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PERSPECTIVES IN REHABILITATION

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Perspectives on tactile intervention for children with cerebral palsy: a framework to guide clinical reasoning and future research

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ABSTRACT

Purpose: Many children with cerebral palsy (CP) are known to experience tactile impairments. Research evaluating specific interventions to manage this is, however, minimal. This paper seeks to consolidate current literature and provide a framework to help clinicians and researchers think strategically about tactile treatment selection and future research planning.

Method: The framework is described via a novel analogy – "*The Apartment Block Theory*". The theory describes the relative effectiveness of three intervention strategies aimed at overcoming a poorly responsive tactile system: (1) *Pressing the buzzer* – providing repeated passive tactile stimulation at the periphery; (2) *Sneaking in the door* – providing active tactile-enhanced motor training that capitalises on the opportunity to provide high-dose tactile input during motor interventions; and (3) *Connecting another way* – providing visually enhanced touch strategies with the aim of enhancing tactile function, which can be compared to phoning the apartment as an alternative to using the buzzer.

Results: Using this theory, the paper describes which sub-groups of children with CP may benefit from each intervention strategy when considering their capabilities in visual, motor, and attention domains.

Conclusions: This theory can assist clinicians to provide effective interventions and researchers to make informed future research decisions to optimise tactile function for children with CP.

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Tactile; sensation; touch; therapy; child; cerebral palsy

➤ IMPLICATIONS FOR REHABILITATION

- Although tactile impairments are reported to be common in children with cerebral palsy, very few successful interventions are reported in the literature.
- Visually enhanced touch is a successful strategy for treating tactile impairments in children with cerebral palsy who have sufficient vision and attention.
- Combining intentional tactile input with upper limb movement training may improve tactile function in children with cerebral palsy who have sufficient movement and attention.
- In children who have complex co-morbidities, including both visual and movement impairments, it may be necessary to consider providing passive tactile stimulation in tactile intervention.

Tactile impairment impacts on upper limb function in a significant proportion of children with cerebral palsy (CP). According to the tactile assessment framework, these deficits may occur in tactile registration and/or tactile perception (which may be spatial, temporal, or textural), and appropriate assessment items for each of these domains have been identified for children with CP [1]. It is known that over 77% of children with unilateral CP have tactile perception deficits and over 40% have concomitant registration and perception deficits [2]. These deficits in tactile registration (as measured by the Semmes Weinstein Monofilaments) and spatial tactile perception (as measured by Single Point Localisation, Double Simultaneous Stimulation, two point discrimination and stereognosis) are also significantly related to motor performance [3]. Although less research has been reported for children with other motor distributions, a significant number of children with diplegia (registration: 20%; stereognosis: 15%) and quadriplegia (registration: 32%; stereognosis: 42%) have been reported to have a deficits as well [4]. Despite this, a recent systematic review of tactile treatment strategies reported that there were no successful strategies reported in the literature specifically for children with CP [5]. This begs the question: what are clinicians to do when children with CP are identified as having tactile impairments? This current perspectives paper presents a strategic framework designed to guide clinicians in more tailored selection of tactile interventions, and to guide researchers in refining the nature and dose of treatments in order to more efficiently and effectively optimise tactile function for children with CP.

What we know

Currently, very little is known about interventions for tactile impairments in children with CP. The 2014 systematic review by Auld et al. identified five studies that measured tactile impairment before and after intervention in children with CP [5]. All five studies included interventions designed to treat motor impairments – that is, none were specifically aimed at reducing tactile deficits. However, each study measured tactile performance as a potential by-product of motor training. These early paediatric studies did not produce improvements in tactile performance, however, the review did identify other successful tactile interventions in

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populations of adults with brain injury (stroke) with potential adaptability for children with brain injury. These interventions were mirror therapy [6] and stimulus specific training [7].

