DOI: 10.1002/ejp.1860

EJP European Journal of Pain

ORIGINAL ARTICLE

Body representation among adults with phantom limb pain: Results from a foot identification task

Emma H. Beisheim-Ryan[1] | **Ryan T. Pohlig**[2] | **Jared Medina**[3] | **Gregory E. Hicks**[1] | **Jaclyn M. Sions**[1]

[1]Department of Physical Therapy, University of Delaware, Newark, Delaware, USA

[2]Biostatistics Core, University of Delaware, Newark, Delaware, USA

[3]Department of Psychological and Brain Sciences, University of Delaware, Newark, Delaware, USA

Correspondence
Jaclyn M. Sions, Department of Physical Therapy, University of Delaware, 540 S. College Avenue, Suite 210JJ, Newark, DE 19713, USA.
Email: megsions@udel.edu

Funding information
This work was supported by the Eunice Kennedy Shriver National Institute of Child Health and Human Development of the National Institutes of Health [T32HD007490] and Promotion of Doctoral Studies I and II scholarships from the Foundation for Physical Therapy Research. The content is solely the responsibility of the authors and does not necessarily represent the official views of the funding institutions.

Abstract

Background: Impaired body representation (i.e. disrupted body awareness or perception) may be a critical, but understudied, factor underlying phantom limb pain (PLP). This cross-sectional study investigated whether adults with lower-limb loss (LLL) and PLP demonstrate impaired body representation as compared to Pain-Free peers with and without LLL.

Methods: Participants (n = 41 adults with PLP, n = 27 Pain-Free peers with LLL, n = 39 Controls with intact limbs) completed an online foot identification task. Participants judged whether randomized images depicted left or right feet (i.e. left–right discrimination) as quickly as possible without limb movement. Using two Generalized Estimating Equations, effects of group, image characteristics (i.e. side, foot type, view, angle) and trial block (i.e. 1–4) were evaluated, with task response time and accuracy as dependent variables ($a \leq 0.050$).

Results: Adults with PLP demonstrated slower and less accurate performance as compared to Controls with intact limbs ($p = 0.018$) but performed similarly to Pain-Free peers with LLL ($p = 0.394$). Significant three-way interactions of group, view and angle indicated between-group differences were greatest for dorsal-view images, but smaller and angle-dependent for plantar-view images. While all groups demonstrated significant response time improvements across blocks, improvements were greatest among adults with PLP, who also reported significant reductions in pain intensity.

Conclusions: Adults with PLP demonstrate body representation impairments as compared to Controls with intact limbs. Body representation impairments, however, may not be unique to PLP, given similar performance between adults with and without PLP following LLL.

Significance

Following lower-limb loss, adults with phantom limb pain (PLP) demonstrate impaired body representation as compared to Controls with intact limbs, evidenced by slower response times and reduced accuracy when completing a task requiring mental rotation. Importantly, 80% of participants with pre-task PLP reported reduced pain intensity during the task, providing compelling evidence for future investigations into whether imagery-based, mind-body interventions have positive effects on PLP.

1 | INTRODUCTION

Each year, approximately 185,000 amputation surgeries are performed to address limb compromise secondary to vascular conditions, infections, trauma, or cancer (Owings & Kozak, 1998). As the number of individuals living with lower-limb loss (LLL) progressively rises (Harding et al., 2020), there is a growing need for specialized clinical care that addresses the unique and debilitating consequences of amputation.

One of the most common and perplexing consequences of LLL is the perception of a phantom limb, that is, sensations perceived as coming from the amputated portion of the limb (Pirowska et al., 2014). A recent meta-analysis estimated 64% of adults with LLL experience painful phantom limb sensations [e.g. aching, electric shocks, painful shortening (Limakatso et al., 2020)]. Phantom limb pain (PLP) appears resistant to localized treatment [e.g. massage, nerve blocks (Hanley et al., 2006)], indicating the importance of evaluating and addressing central nervous system impairments in PLP management.

Body schema impairments, or impaired awareness and representation of the body in space, may underlie PLP. An intact body schema requires integration of complex, multisensory signals (e.g. touch, vision, proprioception) and informs action-oriented tasks, such as movement (Medina & Coslett, 2010; Pitron et al., 2018). When communication between the brain and body is compromised, however, as in the case of peripheral nerve lesions (e.g. amputation), the body schema may become compromised (Lustenhouwer et al., 2020; Moseley et al., 2012). Clinically, body schema impairments can be assessed using a left–right discrimination task (Parsons, 1987), which involves judging the sidedness (i.e. left vs. right) of a body part image. Task completion facilitates mental rotation of one's own body, which is referred to as 'implicit motor imagery' (Bowering et al., 2013). Delayed response time or reduced accuracy may reflect body schema impairments (Parsons, 1987; Parsons & Shimojo, 1987).

