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Functional Plasticity in the Absence of Structural Change: Apraxia and Body Scheme Disorder 10 Years After Childhood Brain Injury

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Abstract

This work presents a case of a young woman with apraxia and a severe body scheme disorder, 10 years after a childhood frontal and occipitoparietal brain injury. Despite specific limitations, she is independent in performing all activities of daily living. A battery of tests was administered to evaluate praxis and body representations. Specifically, the Hand Laterality Test was used to compare RS's dynamic body representation to that of healthy controls ($N = 14$). Results demonstrated RS's severe praxis impairment, and the Hand Laterality Test revealed deficits in accuracy and latency of motor imagery, suggesting a significant impairment in dynamic body representation. However, semantic and structural body representations were intact. These results, coupled with frequent use of verbalizations as a strategy, suggest a possible ventral compensatory mechanism (top-down processing) for dorsal stream deficits, which may explain RS's remarkable recovery of activities of daily living. The link between praxis and dynamic body representation is discussed.

Keywords

rehabilitation, TBI, parietal cortex, Hand Laterality Test, motor imagery

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Praxis is the ability to plan and perform skilled movements using a movement scheme based on stored complex representations and previously learned movements. Apraxia refers to a failure to produce a correct movement in response to verbal command, imitate an action performed by another person, perform movement correctly in response to a seen object, or handle an object correctly.[1,2] Apraxia commonly results in significant impairment in independent daily functioning.

An influential model of praxis[3,4] describes 2 routes of movement planning and execution—a lexical and nonlexical route. In the "lexical route," the meaning of auditory (verbal) or visual (gestural) input is transformed to motor output via a semantic component stored in central semantic memory. In the "nonlexical route," imitation of meaningless and novel gestures is achievable without requiring action semantics. More recently, the 2-route model has been challenged by several lines of evidence necessitating the inclusion of representations of the body. Neural representations of the body are classified according to 3 domains: a dynamic representation of the body in action, a structural representation of the relationships between body parts and a semantic representation of body parts and their function.[5,6] Brain damage can selectively affect 1 or more of these body representations, depending on injury location and severity. Specifically, the parietal cortex is known to be important for generating mental representations of the body, and involvement of superior parietal lobe,[7] intraparietal sulcus, and inferior parietal lobe[8,9] has been implied. Unilateral parietal damage may impair the formation of the dynamic body scheme.[10] In patients with parietal lesions, time estimation for

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performing imagined hand gestures is impaired, as this estimation requires mentally performing the motor task using a dynamic representation of the body.[11] Buxbaum et al[12] describe a patient with apraxia expressed by impairments in imitation of meaningless gestures and tasks requiring spatiomotor transformations of the body, but with preserved ability for tool use. These authors suggest that a dynamic representation of the body (body scheme) is necessary to perform gesture imitation. It has also been suggested that structural knowledge regarding the relative positioning of body parts is required for gesture imitation.[13] Finally, a semantic representation of the body is required for performing meaningful movements.[5]

We present a case of apraxia and body scheme disorder in a 22-year-old woman (RS) who had a severe brain injury with bilateral frontal and occipitoparietal infarctions at the age of 11.5 years. Immediately following injury, RS displayed disrupted body representation in all 3 aspects (see below). However, 10 years later, RS lives a completely independent life and performs activities of daily living with specific limitations. The objective of this study was to describe the recovery process of RS from a severe childhood brain insult to almost full independence in activities of daily living as an adult. RS's residual praxis and body scheme impairments were mapped 10 years postinjury, in order explain her current limitations.

Case Description

RS is a 22-year-old woman who was admitted to hospital at age 11.5 years following a fall from stairs. She was initially in a state of moderate lack of consciousness (Glasgow Coma Scale score 9), with bilateral prefrontal damage in proximity to midline. During her hospitalization in intensive care, she developed high intracranial pressure and was diagnosed with brain edema. Her condition deteriorated and 10 days postinjury, a magnetic resonance imaging (MRI) scan showed additional extensive bilateral occipitoparietal infarctions.