Since that review, five new studies have been published in 2016 for children with CP with variable results. Three studies investigated stereognosis as an outcome. First, Petersen et al. tested the relative effect of three interventions in 63 children with CP (aged 4.4–16 years): (i) surgery with rehabilitation, (ii) upper limb botulinum toxin, and (iii) rehabilitation alone [8]. Stereognosis of 12 common objects was not improved following any of these interventions, which concurs with the results from the systematic review. A second study by McLean et al. measured stereognosis and tactile discrimination in 15 children with CP (mean age 10 years 1 month) [9]. This feasibility study, which was reported as a published conference abstract, involved a single-blind randomized trial of stimulus specific training (i.e., combined tactile/texture discrimination, stereognosis, and proprioception training) for three times per week for six weeks, compared to control (standard care). Although there was a trend towards improvement in stereognosis (Functional Tactile Object Recognition Test) in the treatment group ($n = 9$) compared to the control group ($n = 6$), this was not statistically significant. Further research is required to establish the effectiveness of stimulus specific training for children with CP. A third study by Hobbs et al., measured stereognosis in conjunction with tactile registration and texture perception in 18 children with CP (mean age 10 years 8 months) [10]. This pilot randomised controlled trial, also reported as a published conference abstract, investigated the use of hand vibration delivered via a bimanual track-ball device linked to a gaming system to engage the child in a required dose [10]. The experimental group ($n = 10$) were encouraged to use the gaming system at home for six weeks, while the control group ($n = 8$) used the same gaming system except with the haptic vibration disabled. Results showed no statistical difference in tactile registration, texture perception or stereognosis between the two groups [10]. However, participant satisfaction with the gaming system was reported to be high.

One further study by Kuo et al. [11] investigated texture perception, as well as tactile registration and perception. The study was a randomised controlled trial involving 20 children with CP (age 6–15.5 years) receiving: (i) 82 hours of intensive bimanual intervention plus eight hours of directed play with textured objects with vision occluded, compared to (ii) 82 hours of intensive bimanual training (BMT) plus eight hours of free play with textured objects with vision available [11]. Both groups of children improved their tactile texture perception on a grating orientation task, but there was no significant change in tactile registration or other perception domains [11].

The final study by Auld et al., assessed tactile registration and unilateral and bilateral spatial perception. It piloted the use of mirror-based training for children with CP, on the back of positive results for adults with stroke that were identified by the tactile systematic review [12]. Auld's paediatric study used a robust replicated randomized controlled case series design to show that a single session (54 min) of mirror-based training was able to improve spatial tactile perception for children with unilateral CP aged 7–17 years. Given these promising results, further research is recommended to explore the mirror therapy strategy for larger cohorts of children with CP.

These studies have shown that it is possible to achieve improvements in tactile perception, across spatial and textural domains. At this stage, it is useful to provide a clinical reasoning framework for tactile treatment selection for clinicians, with this framework also providing a stimulus for strategic thinking about future research to improve evidence for tactile interventions, so that children who currently have tactile impairments can receive the treatment they need. To assist the reader, we propose this framework using an analogy – *The Apartment Block Theory*.

The apartment block theory

Have you ever been stuck outside an apartment block? The buzzer at the front door connecting to your friend's apartment is broken, and no matter how much you press it, you cannot get your message through. Perhaps, tactile function is like this? Tactile stimuli have been applied to the skin and resulting tactile messages are trying to travel to the brain, but somewhere in this process the pathway is impeded. A range of barriers are possible along this pathway, and to overcome these we can think about the skin-to-brain pathway like an apartment block. To reach the top floor of the apartment block, you essentially have three options: *(1) Pressing the buzzer repeatedly*, *(2) Sneaking in the door*, or (3) *Connecting another way*. Maybe these same three options could be used to access the tactile building (Figure 1).

Pressing the buzzer repeatedly

First, to get into the "tactile apartment" we can keep *pressing the buzzer* and hope that the message eventually gets through – that is, provide the child with repeated tactile stimuli with the hope that they eventually feel something. Despite historically being a strategy that has been employed by some therapists, there is a lack of evidence in the current literature that this works when delivered independent of other strategies. Although there have been no reports of the effect of passive tactile stimulation on tactile registration or perception in children with CP, it could be anticipated that this strategy would be susceptible to sensory accommodation or sensory overload and unlikely to overcome the issues associated with tactile registration. If we consider our approaches to other sensory systems, we would not expect someone who had a visual registration deficit to improve by being presented with lots of stimuli to look at, and perhaps the same should apply to the tactile sense.

One area of treatment that has been explored in the adult stroke literature is use of electrical tactile stimulation [13–17]. Although obviously providing significantly greater neural input by stimulating peripheral nerves rather than just providing tactile stimuli to the skin surface, these studies demonstrated no improvement in touch performance [5]. That is, merely providing passive peripheral stimulation in what is known to be a central nervous system condition was not enough to impact tactile performance. This may indicate that "*pressing the buzzer*" will be similarly ineffectual in children with CP, however, further research is required to confirm this suggestion. One recent study utilised an innovative approach for "*pressing the buzzer*" by providing repeated stimulation to a child's hands when they were in bimanual contact with an engaging video game [10]. Although this pilot trial reported no significant changes in tactile function in children with CP, the use of engaging formats for providing repeated tactile stimulation warrants further investigation.