In other pain conditions (e.g. low back pain, complex regional pain syndrome), rehabilitative treatments targeting body schema impairments have been shown to ameliorate pain and improve function (Bowering et al., 2013; Louw et al., 2015, 2017; Mendez-Rebolledo et al., 2017; Moseley, 2004a). Recent research has suggested body schema impairments may play a key role in PLP development and persistence (Giummarra et al., 2007, 2010); however, clinical evidence of body schema impairments remains scarce post-LLL.

While adults with upper-limb loss appear to demonstrate body schema impairments (Nico et al., 2004; Reinersmann et al., 2010), it remains unclear if findings are generalizable to LLL, as the somatosensory region representative of the lower-limbs is significantly smaller than that of the upper-limbs (Penfield and Boldrey, 1937). Furthermore, lower-limb prostheses are more functional than upper-limb prostheses (Guo et al., 2017) and may be more easily embodied as one's 'own' limb, potentially reducing body schema changes post-LLL (Boccia et al., 2020).

This study aimed to investigate whether adults with PLP demonstrate body schema impairments, as compared to Pain-Free peers and Controls with intact limbs. We hypothesized adults with PLP would demonstrate increased response time and reduced accuracy when identifying amputated-side foot images during a left–right discrimination task.

2 | METHODS

This cross-sectional pilot study was conducted remotely via an online experimental platform [Gorilla™ (Anwyl-Irvine et al., 2020)]. Participants were recruited from August of 2020 to March of 2021 via the interdisciplinary University of Delaware Amputee Clinic; online advertisements through national organizations (e.g. Amputee Coalition); local prosthetic clinics and limb loss support groups; consent-to-re-contact databases from prior studies in the Delaware Limb Loss Studies Lab; and community events. This study was approved by the University of Delaware Institutional Review Board for Human Subjects Research.

Three groups of participants were recruited: (1) adults with LLL and PLP, (2) age-matched adults with LLL who denied PLP or pain in the remaining portion of their limb (i.e. residual limb pain; Pain-Free) and (3) age-matched, Pain-Free adults with intact limbs (Control). Pain-Free and Control groups were recruited for comparison purposes to investigate whether body schema impairments were specific to PLP, rather than the presence of LLL alone.

For all groups, adults were included if they were English-speaking and-reading (as instructions and self-reported outcome measures were administered in English), had reliable internet service and reported basic computer skills (i.e. ability to type on a computer keyboard and explore the internet independently). Inclusion criteria specific to the PLP and Pain-Free groups included unilateral LLL at or above the transtibial (i.e. below-knee) level that occurred ≥1-year prior and current prosthesis use [as changes in body representation may be affected by prosthesis use (Guo et al., 2017)]. For all participants, exclusion criteria included age >75 years, as older age has been associated with worse performance on the left–right discrimination task (Saimpont et al., 2009); and systemic neuromuscular conditions (e.g. multiple sclerosis, Parkinson's disease), as these may interfere with response time. Exclusion criteria specific to the Pain-Free and PLP groups included congenital amputation aetiology (i.e. amputation present at birth) and formal participation in imagery-based treatment programmes for PLP (e.g. graded motor imagery, mirror therapy), as previous training may affect performance (Ramachandran et al., 2010, 2018; Steenwinkel et al., 2019).

Upon signature of an electronic informed consent form supplied via REDCap (Research Electronic Data Capture) tools (Harris et al., 2009, 2019) hosted at the University of Delaware (which allowed participants to download a copy of the informed consent for their records), participants were emailed a link to Gorilla for access to the study. In Gorilla, all participants provided basic demographic information, including hand and foot dominance. Participants in the PLP group reported pre-task PLP intensity on a numeric pain rating scale [0 = no pain, 10 = worst pain imaginable (Chiarotto et al., 2019)].

2.1 | Left–right discrimination task

All participants then completed a left–right foot discrimination task, which was adopted from the protocol reported by Stone et al. (2019). Instructions were displayed prior to the task, asking participants to sit comfortably with their prosthesis off (if applicable) and their intact foot (or feet) flat on the floor. Participants were instructed to drape a towel or blanket over their legs and to not move their legs, ankles or feet during the task, to reduce the impact of vision and/or movement on task performance. Finally, participants were instructed to judge, as quickly and accurately as possible, whether a foot shown on their computer screen was a left or right foot, indicating sidedness by pressing the computer arrow key corresponding to the foot side (i.e. left arrow key for left foot, right arrow key for right foot).