One month post injury, RS was admitted to the pediatric rehabilitation unit. She was disoriented and cortically blind, with right hemiparesis. She was unable to follow complicated instructions and unable to touch her body parts following verbal instruction or name them.

During rehabilitation (6 months as an inpatient), she showed significant improvement in her visual perception and was partially able to recognize objects in her environment. However, she continued to demonstrate remarkable impairments, including visual field deficit, visual agnosia, simultanagnosia (disturbances of visual orientation), optic ataxia, and ocular apraxia.[14] In addition, she showed left-right disorientation, dyscalculia, and finger agnosia. She was able to identify single letters and rarely read a short 3-letter word but was unable to perform visual tasks on meaningless objects (ie, simple geometric forms). In general, her ability to process meaningful visual stimuli was superior to her ability to process meaningless stimuli. This was also exemplified by her ability to read short words as compared to nonwords. Notably, difficulties in naming her body parts persisted (naming of objects in general was better, but below average; see Table 1),

Table 1. Summary of RS's Neuropsychological Assessments, 6 Months From Injury, 4 Years From Injury, and 10 Years Postinjury.[a]

Measures	Age 12 y 1 mo	Age 15 y 10 mo	Age 22 y 10 mo
Verbal IQ[b]	96	95	110
Information	6	8	10
Similarities	11	13	12
Vocabulary	8	9	14
Comprehension	13	9	11
Performance IQ[b]	NA	NA	NA
Visual organization[c]	−3	−3	−3
Visual discrimination[d]	−3	−3	−3
Visuomotor integration[e]	−3	−3	−3
Verbal fluency[f]	−0.4	−0.3	−0.8
Naming[g]	−1.4	−1	−1.4
Verbal short-term memory[h]	−1	0.7	−1
Verbal long-term memory[i]	0	0.1	0.5
Executive functions[j]		107	109

[a]For Wechsler and Wisconsin Card Sorting Test, scaled scores are reported. For all other tests, z scores are reported.
[b]Verbal and performance IQ: assessed at ages 12 and 15 by Wechsler Intelligence Scales for Children–Revised, at age 22 by Wechsler Adults' Intelligence Scales–3.
[c]Visual organization: Hooper visual organization test.
[d]Visual discrimination: Motor-free Visual Perception Test-3.
[e]Visuomotor integration: Beery-Buktenica Developmental test of Visuomotor integration.
[f]Verbal fluency: phonetic and semantic.
[g]Naming: assessed at age 12 by Boston Naming Test and at ages 15 and 22 by Hebrew Verbal Fluency Test, Kave.
[h]Verbal short-term memory: Digit Span/Wechsler Intelligence Scales.
[i]Verbal long-term memory: Rey Auditory Verbal Memory Test, Delay Score.
[j]Executive functions: Wisconsin Card Sorting Test—total no. of errors.

along with difficulties in identification of body parts following touch and in localization of body parts in relation to one another. As a consequence, RS showed great difficulties in dressing and required verbal mediation (ie, directionality—front/back, right/left). She managed to use familiar objects (cutlery) but could not imitate or identify either meaningless or meaningful gestures without an object. During rehabilitation, muscle strength in her right side improved as well as her ability to perform deliberate and accurate movements, but praxis deficits were still noticeable. She was unable to estimate the distance between herself and objects in her surroundings, such that it was impossible for her to deal with obstacles in her environment and to remain stable in certain postures, in spite of intact basic sensorimotor abilities. Although RS was able to use real objects and to perform meaningful gestures related to her body following verbal instructions ("show me how you would smell a flower," etc), significant difficulty was still noted in imitation of meaningless gestures and activities such as dressing (of herself and others). Naming and localization of body parts were still impaired, but in a follow-up 2 years postdischarge these were reported to be normal.

