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RESEARCH PAPER

Tactile function in children with unilateral cerebral palsy compared to typically developing children

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Purpose: **Tactile deficits have been understudied in children with unilateral cerebral palsy (UCP) using a limited range of tactile assessments. This study aims to characterize performance across a comprehensive battery of tactile registration and perception assessments in children with UCP and typically developing children (TDC).** ***Methods:*** **Fifty-two children with UCP (Gross Motor Function Classification System I = 34, II = 18; Manual Ability Classification System I = 36, II = 16) and 34 TDC were assessed using Semmes Weinstein Monofilaments (tactile registration), and single-point localization, double simultaneous, static and moving two-point discrimination, stereognosis, and texture perception (tactile perception).** ***Results:*** **Children with UCP performed consistently worse with their impaired hand than their unimpaired hand ($Z = 2.77–5.61$; $p < 0.005$). Both hands of children with UCP performed worse than either hand of TDC ($Z = –2.08$ to 5.23; $p = 0.037–< 0.001$). Forty percent of children with UCP had tactile registration and perception deficits, 37% had perception deficits only and 23% had no tactile deficit. The larger the tactile registration deficit, the poorer the performance on all tactile perceptual tests ($r = 0.568–0.670$; $p < 0.001$).** ***Conclusions:*** **Most children with UCP demonstrate poor tactile perception and over one-third also demonstrate poor tactile registration. We contend that tactile dysfunction may contribute to functional impairment and is a possible target for intervention.**

Keywords: Cerebral palsy, children, hemiplegia, perception, sensation, tactile

Implications for Rehabilitation

- Cerebral palsy (CP) is the most prevalent physical disability in childhood, with an incidence of approximately 2 cases per 1000 live births; about 35% of children with CP have unilateral cerebral palsy (UCP).
- Assessment and treatment has been focused on the motor impairment; however, it is known that children with UCP are also likely to have sensory impairment.
- Understanding the nature and severity of sensory, specifically tactile, impairment in children with UCP will assist therapists to direct treatment accordingly and possibly impact the motor impairment.

Introduction

The prevalence, severity and nature of tactile impairments in children with unilateral cerebral palsy (UCP) have been widely studied [1–8], however, findings are inconsistent. That tactile dysfunction is present is not in doubt, but the full profile and severity of dysfunction in this population, and its subgroups, has not yet been resolved. A full battery of tactile registration and perception tests has not been carried out. Questions that remain are: What is the full profile and severity of tactile deficits experienced by children with UCP? In the UCP population, is there a difference in the tactile performance of subgroups of children with left-vs. right-brain injury as is commonly seen in adult stroke populations? Are tactile deficits present in the relatively "unimpaired" limb and if so, to what extent? To clearly determine the magnitude and diversity of tactile deficits, we undertook a comprehensive test battery of tactile registration and perception tests in a single cohort of children with UCP, and compared their results to a cohort of typically developing children (TDC).

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To profile tactile deficits in the UCP population, we identified potential tactile-test items via a systematic review of tactile assessments for children with cerebral palsy (CP). This review utilized a previously developed framework [9] that consists of two phases: registration and perception. Registration is defined as the initial detection of sensory information. Perception is the interpretation of that tactile information. Perception includes three domains: spatial ("where is the stimulus?"), temporal ("when did the stimulus occur?") and modality-specific characteristics ("what is the stimulus?"). Modality-specific characteristics in the tactile domain relate to texture. The perception of texture is composed of processed information originating from perceived spatial and temporal parameters [10]. Items identified in the review were selected for the framework if they had adequately reported validity, reliability and clinical utility for the CP population. We then confirmed the reproducibility of these measures in children with UCP [11].

In literature that compares healthy adults to adults poststroke, differences in tactile performance have been observed, dependent on the side of brain lesion. In particular, lateralization of spatial attention is commonly reported in healthy adults, with the right hemisphere being more active than the left during spatial tasks, regardless of the side of stimulation [12,13]. This lateralization becomes problematic following brain injury, where adults with a right-hemisphere lesion are often unable to attend to stimuli on the left side of space when provided with competing stimuli on the right [14,15]. No research has been undertaken to investigate the degree of lateralization of spatial attention during typical development in children. Little is known about the possible susceptibility to spatial-perception difficulties following right-sided brain injury, and/or the potential for reorganization following early brain lesion, to compensate for spatial lateralization. One recent study showed that moving two-point discrimination (M2PD) was better in children with right hemiplegia than it was in those with left hemiplegia ($Z = 2.2$, $p = 0.03$). However, as stereognosis was not affected by lesion side, the extent of the effect of lesion side remains unclear [16].