After a practice trial was completed, four blocks of images were shown. In each block, 48 foot images were presented twice, in random order, for a total of 96 trials per block. Using an image set from Stone et al. (2019) and Curtze et al., (2010) the 48 images included right and left, human and prosthetic feet, presented from the plantar (i.e. bottom-up) and dorsal (i.e. top-down) position and rotated laterally at six angles (i.e., 0°, or anatomical position; 60°; 120°; 180°; 240° and 300°). The same image set was used for each block, resulting in a total of 384 trials per participant, and each block took approximately 4 min to complete.

Between blocks, a 'break' screen populated, encouraging participants to take a break (of self-selected duration) to reduce the potential impact of mental fatigue. After task completion, participants in the Pain-Free group reported whether non-painful phantom sensations were experienced during the task, and participants in the PLP group rated their PLP intensity during the task on the abovementioned numeric pain rating scale.

2.2 | Self-reported outcome measures

For descriptive purposes, questions from the Limb Deficiency and Phantom Limb Pain Questionnaire (Goller, 2012) were used to assess the presence, frequency and characteristics of phantom limb sensations and PLP. Phantom limb sensation and PLP bothersomeness were captured using a 3-point Pain Bothersomeness Scale, ranging from 0 (not bothered at all) to 2 [extremely bothered (Ephraim et al., 2005)]. Best and worst PLP intensity in the prior 24 h was recorded on the abovementioned numeric pain rating scale and averaged. Finally, the Brief Pain Inventory-Short Form (BPI-SF) Pain Interference Domain (Poquet & Lin, 2016) was used to evaluate the degree to which PLP interfered with daily activities in the previous 24 h. Scores from all seven domains (i.e., general activity, mood, walking ability, normal work, relations with other people, sleep and enjoyment of life), scored from 0 (does not interfere) to 10 [completely interferes (Poquet & Lin, 2016)], were averaged. Reliability and validity of the BPI-SF has been reported in several patient populations (Ehde et al., 2015; Hand et al., 2018; Mendoza et al., 2006). Participants in the PLP group were also asked similar questions regarding the presence, intensity (on average in the past 7 days) and bothersomeness of residual limb pain for characterization purposes.

2.3 | Data management and statistical analyses

Prior to conducting statistical analyses, overall accuracy was evaluated per participant per block to identify and remove participants whose accuracy could be considered at or near chance (i.e., <60%). Furthermore, based on text entries regarding postural changes during the task, participants were removed if they reported frequent movement of the limbs to assist with image identification. Finally, for response time analyses, incorrect trials were excluded, and response times <200 ms and >2.5 standard deviations from each participant's mean per block were identified and removed. Response time cut-points were imposed to limit the influence of responses falling outside a realistic response time window (e.g. exceptionally fast responses due to accidental key press, exceptionally slow responses due to distraction) and were based on comparison to prior literature regarding left–right discrimination (Lee et al., 2021; Parsons & Shimojo, 1987; Stone et al., 2019).

In SPSS 26 (IBM), two, separate Generalized Estimating Equations [GEE (Liang & Zeger, 1986)], each with an exchangeable working correlation matrix, were used to evaluate response time and accuracy. For response time, the model was specified with a gamma distribution and log link function and naïve (i.e., model-based) errors, given response time's positive skew (all Shapiro–Wilk tests: $p < 0.001$). For accuracy, the model was specified with a binomial distribution (i.e., 0 = incorrect, 1 = correct) with logit link function and robust errors, such that overall accuracy was calculated by the model as a proportion of the total number of correct trials per group and condition. GEE is an extension of the Generalized Linear Mixed Model that provides accurate parameter estimates for repeated measures or clustered data, even if the correlation structure is mis-specified (Griswold et al., 2013). This is because the correlation matrix is treated as a nuisance parameter, and estimation is based on quasi-likelihood (Griswold et al., 2013). GEEs can be contrasted with Generalized Linear Models: given a set of predictors, the marginal mean is modeled instead of a mean that is conditional on random effects (Heagerty & Zeger, 2000). GEEs account for within-group, non-independence of data and missing data [e.g. trials removed due to imposed response time cut-points (Hubbard et al., 2010)].