Ten years postinsult at the age of 22, no additional structural changes occurred in RS's MRI scans compared to her postinjury scan (current scan in Figure 1). Functionally, however, RS recovered remarkably and lives a completely independent life. Her neuropsychological profile at 3 different time points

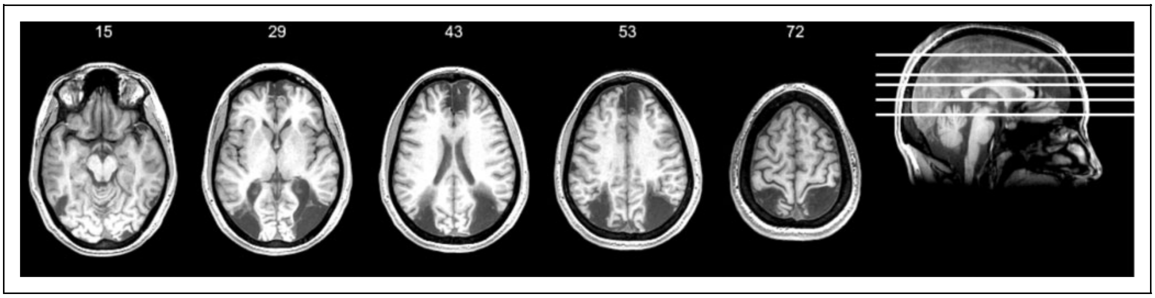

Figure 1. Current structural state as revealed by recent magnetic resonance imaging (MRI). Axial slices reveal gliosis and extensive cystic encephalomalacic changes in bilateral occipital, parietal, and anterior frontal lobes. No significant changes were shown compared to prior (postinjury) scans.

Table 2. Summary of Evaluations Administered to RS.

Purpose of assessment	Assessment	Includes
Praxis	Dynamic Occupational Therapy Cognitive Assessment for Children (DOTCA-Ch)[15]	Meaningless gesture and sequence imitation (verbal cues, demonstration)
		Meaningful actions with and without objects
Body representations		
Dynamic body scheme	Hand Laterality Test[16]	Mental rotation task—laterality judgment following presentation of visual stimulus of hand
Structural description	Pointing to body parts following verbal instruction (Cratty and Sams's assessment of body image for blind children[17])	Point to own body parts
		Point to examiner's body parts
	Positioning of body parts	Name adjacent body parts
		Determine relative positioning of body parts
Semantic representation	Naming of body parts	Name own and examiner's body parts following touch
	Naming the function of body parts	

(6 months after injury, 4 years and 10 years after injury) is described in Table 1. RS has residual visuospatial problems (tunnel vision and visuospatial disorientation), but is completing her studies in college to become a special education teacher. She performs all basic and instrumented activities of daily living without assistance; she cooks, cuts vegetables for a salad, etc. Still, RS reports persisting difficulties with novel tasks, specifically those involving her body. For example, when buying new shoes, she is unable to determine which is the right and which is the left; novel items of clothing require time and practice until she is able to use them. She reports an inability to dance, and tying a scarf on her hair involves extensive practice. Thus, specific difficulties with praxis and body representation persist even 10 years postinsult. Interviews with her reveal a young woman with exceptional determination and character. RS verbalizes her difficulties by stating that following her injury, "a new body was given" to her and she needs to familiarize herself with it anew. A comprehensive evaluation of praxis and body scheme was performed as detailed below, in order to identify the focus of these difficulties.

Evaluation of Praxis and Body Scheme in Patient RS

Praxis and body representation evaluations took place during a single, 1-hour session at the pediatric rehabilitation department. A summary of the evaluations is detailed in Table 2. RS gave informed consent according to the hospital's ethics committee requirements.

Praxis

Praxis was evaluated using the Dynamic Occupational Therapy Cognitive Assessment for Children (DOTCA-Ch).[15] A pediatric evaluation was selected because of its increased level of detail and sensitivity for praxis function, in comparison to the adult version (Loewenstein Occupational Therapy Cognitive Assessment).