We hypothesized that: (1) impairments in tactile registration and perception are associated with the presence of UCP; (2) in TDC, the performance of tactile registration and perception is influenced by hand dominance; and (3) in children with UCP (a) tactile registration and perception performance are associated with the presence of unilateral hand impairedness, (b) there is an association between the severity of registration deficits and the severity of perception deficits, and (c) performance in spatial-tactile perception is influenced by the side of the brain lesion. In addition, it is anticipated that in children with UCP, three profiles of performance would be observed: (1) children with deficits in both tactile registration and perception; (2) children with no registration deficits, but primary deficits in perception; and (3) children who have no registration or perceptual deficits.

Methods

Study design and participants

We conducted a cross-sectional study with TDC and children with UCP aged 8–18 years. Ethical approval was granted by The University of Queensland, the Cerebral Palsy League and the Royal Children's Hospital, Brisbane, Australia.

Participants

Children who were registered on the two main databases of children with CP in Queensland: the Cerebral Palsy League database and the Royal Children's Hospital, Department of Paediatric Rehabilitation database, were eligible. All children on these databases have a confirmed diagnosis of UCP. The exclusion criteria were: an inability to understand and/or follow test instructions due to intellectual or behavioral difficulties; receipt of upper-limb intramuscular Botulinum Toxin A injections within 3 months prior to recruitment (data showed no children in this study had Botox within 12 months); previous upper-limb orthopedic surgery; and/or uncorrected visual impairment. Recruitment information was mailed to all families, who were then followed up via a phonecall.

TDC volunteered in response to advertisement fliers and e-mail circulars. Children were excluded if they had any known impairment in intellect, upper-limb performance, behavior, tactile performance or known uncorrected visual impairment.

Assessment procedure

To evaluate tactile function, a physiotherapist who was experienced in the evaluation of children with CP performed the assessments at the child's home, the Royal Children's Hospital, Brisbane or a Cerebral Palsy League venue. Prior to assessment, the dominant hand for each child was established using the Edinburgh Handedness Inventory [17] and upper-limb function was classified using the Manual Abilities Classification System [18]. Children were screened for intellectual difficulties that may affect their understanding of the tasks using the Kaufman Brief Intelligence Test [19]. The full battery of tactile assessments was completed in less than 30 minutes. Tactile function was assessed by one test of tactile registration, five tests of spatial-tactile perception and one test of texture perception. The available clinimetric properties for each of these tactile assessments have been established in a previous systematic review [9]. The exact methodology for each tactile test has been reported in a previous study [11]. All the tests of tactile registration and perception have satisfactory reproducibility (agreement and test-retest reliability) [11].

Tactile registration was performed using the full 20-item Semmes Weinstein Monofilament (SWM) kit and original test methodology [20]. The outcome measure was the lowest monofilament (value) at which the child was able to correctly identify at least one touch (out of a possible three) on all four tested fingers. Single-point localization was assessed using the largest (suprathreshold) SWM and was scored as the number correct out of 12 and double simultaneous as the number correct out of 24 [21]. Both M2PD and static 2PD (S2PD) were assessed using the Disk-Criminator® [22,23]. Texture perception was tested using the AsTex®, a perspex board that displays tactile gratings of reducing tactile discrimination index [24]. The averaged scores from three fingers were converted to the tactile discrimination index for each finger using the chart available with the test kit. Motor-enhanced tactile perception was assessed using stereognosis with nine common objects, with the outcome being the number of correct responses out of a possible maximum of nine [25].