The GEE models included the following main effects: *group* (Control, Pain-Free, PLP), *block* (1, 2, 3, 4), *side* [amputated (or non-dominant, for Controls), intact (or dominant, for Controls)], *view* (dorsal, plantar), *type* (human, prosthetic), *angle* (0°, 60°, 120°, 180°, 240°, 300°), *repetition* (first image repetition per block, second image repetition per block), two-way interactions with group (*group* * *block*, *group* * *side*, *group* * *view*, *group* * *type*, *group* * *angle*) and the three-way interaction of *group* * *view* * *angle*. All image factors were entered into the model as nominal variables. The three-way interaction tests whether group performance differs when viewing images requiring greater implicit motor imagery [i.e., plantar vs. dorsal view and rotations further from 0° (Schwoebel et al., 2001)]. The inclusion of *group* * *side* and *group* * *type* interactions tests for differential group performance by the type of foot depicted or when the affected side was presented (for adults with LLL). Finally, as impairments in left–right discrimination may improve with practice differently between groups (Reinersmann et al., 2010), a *group* * *block* interaction was evaluated. Post hoc pairwise comparisons were assessed using Fisher's Least Significant Differences to account for multiple comparisons while preserving the error rate ($a = 0.050$).

3 | RESULTS

Among the 411 individuals (i.e., 358 adults with LLL, 53 adults with intact limbs) contacted for this study, 195 were screened for inclusion (Figure 1

). Of the individuals who were screened, 136 adults (i.e., 96 with LLL, 40 Controls) were eligible and provided with electronic informed consent forms. In total, of the 114 participants who completed the study, 105 were included in final analyses (37 Controls, 27 Pain-Free, 41 PLP).

3.1 | Participant characteristics

Participants were predominantly right-handed, right-footed and middle-aged (Table 1). Females comprised 38%–45% of all groups. Participants with LLL predominantly had a transtibial (i.e., below-knee) amputation that had occurred several years prior (median: 8 years), and 56%–64% of participants had used a prosthesis for >5 years (Table 2). No significant differences in demographics or amputation-specific characteristics were present between groups ($p > 0.050$), with the exception of weight, which was significantly higher among adults in Pain-Free and PLP groups as compared to Controls.

Additional phantom limb and prosthesis use characteristics are provided in Table 2. Among adults with LLL, non-painful phantom limb sensations were similarly prevalent in both PLP and Pain-Free groups, but 69%–85% of participants reported they were not bothersome (Table 2). Among adults in the PLP group, residual limb pain was reported by 26 (63%) of participants and was largely considered ‘somewhat’ bothersome. Furthermore, average PLP intensity in the past 24 h was mild, but 80% of participants considered PLP ‘somewhat’ or ‘extremely’ bothersome. PLP most commonly occurred ‘1–3 times per week,’ and lasted ‘several minutes but <1 hour.’ Pain interference in the past week was relatively low (median: 1.29).

In the Pain-Free group, 5 (19%) participants reported experiencing phantom limb sensations during the task. Participants described phantom sensations as, ‘tingling,’ or feeling as though specific body parts (e.g. ‘big toe,’ ‘ankle’) were present and moving. Interestingly, one participant in the Pain-Free group reported phantom limb sensations of ‘pulsating/throbbing,’ and reported that the task increased pain in his residual limb (intensity: 5/10), which he did not experience on a day-to-day basis.

The majority of the PLP group ($n = 25$; 61%) reported mild pre-task PLP (median intensity: 2/10). Among the 25 participants who reported pre-task PLP, 20 (80%) reported a reduction in PLP intensity during the task. Moreover, 16 (64%) reported complete relief from their pre-task PLP (i.e., 100% reduction in pain intensity, ranging from