Meaningless posture and movement imitation. RS was required to imitate 3 meaningless static postures and 5 meaningless movement sequences following the examiner's demonstration and 4 meaningless postures following verbal cues from the examiner (eg, "put one hand on the other shoulder and the second hand behind your back"). RS was able to imitate 1 of 3 static postures (33% success) and 1 of 5 movement sequences (20%). However, she was successful in imitating all 4 postures following verbal cues (100%).

Meaningful actions with and without objects. RS was asked to perform 4 meaningful actions with objects (paper and an envelope, a maze, etc) and 5 meaningful actions with no object (eg, "Imagine there is a loaf of bread on the table. Show me how

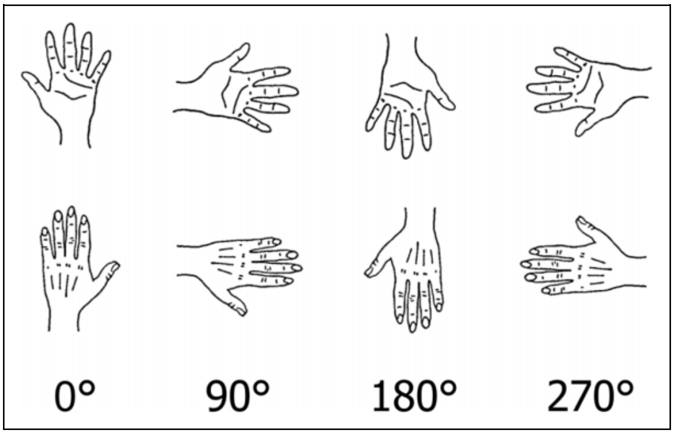

Figure 2. Experimental conditions for the Hand Laterality Test.[16] Subjects viewed images of hands in palmar (top row) or dorsal (bottom row) view, presented in 1 of 4 rotation angles. Subjects were asked to determine whether the hand was right or left, and decision time was recorded.

you slice the bread and spread it"). RS was successful in performing 2/4 actions with physical objects (50%) and 1/5 actions with no object (20%). Her difficulties in performing this task involved using her body as the object (eg, her finger as the knife to slice the bread).

Evaluation of body representations was performed according to 3 domains as described in the literature. Where possible, existing standardized clinical evaluations were used.

Dynamic body scheme

Dynamic body scheme was evaluated using a computerized version of the Hand Laterality Test[16] and compared to reference data from N=14 female volunteers, aged 28.3 ± 4.9 years. A stimulus of a right or left hand was presented, rotated clockwise at 1 of 4 angles (0°, 90°, 180°, 270°) and 1 of 2 orientations (dorsal, palmar) (Figure 2). Subjects, sitting in front of the screen with their hands facing down, were requested to press a key indicating whether the image was of the right or left hand (all control subjects and RS were right-handed). The time required to make a decision was recorded. Each combination of angle and orientation was presented twice, for a total of 32 attempts. A fixation cross was presented for 2 seconds between stimuli. The task was coded using custom-written Matlab code (Matlab R2015b, Natick, MA) with the PsychToolbox-3 extension.[18]

RS's and controls' performance in the Hand Laterality Test is presented in Figure 3. Reaction time of healthy subjects in this task correlates with the difference between the position of the hand in space and the current position of the subject's hand (orientation angle), suggesting that the strategy used to solve the task is a mental rotation of the subject's hand to the orientation of the stimulus.[19] In the current study, controls were correct on 86% of cases (72%-100%) and took 2.23 ± 0.5 seconds to answer. As in previous studies,[17,20] our results from controls revealed a dependency of reaction time on orientation angle ($F_{3,39}$ = 24.80, P < .001), as well as shorter reaction times for