Statistical analysis

The distribution of the data was examined, and due to the nonsymmetrical nature of the data, summary statistics are presented using median and interquartile range for each tactile test in the UCP and TDC groups. The percentage of children with UCP who had deficits on each tactile test was calculated, using the score at which the lowest 5% of the TDC group performed as the cutoff. We investigated the data and since the assumptions of linear regression models (normality of residuals and constant variance of residuals) were not met we analyzed our data using nonparametric tests. To test the first hypothesis, that presence of CP is associated with impairment in tactile performance, the differences between the tactile performance of both upper limbs of children with UCP and TDC were tested using the Wilcoxon rank sum test. To examine the differences between the dominant and nondominant hands within groups, the Wilcoxon signed rank test was used (Hypotheses 2 and 3a). To test whether there was an association between the severity of registration and perception deficits (Hypothesis 3b), Spearman's correlation coefficient was calculated for the outcomes of each tactile test. The differences in tactile performance between children with left- and right-sided brain injury (Hypothesis 3c) were tested using Wilcoxon rank sum tests. The relationship between registration deficits and perception deficits was evaluated by determining perceptual performance at each level of registration performance, and by calculating the odds ratio of having a perceptual deficit when there is also a registration deficit, compared with when there is no registration deficit. Relationships between age and tactile results were investigated using Spearman's correlation coefficients. A p value of 0.05 was considered statistically significant. All statistical tests are based on two-sided comparisons. There was no adjustment for multiple comparisons.

Results

Participants

Database audits identified 253 potential participants with UCP. Fifty-nine children provided informed consent to participate in the study and 52 of those met the criteria (median age = 12 years; range 8–17 years; 29 male; 23 left UCP; Gross Motor Function Classification System I = 34; II = 18; Manual Ability Classification System I = 36; II = 16). The reference population comprised 34 TDC (median age = 9 years; range 5–17 years; 20 male; nine left-handed).

Comparison of children with UCP and TDC

The impaired hand of children with UCP performed worse than the nondominant hand of TDC in all tactile tests except M2PD (as seen in Table I and Table II). Similarly, the unimpaired hand of children with UCP showed poorer performance than the dominant limb of TDC on all tactile tests. Further, for each tactile test except M2PD, the unimpaired (i.e. best performing hand) of the UCP group showed significantly poorer performance than either hand of the TDC (Table II). There were no differences on any tactile tests between the dominant and nondominant hands of TDC.

Children with UCP

Performance was worse using the impaired hand than using the unimpaired hand on all tactile tests in children with UCP (Table II). Forty percent of children with UCP (n = 21) had both registration and perception deficits, 37% (n = 19) had perceptual deficits alone and 23% (n = 12) had normal tactile function. The percentages of those children with UCP who had an impairment in each perceptual test were: single-point localization: 40% (n = 21); double simultaneous: 58% (n = 30); S2PD: 31% (n = 16); M2PD: 27% (n = 14); stereognosis: 63% (n = 33); AsTex® (index finger): 17% (n = 9). Registration performance (SWM) of the impaired hand and perception as measured by all other tactile tests were moderately correlated ($r = 0.56–0.67$; $p < 0.001$) (Table III). Tests of spatial

Table I. Tactile performance (median and IQRs) on tactile assessment items in children with UCP and TDC.

		UCP				TDC			
		Impaired		Unimpaired		Nondominant		Dominant	
Test	Range (lowest to highest) (min unit)	Median	IQR	Median	IQR	Median	IQR	Median	IQR
SWM	1–20 (1)	4	2–6	3	2-5	2	1-2	2	1–3
SPL	1–12 (1) correct	11	6–12	12	11–12	12	12–12	12	12–12
S2PD	15–2 (1) mm	3	3–7	2	2–3	2	2-2	2	2–2
M2PD	15–2 (1) mm	3	2–6	2	2–2	2	2-2	2	2–2
DS	1–24 (1) correct	17	12–22			23.5	22–24		
Stereognosis	1–9 (1) correct	7	4–9	9	8–9	9	9–9	9	9–9
AsTex®-index finger	2.5 (0.03)	0.60	0.40–0.83	0.44	0.24–0.68	0.27	0.21–0.64	0.27	0.21–0.47
AsTex®-thumb	2.5 (0.03)	0.64	0.37–0.90	0.44	0.27–0.67	0.31	0.21–0.44	0.27	0.21–0.47
AsTex®-5th	2.5 (0.03)	0.60	0.36–0.77	0.44	0.27–0.67	0.31	0.21–0.50	0.31	0.21–0.47

IQR, interquartile range; UCP, unilateral cerebral palsy; TDC, typically developing children; SWM, Semmes Weinstein Monofilaments; SPL, single-point localization; DS, double simultaneous; S2PD, static two-point discrimination; M2PD, moving two-point discrimination.