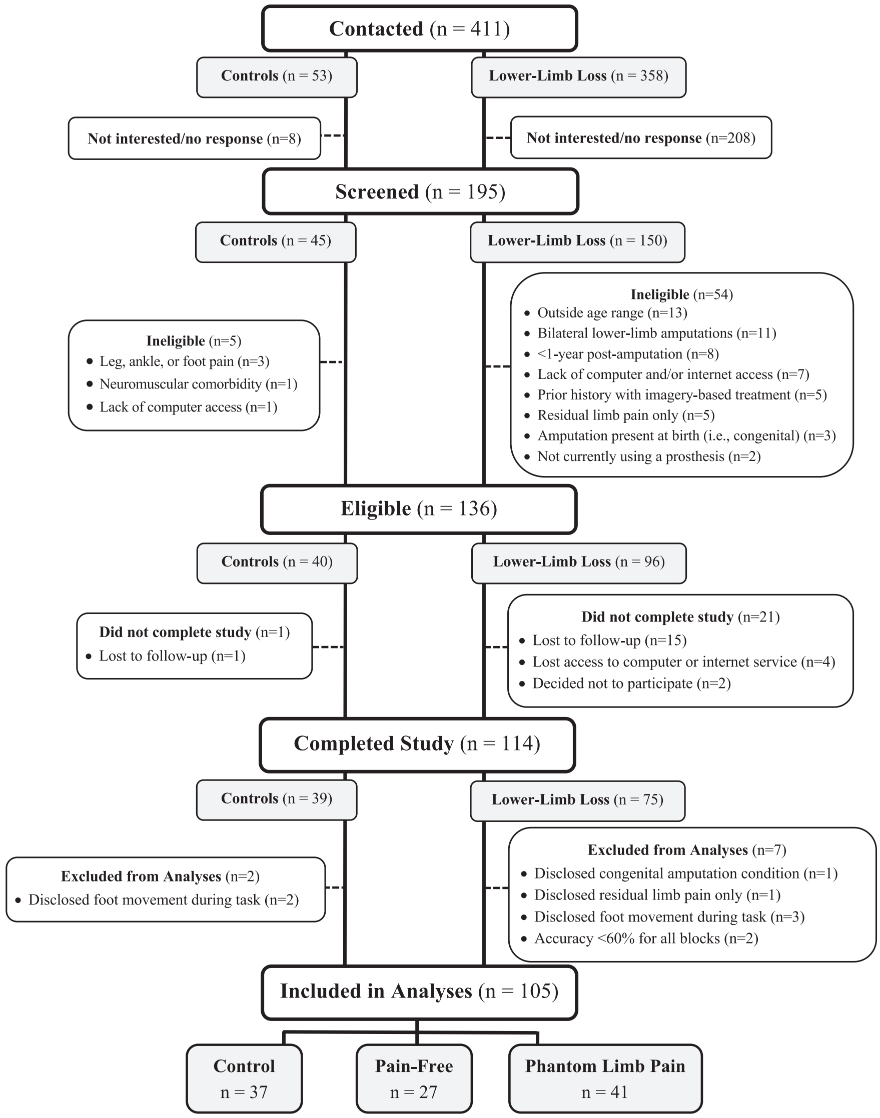

FIGURE 1 Participant inclusion flow diagram

1-to-8-point reductions). Four participants with pre-task PLP reported a worsening of pain during the task, with increases in pain ranging from 25% to 50% of their pre-task pain [equivalent to a 1-to-2-point change, which fails to surpass test-–retest measurement error previously reported for the numeric pain rating scale(Childs et al., 2005)]. Finally, among participants in the PLP group who denied pre-task PLP ($n = 16$), 13 (81%) did not experience any PLP during the task, while three reported an increase in PLP intensity ($n = 1$ with a 1-point increase; $n = 2$ with a 2-point increase).

3.2 | Response time

Among analysed participants, incorrect trials comprised 9.7% of all trials (i.e., 3882 out of 40,033) and were removed from response time analyses. Additionally, 3.0% of correct trials were removed due to response times falling below (36) or above (1036) established cut-points. Overall group means are presented in Table 3, and model results are presented in Table S1.

There were significant main effects of *group*, *block*, *view*, *angle* and *repetition* (Table S3 for detailed post hoc comparisons of main effects). Overall, adults with PLP and Pain-Free participants were slower to respond than Controls [mean difference = 248 and 357 ms respectively, $p = 0.004$–0.018], but differences between LLL groups were nonsignificant ($p = 0.394$). Response time was faster with each subsequent block (mean difference = 67–382 ms, $p < 0.001$) and for dorsal-view images as compared to plantar-view images [mean difference = 407 ms, $p < 0.001$). Response times slowed as image rotation increased (i.e., as compared to 0° and 300° orientations, response times were 188–564 ms slower at images rotated 60°, 120°, 180° and 240°, $p < 0.001$). Finally, participants were 66 ms faster upon viewing the second image repetition within each block ($p < 0.001$).

While *group* * *view* and *group* * *angle* interactions were each significant, the significant *group* * *view* * *angle*

TABLE 1 Participant characteristics

Variable	Control ($n = 37$)	Pain-free ($n = 27$)	Phantom limb pain ($n = 41$)	p
Demographics				
Sex, female[a]	16 (43%)	10 (37%)	19 (46%)	0.667
Age, years[b]	56 (43, 61)	60 (48, 65)	58 (48, 66)	0.363
Height, m[b]	1.75 (1.65, 1.83)	1.75 (1.65, 1.80)	1.73 (1.60, 1.79)	0.688
Weight, kg[c,d]	79.2 (14.8)	87.4 (23.0)	89.0 (22.0)	**0.026**
Race, Caucasian/White[a]	32 (86%)	25 (93%)	39 (95%)	0.384
Ethnicity, Non-Hispanic[a]	35 (95%)	26 (96%)	40 (98%)	0.791
Hand and foot dominance				
Hand dominance, right[a]	34 (92%)	20 (74%)	30 (73%)	0.190
Foot dominance, right[a]	29 (78%)	22 (81%)	32 (78%)	0.369

Abbreviation: LLL, lower-limb loss.

[a] Data presented as n (% of sample).

[b] Data presented as median (25th, 75th percentile).