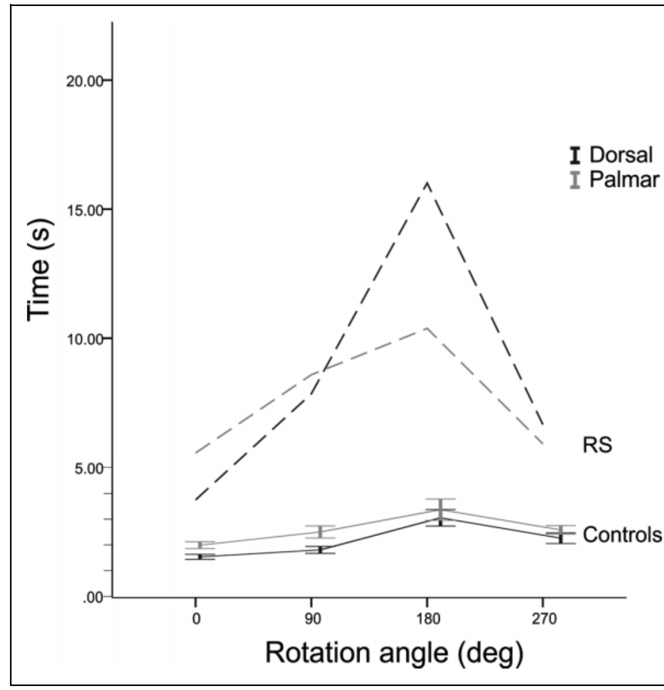

Figure 3. Decision times in the Hand Laterality Test for RS (dashed line) and controls (solid line) as function of rotation angle, in dorsal (black) and palmar (gray) views. Error bars for control data mark 1 standard error of the mean.

dorsal vs palmar views (RM-ANOVA; $F_{1,13}$ = 20.87, P < .002). RS was correct on 59% of the cases, with no difference between orientation angles (Kruskal-Wallis H test; $\chi^2(3)$ = 2.38, NS) or type of view (dorsal/palmar; $\chi^2(1)$ = 0.01, NS). However, the relationship between reaction time and angle that was identified for controls was maintained in RS's data ($\chi^2(3)$ = 20.02, P < .001), such that angles that were biomechanically "awkward" were associated with longer response times. This suggests that mental rotation of the arm was attempted by RS to solve the task. Indeed, when asked about her strategy in solving this task, she said she attempts to rotate the hand but then tries to find "hints" such as which direction the thumb is facing.

Body structural description

Body structural description was assessed by asking RS to touch body parts following verbal instruction to do so. Body parts were selected according to Cratty and Sams's assessment of body image for blind children[21]: 5 simple body parts (eg, right arm, left hand), 5 more complex body parts (eg, right wrist, left thigh), 5 parts of the face (eg, left ear, nose), and 5 fingers. RS was asked to point to her own body parts and subsequently—point to body parts of the examiner according to similar instructions. Additionally, RS was asked to name the body part adjacent to a given body part (eg, "what body part is below the thigh?"), as well as to determine relative positioning of body parts (eg, "is the knee above or below the shin?").

RS made no errors pointing to her own or the examiner's body parts (20/20, 100%). However, she continuously verbalized her strategy of solving the task (eg, "if it is your right hand it is supposed to be on my left"). RS was able to correctly name the adjacent body parts (10/10, 100%) and answered 9/10 questions of relative positioning correctly (90%), where the question she answered wrong was related to the position of the thumb with respect to the wrist.

Semantic representation of body parts

Semantic representation of body parts was evaluated using (1) naming of own and examiner's body parts[22] and (2) naming of the function of body parts.[23] RS was requested to name 10 body parts (eg, nose, ear, hair, neck, etc) following the examiner's pointing to the body part on RS's body or the examiner's body.

RS named the body part correctly in all cases (20/20, 100%). RS was also successful (100%) in naming of the function of 5 body parts (mouth, foot, ear, knee, and elbow).