Table II. Comparisons between hands and between groups in children with UCP and TDC.

	Wilcoxon signed rank for difference Z-scores between hands				Wilcoxon rank sum between groups					
	TDC group		UCP group		Impaired vs. nondominant		Unimpaired vs. dominant		Nondominant vs. unimpaired	
Test	Z-score	p value	Z-score	p value	Z-score	p Value	Z-score	p value	Z-score	p value
SWM	– 1.13	0.258	2.78	0.005	– 5.23	< 0.001	– 3.37	< 0.001	– 4.38	< 0.001
SPL	– 0.68	0.495	– 4.03	< 0.001	4.24	< 0.001	3.02	0.002	2.57	0.010
DS					5.52	< 0.001				
S2PD	– 0.58	0.564	5.62	< 0.001	– 2.52	0.012	– 6.36	< 0.001	5.52	< 0.001
M2PD	– 1.0	0.317	5.17	< 0.001	– 1.34	0.182	– 5.34	< 0.001	– 1.72	0.086
Stereognosis	0	NA	– 5.19	< 0.001	2.82	0.005	5.45	< 0.001	2.82	0.005
AsTex®-In	0.78	0.434	3.77	< 0.001	– 2.34	0.019	– 3.19	< 0.001	– 1.66	0.097
AsTex®-Th	– 0.31	0.753	4.17	< 0.001	– 2.08	0.038	– 3.61	< 0.001	– 2.09	0.036
AsTex®-5th	– 0.47	0.636	3.73	< 0.001	– 2.20	0.028	– 3.63	< 0.001	– 2.14	0.032

UCP, unilateral cerebral palsy; TDC, typically developing children; SWM, Semmes Weinstein Monofilaments; SPL, single-point localization; DS, double simultaneous; S2PD, static two-point discrimination; M2PD, moving two-point discrimination; AsTex®-In, AsTex®-index finger; AsTex®-Th, AsTex®-thumb; AsTex®-5th, AsTex®-5th digit.

Table III. Relationships between different tactile assessments in the impaired hand of children with UCP (n = 52) using Spearman's correlations.

	Registration (SWM)	Single-point localization	Double simultaneous	Static 2PD	Moving 2PD	Stereognosis
Single-point localization	R = –0.67 p < 0.001					
Double simultaneous	R = –0.57 p < 0.001	R = 0.76 p < 0.001				
Static 2PD	R = 0.60 p < 0.001	R = –0.64 p < 0.001	R = –0.50 p = 0.001			
Moving 2PD	R = 0.57 p < 0.001	R = –0.61 p < 0.001	R = –0.52 p < 0.001	R = 0.76 p < 0.001		
Stereognosis	R = –0.66 p < 0.001	R = 0.74 p < 0.001	R = 0.75 p < 0.001	R = –0.74 p < 0.001	R = –0.61 p < 0.001	
AsTex®	R = 0.60 p < 0.001	R = –0.51 p < 0.001	R = –0.31 p = 0.059	R = 0.43 p = 0.008	R = 0.23 p = 0.174	R = –0.53 p < 0.001

UCP, unilateral cerebral palsy; SWM, Semmes Weinstein Monofilaments; 2PD, two-point discrimination; R= correlation.

perception for the impaired hand were related to each other (r = 0.55–0.76; p < 0.001). Texture perception (AsTex®) of the impaired index finger showed moderate correlations with registration, single-point localization, S2PD and stereognosis (Table III). Notably, the largest correlation between tests was only 0.76 (between S2PD and M2PD), which suggests that no two tests are sufficiently similar for one to be disregarded, on statistical grounds, from the assessment framework.

Having a registration deficit increases the likelihood of having a perceptual deficit, as demonstrated by the reported odds ratios (Table IV). Odds ratios were lower for the more difficult tests (e.g. double simultaneous), as there were many children who exhibited deficits on these items even with normal registration performance.