[c] Data presented as mean (standard deviation).

[d] Self-reported weight (with prosthesis donned for adults with LLL).

Bolded p-value indicates statistically significant between-group difference.

interaction indicated between-group differences were dependent on both angle and view (Figure 2). Post hoc pairwise comparisons indicated the Control group was significantly faster than PLP and Pain-Free groups at all dorsal-view angles (mean differences = 177–349 ms; standard error [SE] = 79–143, $p < 0.001$–0.025); however, for plantar-view images, differences between Controls and LLL groups were dependent upon the angle of rotation. Specifically, while Controls were significantly faster than adults with PLP when identifying plantar-view images rotated 0°, 240° and 300° (mean differences = 211–280 ms; SE = 100–119, $p = 0.025$–0.035), differences between Controls and adults with PLP were smaller and nonsignificant for plantar-view images rotated 60°, 120° and 180° ($p = 0.065$–0.100). Furthermore, differences between Controls and Pain-Free adults were smaller and nonsignificant for plantar-view images rotated 120° ($p = 0.105$), but Controls were significantly faster than Pain-Free adults at all other plantar-view angles. No response time differences were observed between adults with PLP and Pain-Free peers, regardless of view or angle ($p > 0.070$).

Additionally, between-group differences were observed for rotation costs, that is, differences in response time between the easiest orientation (0°) and the most difficult orientations (180° in the dorsal view and 120° in the plantar view). Adults with PLP demonstrated a greater rotation cost than Pain-Free peers for both dorsal-view images (i.e., a 58% increase in response time vs. a 47% increase among Pain-Free peers) and plantar-view images (i.e., a 46% increase in response time vs. a 28% increase among Pain-Free peers). Controls demonstrated similar rotation costs to adults with PLP (i.e., 50% and 56% increases in dorsal and plantar views respectively).

The PLP group also showed the least exaggerated differences when comparing dorsal-view images to plantar-view images at 120° and 180°. At 120°, adults with PLP were, on average, 521 [SE = 42, 95% confidence interval (CI) = 438–604] ms slower when identifying plantar-view images, while Pain-Free adults and Controls were, on average, 590 (SE = 53, 95% CI = 485–695) and 624 (SE = 43, 95% CI = 539–709) ms slower respectively. At 180°, adults with PLP were, on average, 145 (SE = 33, 95% CI = 81–210) ms slower when identifying plantar-view images, while Pain-Free adults and Controls were, on average, 235 (SE = 42, 95% CI = 153–317) and 214 (SE = 30, 95% CI = 155–273) ms slower respectively. Conversely, the Pain-Free group demonstrated greater differences between plantar and dorsal views at 0°, 60° and 240° (mean difference = 439–726 ms) as compared to PLP and Control groups (mean difference = 273–625 ms).

In addition to the three-way interaction, there was a significant *group* * *block* interaction (Figure 3). While adults with PLP and Pain-Free participants performed similarly in all blocks ($p > 0.222$), adults with PLP improved the most as blocks progressed (mean difference = 493 ms, SE = 28, 95% CI = 438–548) versus Pain-Free peers (mean difference = 365 ms, SE = 29, 95% CI = 309–421) or Controls (mean difference = 297 ms, SE = 20, 95% CI = 259–336). Pairwise comparisons also evidenced significant response time differences between Controls and both adults with PLP and Pain-Free peers in all blocks ($p < 0.001$–0.046).

TABLE 2 Amputation-specific characteristics among adults with LLL ($n = 68$)

Variable	Pain-free ($n = 27$)	Phantom limb pain ($n = 41$)	p
Amputation-related details			
Amputated side, right[a]	14 (52%)	25 (61%)	0.457
Level[a]			
Transtibial	18 (67%)	21 (52%)	0.204
Transfemoral	6 (22%)	18 (44%)	
Knee disarticulation	2 (7%)	1 (2%)	
Hip disarticulation	0 (0%)	1 (2%)	
Rotationplasty	1 (4%)	0 (0%)	
Etiology[a]			
Trauma	9 (33%)	16 (39%)	0.669
Cancer	5 (18%)	9 (22%)	
Infection	4 (15%)	8 (19%)	
Diabetes	4 (15%)	4 (10%)	
Peripheral vascular disease	1 (4%)	2 (5%)	
Other/multiple reasons	4 (15%)	2 (5%)	
Time since amputation, years[b]	8 (3, 26)	8 (4, 20)	0.869
Prosthesis-related details			
Prosthesis experience, >5 Years[a]	14 (52%)	26 (63%)	0.343
Daily prosthesis wear time[a]			
<25% of waking hours (1–3 h)	0 (0%)	2 (5%)	0.179
25%–50% of waking hours (4–8 h)	0 (0%)	3 (7%)	
>50% of waking hours (>8 h)	3 (11%)	8 (20%)	
All waking hours (12–16 h)	24 (89%)	28 (68%)	
Non-painful phantom limb sensations			
Phantom limb sensations present[a]	20 (74%)	35 (85%)	0.201
Bothersomeness[a]	$n = 20$	$n = 35$	
Not bothered	17 (85%)	24 (69%)	0.363
Somewhat bothered	3 (15%)	10 (28%)	
Extremely bothered	0 (0%)	1 (3%)	
Residual limb pain			
Residual limb pain present[a]	–	26 (63%)	–
Average intensity in past 7 days, 0–10[b]	–	$n = 26$ 3 (2, 5)	–
Bothersomeness[a]		$n = 26$	
Not bothered	–	9 (34%)	–
Somewhat bothered	–	15 (58%)	–
Extremely bothered	–	2 (8%)	–
Phantom limb pain			
Average intensity in past 24 h, 0–10[b]	–	$n = 40$ 2 (1, 5)	–
Bothersomeness[a]		$n = 40$	