Discussion

In this study, we present a 10 years' follow-up assessment of praxis and body representations in a patient following childhood bilateral frontal and significant occipitoparietal brain damage. Assessment revealed persistent deficits in praxis as well as major deficits in dynamic body scheme, but restored semantic body representation and relatively intact structural body representation. Compared with her initial postinsult performance, at 10 years postinsult RS displays remarkable independence in activities of daily living but is still unable to perform tasks relying heavily on dynamic representations of her own body (eg, dance) and/or that of other individuals (eg, change a diaper). In our praxis assessment, RS performed poorly on gesture imitation tasks (unless verbal cues were provided). RS performed remarkably well on tasks requiring semantic knowledge of the human body, whereas in tasks that required structural body description RS performed well but was constantly verbalizing the task—that is, using semantic information as cues to solve the task.

RS's dynamic body representation was evaluated by the Hand Laterality Test.[16] This test of mental imagery has been used extensively in healthy individuals[18,20,24,25] indicating consistently that biomechanical constraints of arm movement dictate the time required to perform laterality judgement of a visual stimulus (hand). Variants of this task were used to assess dynamic body representation in people with brain damage following injury. For example, people with unilateral damage to the left hemisphere were shown to perform hand laterality judgement less accurately than people with right hemisphere damage or controls.[26] However, in the current study the Hand Laterality Test was used as means to evaluate a severe dynamic body scheme disorder in a person who has developed compensatory strategies following an extensive childhood brain lesion and has achieved exceptional functioning in everyday life. These strategies use semantic information (see below), but our use of the Hand Laterality Test quantified the extent of the deficit in dynamic body scheme without enabling compensatory strategies of other domains, which were relatively preserved.

Our results support the work of Buxbaum et al,[12] who showed that the "2-route" model of praxis[3] cannot fully account for ideomotor apraxia. RS's praxis impairment decreased her ability to perform meaningful as well as meaningless gestures. Similarly to the results of Buxbaum et al, RS also showed a significant impairment in dynamic body scheme. However, RS's praxis abilities were improved when performance was initiated by verbal cues, as well as when actual tools were involved. Thus, RS was able to access stored gesture representations when provided with verbal hints or tools. Similarly to the patient described by Buxbaum et al,[12] RS was less accurate than control subjects when performing the Hand Laterality Test. However, the current study evaluated both the accuracy and timing of laterality judgment and showed that despite long latencies, the typical relationship between angle and response time was maintained. Thus, RS was not completely unable to perform mental rotation but rather did so with diminished effectiveness. Indeed, people with traumatic brain injury are able to perform mental imagery tasks such as the Hand Laterality Test,[27] but their reaction times are significantly longer than controls. It is difficult to compare performance of people with parietal lesions to those of other patients on this task because of lack of information on lesion location and severity in previous reports.[27] However, RS's extensive injury may at least partially explain her reaction times, which were twice as long as those reported by Oostra et al.[27] It could be argued that RS's longer response times in the Hand Laterality Test are associated with her residual visual deficits. However, if indeed this is the case, it can be expected that equally long response times would be registered for all rotation angles of stimuli (because the same image needs to be processed) and this was not the case (Figure 2). Thus, we argue that the origin of RS's slow response is in her diminished capability of performing mental rotations, which are attempted nonetheless (as she also attested when asked).

The ability to use mental representations of the body is important for praxis.[28,29] Specifically, it is the dynamic body representation that is crucial for praxis functions.[30] Indeed, RS's impaired performance on the Hand Laterality Test is evidence of significant damage to her ability to construct a dynamic body representation due to injury in the occipitoparietofrontal pathway (dorsal stream). This damage is clearly visible in RS's recent MRI scans, which are evidence that no structural plasticity occurred in the 10 years following her injury. RS's dorsal stream deficits involve both aspects of perception as well as action (both dorsodorsal and ventrodorsal streams), as evidenced by her residual deficits in visual performance (eg, optic ataxia) as well as in reaching and object manipulation.[31] However, the fact that RS displayed a pattern of mental rotation similar to that of controls (albeit with lowered accuracy and longer response times), along with her ability to perform activities of daily living, suggests remarkable functional plasticity. It may be that spontaneous recovery processes, together with extensive rehabilitation and RS's high motivation and intellectual abilities, led to the ability of a different neural pathway, such as the ventral stream, to compensate for dorsal stream deficits. A similar compensatory mechanism of an intact ventral stream has been shown in the case of visual processing among patients with simultagnosia due to posterior parietal damage.[32] Supporting this hypothesis is the fact that RS's ability to construct a semantic representation of the body has improved dramatically since her injury and has reached a normal level. Additionally, RS constantly used verbalizations to solve tasks associated with body representations. Indeed, although her injury includes prefrontal damage, it is mostly ventromedial, and her orbital frontal cortex is intact (Figure 1). The intact orbital frontal cortex, associated with determining object identity,[33] can assist RS in object identification, and semantic information can be used to determine connections between objects, including body parts (top-down processing). This compensation, if indeed it is in effect here, may be less than optimal, as RS still complains of an inability to perform activities of daily living that require on-line representation of her body or that of others (eg, dance or change a diaper). However, the fact that RS lives an independent life despite significant apraxia suggests she may be able to use semantic information to compensate for deficits in dynamic and structural body representations also in her everyday life.

