma vez por semana, durante 6 semanas. Dentro dos estímulos a apresentar estão incluídos:

a) estímulos inanimados auditivos, visuais, gustativos, olfactivos e tácteis,
b) estímulos animados incluindo um familiar e um cão devidamente certificado pela Associação Ânimas que estará sempre acompanhado pelo seu Instrutor.

Durante a apresentação dos estímulos, todos os comportamentos e todas as alterações fisiológicas serão registadas através de uma câmara de filmar e de um equipamento específico para o efeito. Não são conhecidos quaisquer riscos associados aos procedimentos deste estudo. Todos os dados obtidos serão usados apenas para fins de investigação, ficando guardados, durante dois anos, em suporte informático e protegidos por palavra-chave, do conhecimento único e exclusivo dos investigadores responsáveis. Após este período, os dados serão destruídos. No caso de apresentação do material video em reuniões científicas será sempre ocultada a face do(a) __________________ não tornando possível a sua identificação, a não ser que expressamente o autorize. Este estudo não representa qualquer tipo de tratamento, mas o/a __________________ poderá ser beneficiado por um melhor conhecimento do seu comportamento e reacções, que poderá fornecer a quem o/a trata indicações úteis. Além disso, este melhor conhecimento pode ser benéfico para outras crianças na mesma situação.

Em qualquer momento do estudo será possível anular este consentimento e ter acesso aos resultados até então obtidos. A participação é voluntária, não existindo despesas ou compensações financeiras relacionadas com a participação do(a) __________________ neste projecto de investigação.

Se surgir alguma questão acerca do estudo pode sempre contactar-me através do número de telefone 918157456 ou à minha orientadora, a Professora Doutora Liliana de Sousa, através do número 222062200.

Em anexo está o consentimento informado, livre e esclarecido para ser assinado, em duplicado, caso não tenha ficado qualquer dúvida.
AO REPRESENTANTE LEGAL

Por favor, leia com atenção todo o conteúdo deste documento. Não hesite em solicitar mais informações se não estiver completamente esclarecido. Se entender que o/a __________ pode participar neste estudo, então assine este documento.

Termo de Consentimento Informado, Livre e Esclarecido

Eu, ____________________________, portador do Bilhe de Indentidade Nº ________________ declarei que fui suficientemente esclarecido(a) por Mariely Lima sobre o estudo acima referido e que autorizo a participação no mesmo do/da ____________________________, de quem sou o/a representante legal.

___________________________________ Data _______/______/______
Assinatura do Representante Legal

___________________________________ Data _______/______/______
Assinatura da Investigadora Responsável

Obrigada por colaborar comigo nesta investigação.

Este projecto foi aprovado pelo Instituto de Ciências Biomédicas Abel Salazar da Universidade do Porto e ainda pela Fundação da Ciência e Tecnologia para atribuição de bolsa de doutoramento. Este projecto decorrerá sob a orientação da Professora Doutora Liliana de Sousa e co-orientação da Professora Doutora Isabel Amaral.
PARECER

Acerca do Projecto apresentado pela Licª Mariely Lima, doutoranda pelo Instituto de Ciências Biomédicas Abel Salazar

O projecto, claramente exposto no documento apresentado, tem como objectivo registar e descrever as respostas comportamentais, do pulso e da resistência cutânea a estímulos uni – e multisensoriais, em crianças com multideficiências graves.

O Projecto reúne as condições necessárias para ser avaliado, tem relevância e é orientado por investigadoras idóneas e experientes.

No que concerne à sua avaliação ética, há aspectos a considerar que devem ser integrados numa visão de conjunto. Assim, tratando-se de crianças com deficiência, duplamente vulneráveis e, que presume, incapazes de manifestar anuência a qualquer proposta, o consentimento informado terá de ser dado pelos Pais ou responsáveis, representantes legais. Esta situação é vulgar, mas leva sempre a uma cautelosa aproximação à emissão de opinião. Em segundo lugar, a investigação proposta não resultará em benefício terapêutico para as crianças. Em terceiro lugar, a fixação por filmagem das reacções comportamentais levanta problemas de respeito pela privacidade e dignidade das crianças.

Na folha informativa do consentimento informado deve constar a possibilidade de revogação e não de anulação do referido consentimento.

Estas reservas poderiam indiciar uma atitude de rejeição da investigação proposta, mas há aspectos positivos a considerar:

a) Não são de esperar quaisquer consequências negativas ou reacções adversas;

b) Poderá haver benefício para as crianças examinadas (e para outras em situação similar) por se ficar a conhecer melhor os comportamentos e reacções desencadeados por determinado tipo de estímulo, que poderá ser seleccionado para utilização ulterior, no acompanhamento da criança (por ex. um estímulo que induz uma reacção de agradar poderá ser aplicado com determinada frequência);

c) O avanço científico neste campo pode levar, eventualmente, a melhorias no tratamento e reabilitação.
d) Está garantida a privacidade e a destruição dos registos ao fim de um período razoável. Todavia, essa restrição deve ser formalmente comunicada à instituição dos investigadores.

e) É de esperar que as visitas e contactos durante 6 semanas possam ser úteis aos pais e/ou cuidadores, pelo acompanhamento e solidariedade que comportam.

A CEUP aguarda que as investigadoras lhe enviem um relatório acerca dos resultados obtidos e/ou dificuldades com relevância ética porventura surgidas durante a execução do projecto.

Nestas condições, a Comissão de Ética da Universidade do Porto dá a sua aprovação a este Projecto.

Porto, 7 de Outubro de 2010

Walter Osswald