(Continues)

TABLE 2 (Continued)

Variable	Pain-free (*n* = 27)	Phantom limb pain (*n* = 41)	*p*
Not bothered	–	8 (20%)	–
Somewhat bothered	–	29 (73%)	
Extremely bothered	–	3 (7%)	
Frequency in the past week[a]		*n* = 40	
Never	–	4 (10%)	–
1–3 times per week	–	17 (43%)	
4–6 times per week	–	7 (18%)	
Once per day	–	3 (7%)	
Multiple times per day	–	8 (20%)	
Constant pain	–	1 (2%)	
Duration in the past week[a]		*n* = 39	
<1 minute	–	12 (31%)	–
Several minutes but <1 h	–	17 (44%)	
Several hours	–	6 (15%)	
Several days	–	4 (10%)	
BPI-SF pain interference domain, 0–10b	–	*n* = 40 1.29 (0.43, 3.39)	–

Abbreviations: BPI-SF, Brief Pain Inventory-Short Form; LLL, lower-limb loss.
[a]Data presented as *n* (% of sample).
[b]Data presented as median (25th, 75th percentile).

TABLE 3 Left–right discrimination task performance across groups

Group	Overall mean	SE	95% confidence interval	
			Lower bound	Upper bound
Response time (ms)				
Control (*n* = 37)	1363	69	1234	1506
Pain-free (*n* = 27)	1720	102	1531	1932
Phantom limb pain (*n* = 41)	1611	78	1465	1770
Accuracy (proportion of correct responses)				
Control (*n* = 37)	0.96	0.007	0.94	0.97
Pain-free (*n* = 27)	0.93	0.015	0.89	0.95
Phantom limb pain (*n* = 41)	0.93	0.014	0.90	0.95

Abbreviation: SE, standard error.

Finally, no significant main or interaction effects of *side* were observed. There was no significant main effect of *type*; however, there was a significant interaction effect of *group * type*, indicating adults with PLP were 35 ms (SE = 10, 95% CI = 15–54) faster when identifying prosthetic versus anatomical feet ($p = 0.001$).

3.3 | Accuracy

Similar to previous studies investigating left–right discrimination of feet (Coslett et al., 2010a; Curtze et al., 2010; Stone et al., 2019), task accuracy was high (proportion of correct responses: 0.94, see Table 3 for overall means and Table S2 for model results).

In the accuracy model, significant main effects mirrored those of the response time model. Differences between Controls and both adults with PLP and Pain-Free participants were similar (0.03, i.e., adults with LLL in both groups were 3% less accurate than Controls on average), but differences only reached significance between Controls and adults with PLP ($p = 0.038$ vs. $p = 0.070$ among Pain-Free participants). Accuracy was lower in block 1 compared to subsequent blocks (mean