One factor that may be associated with RS's remarkable outcome is her age at the time of injury. Age at injury has been related to positive cognitive outcome among children with brain injury; children injured prior to school age often demonstrate slower recovery patterns and poorer cognitive outcomes.[34] This may be because most cognitive abilities improve extensively during early childhood (until the age of 11-12 years) and are followed by a more gradual improvement rate until they are well stabilized during adulthood (18-20 years).[35] RS was injured at the age of 11.5, when most of her cognitive skills have been acquired. This fact may have assisted her in using various compensation strategies to achieve a high level of everyday functioning.

Our results may be informative for clinicians facing similar cases. The gap between RS's high level of everyday performance and the results of her assessments reported here highlights the importance of a detailed assessment of praxis and of the 3 representations of the body. The majority of evaluations performed here (Table 2) are taken from standardized tests, and the Hand Laterality Test may be coded using off-the-shelf software (Matlab, PsychToolbox). By examining all aspects of praxis and body representations, clinicians may be able to provide a more complete overview of the impairment in body scheme and its underlying mechanism and subsequently design specific remedial or compensatory strategies (e.g. semantics) to achieve functional independence in these cases.

This study has several limitations. First, the evaluation protocol of body representations reported here was not performed early after injury but only 10 years after. Thus, although we were able to verify RS's anatomical status to determine that no anatomical changes occurred within a 10-year period, we were unable to compare RS's performance on the same tests to directly evaluate functional recovery. However, since RS was initially unable to identify body parts at all, nor maintain gaze due to ocular apraxia, performance of the same test battery would have been impossible at an early stage of recovery. Second, the evaluation presented here of the Hand Laterality Test would have been more meaningful if functional MRI were used in conjunction with the motor imagery task. Indeed, a recent study used this task and demonstrated specific areas of brain activation associated with it in young healthy adults.[20] Unfortunately, the latencies of RS's responses in the Hand Laterality Test were too long to enable identification of temporally relevant brain activation areas. An additional limitation to this study is related to the extensiveness of RS's injury. Alongside the parietal injury that contributes to the impairment in dynamic body scheme, RS has prefrontal damage, which may contribute to her praxis impairment. However, as her neuropsychological evaluations show, her executive functions and verbal short- and long-term memory functions are intact and no indication of executive function impairments in daily life have ever been reported—a fact that probably assists her in compensating for her extensive injury and in achieving a normal level of performance in everyday life.

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Author Contributions

TK and JL have equally contributed to this work. TK and JL: study design, data analysis, manuscript writing, reviewing, and editing. OB: study design, data collection, manuscript reviewing, and editing. AAJ: manuscript writing, reviewing, and editing. AL: data collection, image analysis, manuscript reviewing, and editing. GT: data collection, image analysis, manuscript reviewing, and editing. TS: manuscript writing, reviewing, and editing.

Declaration of Conflicting Interests

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Ethical Approval

The study has been approved by the institutional ethics committee (3073-16-SMC).

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