Sneaking in the door

Tactile treatment is often found "*sneaking in the door*" with upper limb movement training. Some treatment approaches, such as constraint induced movement training (CIMT) or BMT involve providing movement-focussed therapy with a secondary anticipation that tactile function may also improve by opportunistically

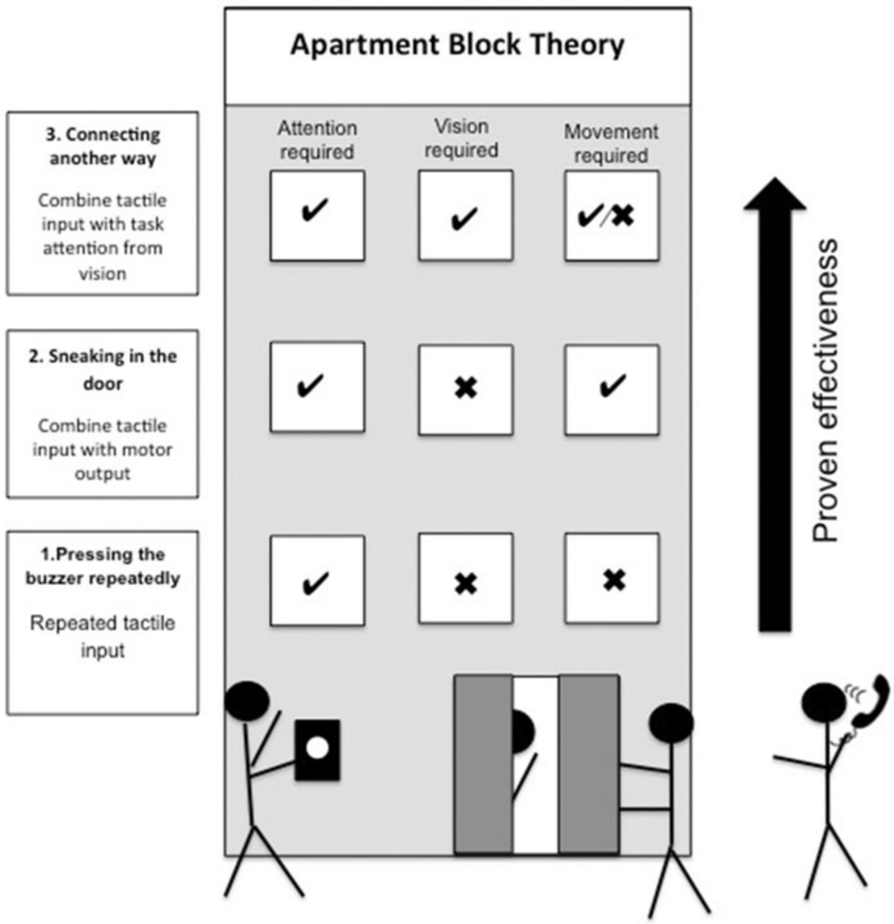

Figure 1. Apartment block theory for treating the tactile system in children with cerebral palsy.

sneaking in the door as motor output occurs. Four studies investigating CIMT or BMT in children with CP have investigated tactile performance before and after intervention. Three studies showed no statistical improvement in tactile performance. In the first study, children showed no improvement in two point discrimination following a two-week intensive CIMT block (70–84 hours) plus six months of home program [18]. In a second study, improvements in two point discrimination in the impaired hand were shown after six hours of CIMT, however, this was not examined statistically [19]. In a more recent study, 60 children were randomised to either 60 hours of CIMT or BMT, with both groups demonstrating no improvement in two-point discrimination or stereognosis [20]. Thus it seems that even at high doses, tactile stimuli unintentionally *sneaking in the door* with CIMT or BMT leads to very limited or no improvement in tactile performance.

The lack of significant improvement in tactile performance with movement training in these three studies is perhaps not that surprising, when considering the perfect positioning, timing and attentional capacity that is needed for tactile function to *sneak in the door* with movement. First, the hand needs to be able to be placed in contact with a tactile stimulus in an optimal manner. Second, the door must be open long enough for the afferent tactile input to sneak in the door opportunistically as the efferent motor output is occurring. Third, the door needs to be opened wide enough for higher processing centres to attend to and identify the tactile stimulus simultaneous to completing the motor task. If, during the movement task, the child is primed to share their attention between the quality of their motor output and the congruent features of the tactile input by careful instruction, then the child may achieve both the motor task as well as perception of the tactile features. However, if the child's attention is focussed entirely on the characteristics of the motor performance, it is unlikely that the tactile input, though it is present at the same time, will fit through the attentional door. There needs to be absolute congruence between tactile input and motor output and optimal allocation of location, time and attention for *sneaking in the door* to facilitate tactile perception successfully.