In the children with UCP, there were no significant performance differences between children with left vs. right UCP on any individual tactile test (Table V). However, the percentages of children with registration and perception deficits differed according to side of UCP. Although the number of children with normal function was similar (left 22%, n = 5/23; right 24%, n = 7/29), the combined registration and perception deficit was more pronounced in children with left UCP (52%, n = 12/23) than in those with right UCP, (31%, n = 9/29), whereas the less-severe perception-only deficit was more common in children with right UCP (45%, n = 13 vs. left 26%, n = 6). Double simultaneous performance was slightly better with age (r = 0.3552; p = 0.0265) for children with UCP, but there were no other age-dependent effects.

Discussion

This study is the first to clearly document that over 75% of children with UCP have tactile deficits, and over 40% have deficits in both tactile registration and perception. The results confirm that tactile performance was poorer on the impaired hand of children with UCP than it was on their unimpaired hand, and worse than both hands of TDC. However, tactile deficits do extend to the unimpaired hand as well – performance was worse on the unimpaired hand of children with UCP than it was on either hands of TDC. Specifically, 53.8% of the 52 participants with UCP demonstrated some form of tactile impairment in the unimpaired limb, using the cutoffs derived from the sample of TDC. Bilateral tactile impairment would have significant implications for bimanual function, which is essential for most activities of daily living.

Tactile registration

Tactile registration deficits are clearly associated with an increased likelihood of tactile perception deficits. This indicates that, fitting with the defined framework, a deficit in tactile

Table IV. Odds ratios for the presence of impaired tactile perception based on presence of impaired registration (with SWM category 1 = sensation within normal limits, SWM category 2–4 = sensation outside normal limits) and performance on perceptual tests at each level of tactile-registration impairment.

Total n = 52; sensation within normal limits = 31	Cutoff for impaired perception	Odds ratio	95% CI	Normal sensation 1 (n = 31)		Diminished light touch 2 (n = 9)		Diminished protective sensation (n = 9)		Loss of protective sensation 4 (n = 3)	
				%	n	%	n	%	n	%	n
Spatial											
SPL	< 10	20.8	(4.8–89.1)	16.1	5	55.5	5	88.8	8	100	3
S2PD	< 4	8.6	(2.2–33.8)	12.9	4	11.1	1	88.8	8	100	3
M2PD	< 4	18.7	(3.4–99.6)	9.6	3	11.1	1	77.7	7	100	3
DS	< 20	7.8	(1.8–32.4)	41.9	13	77.7	7	88.8	8	100	3
Stereognosis	< 9	11.5	(2.2–58.2)	45.2	14	77.7	7	100	9	100	3
Textural											
AsTex® index	< 0.9	3.6	(0.7–16.5)	9.6	3	11.1	1	44.4	4	66.6	2

SWM, Semmes Weinstein Monofilaments; SPL, single-point localization; DS, double simultaneous; S2PD, static two-point discrimination; M2PD, moving two-point discrimination; CI, confidence interval.

Table V. Difference between the impaired hand of children with left vs. right UCP on tactile assessment.

	Median (interquartile range)		Wilcoxon Rank Sum Test between groups	
Tests	Left UCP (n = 23)	Right UCP (n = 29)	Z-score	p value
Semmes Weinstein Monofilaments	5 (2–8)	4 (2–5)	1.05	0.29
Single-point localization	11 (6–12)	11 (7.5–12)	–0.89	0.38
Double simultaneous	16 (9–21)	18 (12–22)	–1.19	0.24
Static 2PD	3 (3–8)	3 (3–6.5)	–0.51	0.61
Moving 2PD	3 (2–7)	4 (2.5–4)	–0.07	0.95
Stereognosis	7 (2–9)	7 (5–9)	0.15	0.88
AsTex®-index finger	0.64 (0.27–0.96)	0.6 (0.44–0.78)	–0.06	0.96
AsTex®-thumb	0.64 (0.27–0.8)	0.64 (0.47–0.91)	–0.71	0.48
AsTex®-5th digit	0.64 (0.37–0.86)	0.6 (0.4–0.74)	0.44	0.66

UCP, unilateral cerebral palsy; 2PD, two-point discrimination.

registration is associated with an inevitable deficit in tactile perception (Table IV). If a child has reduced tactile registration, he/she has reduced ability to detect tactile stimuli. This, in turn, means that the child is unable to perceive the object characteristics, which reduces his/her ability to understand and utilize sensory information and thereby disrupts his/her interactions with objects in their environment. It would seem important then to assess tactile function with regard to both tactile registration and perception. The full nature or severity of tactile impairment in UCP cannot be understood without an assessment of tactile registration and these results indicate the extent of tactile registration deficits in this population and the impact they have on tactile perception.