One study has shown that combined tactile-motor therapy (i.e., bimanual therapy with intentional tactile components), which is consistent with the *sneaking in the door* approach, can improve both tactile and motor performance. In the Kuo et al. [11] study introduced above, all participants received 82 hours of Hand-Bimanual Training (HABIT) [21] training, along with an additional eight hours of textured-object training (without vision) (HABIT-+T Group) or eight hours of textured-object play (with vision) (HABIT-Control Group) [11]. Results show that both groups improved their grating orientation skills (texture perception), irrespective of the mode of textured-object interaction, however, there was no statistical improvement in other tactile registration or perception domains. These results support the authors conclusion that "tactile impairments can be improved when the tactile input is structured in the environment" (p. 138) [11], specifically that high-dose HABIT plus targeted textured-object interaction may have a specific positive impact on texture perception. However, similar to other previous studies that utilise a mixed intervention approach [9], further research is needed to determine whether the impact on texture perception is most attributable to the 82 hours of HABIT or the eight hours of textured-object interaction, or both combined.

This study shows that use of structured motor tasks that are weighted heavily in the tactile domain may be able to ensure sufficient congruence between motor output, tactile input and tactile attention to improve texture perception.

The different use of vision in the HABIT + T versus HABIT + Control groups in the eight hours of textured-object training deserves consideration. In the HABIT + T group, tactile training was provided without vision – essentially a form of Visually Constrained Touch – which works theoretically by encouraging individuals to channel their attention towards the tactile properties of objects by removing the ability to rely on visual cues. In the HABIT + Control group, children were provided with the same tactile stimuli, however they were able to obtain additional information via visual cues during tactile exploration. Despite the differences in the availability of vision, both groups demonstrated similar improvement in texture perception. This does raise the question as to exactly what role vision plays in tactile treatment.

Connecting another way

Alternatively, we can connect a different way – to get into the apartment block you may phone your friend and inform him that you are there – so he can look out his window and say, "I can see you there – I'll let you in." In the same way, to enhance tactile registration and/or perception we may ask a child to visually attend to their hands during tactile stimulation, in order that they can use visual information to inform tactile brain regions that a tactile stimulus is present. This is referred to as visually enhanced touch or the VET effect, and is proposed to occur when vision modulates activity in the primary somatosensory cortex via bimodal neurons that respond to both visual and tactile information in the same corresponding area of skin [22]. Demonstrated primarily in adults to date, the VET effect does not require the vision to be informative about the nature of the touch (e.g., using vision to identify whether one is touched with one or two stimuli in a two-point discrimination trial) [23], but rather capitalises on the modulation of early somatosensory processing in the primary somatosensory cortex by viewing one's own body [24], in-turn reducing the tactile field size of involved neurons and enhancing tactile acuity [23].

The VET effect is strongest when the viewed body part is (i) identified as belonging to the individual [25], (ii) when a sufficient portion of the body can be seen [23], (iii) when vision of the body part is magnified [26] and (iv) when the task being attended to is at the limits of performance for the individual [27]. In one study, non-informative vision of the hand has been shown to improve tactile acuity in both the hand and the face of the individual, but not the foot [28], possibly due to the significant functional relationship of the hand and face [22]. Similarly, the VET effect is more commonly seen in those body parts that are often viewed by the individual (e.g., the face in preference to the neck [29]). The effect also seems to be greatest in those individuals with lower baseline performance [30]. This research highlights several valuable intervention conditions that should be utilised when setting up a task designed to capitalise on the VET effect.

The VET effect can also be capitalised on with illusory vision or mirror-based training. In one study in adult patients with complex regional pain syndrome, the use of mirror-based training demonstrated an improvement in two point discrimination ability that lasted at least 48 hours [31]. Similarly, mirror-based training in adults with peripheral nerve damage has been shown to significantly improve tactile function in areas of chronic numbness [32]. Further, an RCT in 36 adult stroke patients demonstrated that mirror-based training involving watching the unimpaired limb (in the apparent position of the impaired limb) perform a series of hand movements on verbal command could improve tactile registration as measured by the Fugl Meyer assessment [6].

Mirror-based training has recently been demonstrated to improve spatial tactile perception in children with CP [12]. The treatment design involved two mirror-based tasks: (i) a visually-non-informative tactile perception task, and (ii) a visually-informative hand position copy task. The protocol required minimal equipment or cost, and was well-tolerated by children. This "*connecting another way*" training was extremely efficient, leading to changes in spatial tactile perception after a single treatment session of only 54 minutes in duration, which is significantly less than the doses required for "*sneaking in the door*" approaches to improve tactile performance in children with CP. Children demonstrated changes in single point localisation and double simultaneous, two tactile test items appropriate for children with CP [1,33] and shown to be most significantly related to both unimanual and bimanual upper limb motor performance, respectively [3]. Given these promising results and the great potential this type of training has to utilise technology in a game-oriented format with easy translatability to the paediatric population, further research is warranted.

One critical aspect of the "*connecting another way*" method of accessing the tactile building is how it differs to what therapists may have automatically and historically done in tactile treatment. Previously there may have been a tendency to tell children to close their eyes, or blind fold them so that they could concentrate on what they were feeling – essentially Visually Constrained Touch, where success of the VET effect relies on the therapist ensuring that children sustain visual attention when receiving the stimulus. Planning therapy sessions that utilise the VET effect for tactile tasks at the limits of performance is recommended. In addition, future studies with children with CP are required to elucidate subgroups of children for whom the VET effect is potentially beneficial. For now, though, the most critical component to realise is the need to continuously and consciously capitalise on vision in tactile training endeavours.

Choosing the right approach

When choosing the right approach it is necessary to consider the combination of both best practice evidence and what may be feasible for a particular child given their specific combination of neurological, sensory, motor, and cognitive impairments. This performance profile will influence clinical decision making regarding the best approach, or which further research on tactile interventions is required. Based on the current literature, and using the "apartment block" analogy, several principles may be considered when designing tactile treatments:

1. *In the best-case scenario, when working with children who have sufficient attention and vision, it is possible to capitalise on the VET effect (i.e., "connecting another way")*. This approach, which integrates visual attention with concurrent tactile stimuli, has the strongest positive evidence for improving tactile function in adult and paediatric populations. This strategy can be delivered via a direct visual observation method, or a mirror-based training approach, and will be most effective for children who have sufficient: (i) vision to see their hand, (ii) ability to maintain attention to task, and (iii) movement to open the hand.
2. *In children who have visual difficulties, but still have sufficient attention and movement, then the best option is to combine tactile input with motor intervention (i.e., "sneaking in the door")*. This approach, which involves intensive bimanual motor training combined with texture and stereognosis

training, may lead to improvements in tactile performance in children with CP. Texture and stereognosis training may be provided through exposure to various textures, surfaces, objects, sizes, and shapes. Bimanual motor training is recommended to ensure "best value" from treatment sessions. However, further research is required to investigate unimanual training with tactile-loaded activities, as well as the nature and dose of these activities.

3. *In children who have complex co-morbidities, including both visual and movement impairments, it may be necessary to consider providing passive tactile stimulation (i.e., "pressing the buzzer")*. In this strategy, a therapist or device is used to passively provide tactile stimuli, since children lack sufficient visual and motor function to participate in active intervention. Similar to sneaking-in-the-door, texture and stereognosis training may be provided through exposure to various textures, surfaces, objects, sizes, and shapes. However this is not accompanied by motor involvement in the task. Further research is needed to investigate the effectiveness of more deliberate spatial and temporal stimulation, perhaps delivered via vibration, brushing, monofilaments, or other means.

Irrespective of the strategy chosen, it is important to plan appropriate monitoring of sensory engagement, tolerance and functional outcomes. When delivering any tactile intervention, it is important to monitor for sensory overload since repeated stimulation may inadvertently create or exacerbate tactile hypersensitivity problems. Secondly, it is critical to always consider safety. Since a significant proportion of children with CP have an impairment in tactile registration [2], it is important to educate families and teachers about the potential safety risks of certain tactile stimuli (e.g., rough sandpaper) and to consider this when providing tactile stimuli as part of therapeutic interventions. Finally, it is important to reflect on the tactile assessment framework to identify the measure of tactile function that is most likely to be responsive to the chosen intervention. Likewise, if there are parallel gains to be made in motor function or visual perception, then these outcomes should also be measured before and after intervention. Each of these areas would benefit from further research in order to inform clinical decision making.

Conclusions

It has been known for some time that tactile impairment is prevalent in children with CP, but evidence underpinning treatment recommendations is still in development. This paper proposes a framework for clinical reasoning and future research in tactile intervention based on the evidence to date. Considering this literature, clinicians are encouraged to explore the benefits of VET for children with sufficient visual, motor, and cognitive skills; tactile-enhanced motor training for children with visual impairment; or repeated stimulation in children with more complex comorbidities involving both visual and motor impairment. Further research is needed in larger and more diverse cohorts to determine the nature, timing and dosage of each of these treatment strategies for optimal effect.

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