Tactile perception

For children with UCP, performance on all spatial tests was related, which demonstrates the similarity in item construct. As the highest correlation between tactile variables was 0.76, this indicates that there is no redundancy in the test battery – no two variables are sufficiently related to disregard one of them. Further, if time permits, each item should be included because it examines a different aspect of tactile function, ranging from "easier" unilateral, unidermatomal spatial-tactile perception (single-point localization), through to the most challenging, bilateral spatial-tactile perception (double simultaneous). However, if time only permitted the inclusion of a small number of tests, it would be best to retain at least one test of registration (SWM), one test of unilateral spatial perception (single-point localization in preference to the less discriminative unilateral 2PD items) and one test of bilateral spatial perception (double simultaneous).

Performance on the AsTex®, the only test to investigate the modality-specific characteristic of texture, was moderately correlated to results on registration (SWM), single-point localization, S2PD, double simultaneous and stereognosis. These results imply that children who are unable to register or to be aware of tactile information also perform poorly on texture perception (presumably due to their inability to be aware of the stimulus, regardless of their ability to perceive the tactile gratings) and that this is significantly related to performance on spatial perception tests.

As the brain lesion in UCP can significantly vary between children, it is possible for a perceptual deficit to exist in the absence of a deficit in tactile registration. That is, a child may be able to detect sensory information, but unable to understand the location, shape or timing of the information. In this circumstance, assessing only tactile registration would provide the erroneous impression that there is no tactile impairment. It would also seem important to carry out tactile perception assessments in children who have impairments in registration to examine the extent of these relative to the registration impairment. If a child, however, has a tactile-registration deficit that categorizes him/her in the most severe SWM category (category 4: loss of protective sensation), the results from this study show that the child will definitely have deficits in all aspects of tactile perception (Table IV), which makes assessment of tactile perceptual function in that child redundant.

Our study indicated that there were no significant differences between children with left vs. right UCP on any specific tactile test. However, when observing the median and interquartile range values and the percentage of tactile registration and perception impairment, it can be seen that there is a trend for those with right-sided brain lesions (left UCP) to have more severe sensory deficits. For the median scores, the most notable difference is for double simultaneous – the bilateral spatial-tactile perception test. This supports results from the adult literature, where there is evidence that the right hemisphere is dominant for spatial attention to sensory stimuli [12,13,26,27]. The lack of significance of these data by comparison to the results we tend to see in the adult stroke population may indicate the reorganization potential of the developing brain, in that the typical hemispheric lateralization of function is altered in early brain injury. The difference between children and adults could also be related to a difference in lesion type/location. Most adult lesions are in the region of the middle cerebral artery, primarily impacting cortical function, which by its very nature is more lateralized to specific brain hemispheres. However, injuries sustained before birth, as is the case for many children with UCP, more commonly affect periventricular regions. Alternatively, it may be that this study was underpowered to identify a difference between those with left vs. right UCP. The slight trend identified in this study does, however, warrant further investigation of the potential differences between children with left vs. right UCP in a larger sample with neuroimaging data to elucidate that nature of the brain lesion and its reorganization.

One limitation with this study is the small range of severity of motor impairment, as indicated by the MACS, displayed in the sample. The children with UCP who participated in the study only represented MACS levels I and II. A sample including children with MACS III may have led to more comprehensive results and could perhaps show that tactile deficits are in fact underestimated in the current study.

Conclusion

The majority of children with UCP have tactile deficits. At least 40% of our sample had both registration and perception deficits and a further 37% had primary perception deficits. Tactile performance in the impaired limb can be expected to be poorer than both the unimpaired hand and either hand of peers with typical development. Tactile-registration deficits are associated with tactile perception deficits and children who demonstrate severe registration deficits (i.e. a loss of protective sensation) also demonstrate very poor tactile perception. There was a trend for children with left UCP to perform worse on spatial-tactile perception tests, than children with right UCP. These results highlight the extent of tactile registration and perception impairments in children with UCP, the potential differences between children with left vs. right UCP and the need for further research on how this information could lead to more targeted treatments for these children.

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