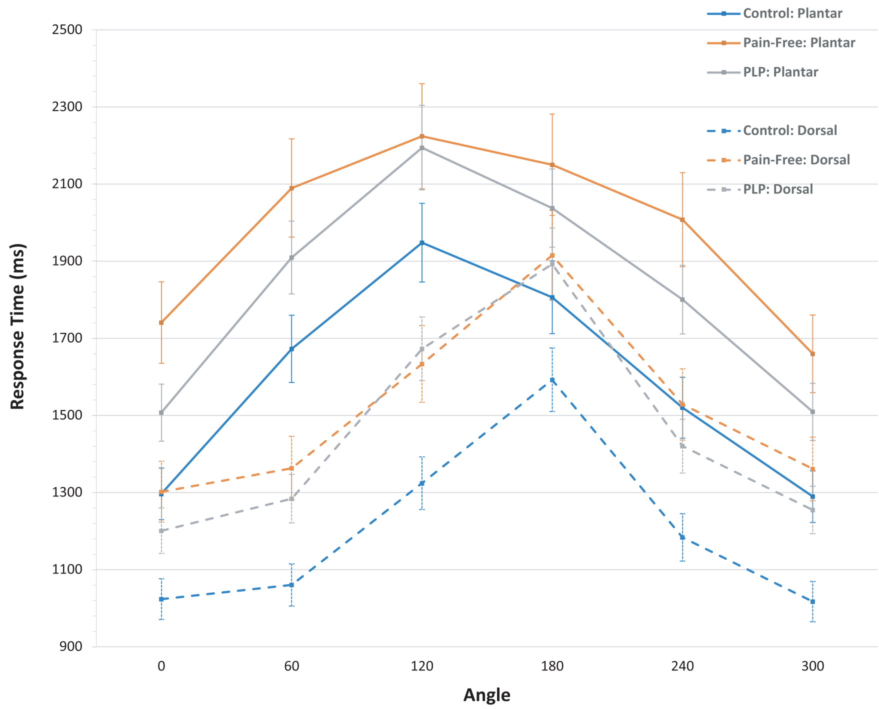

FIGURE 2 Mean response times for Control, Pain-Free, and phantom limb pain (PLP) groups are presented by angle and view. Controls were significantly faster than Pain-Free adults and adults with PLP at all angles in the dorsal view, but between-group differences in the plantar view were dependent upon the angle of rotation. Specifically, in the plantar view, adults with PLP were significantly slower than Controls at 0°, 240° and 300°, but differences were smaller and nonsignificant at 60°, 120° and 180°. Furthermore, Pain-Free adults were significantly slower than Controls when identifying plantar-view images at all angles except 120°. No differences were observed between Pain-Free and PLP groups at any angle or view

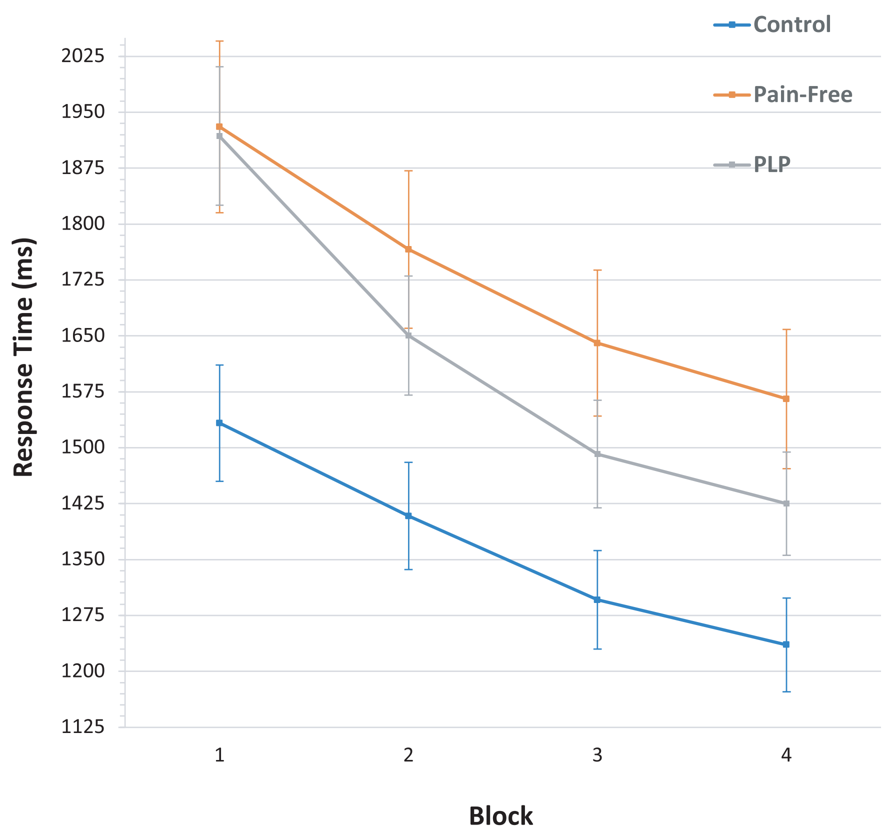

FIGURE 3 Mean response times for Control, Pain-Free and phantom limb pain (PLP) groups are presented by block (1–4). Both participants with PLP and Pain-Free peers were significantly slower than Controls in all four blocks. No significant differences were observed between adults in the PLP and Pain-Free groups during any block. Participants in the PLP group demonstrated larger improvements in response time between blocks 1 and 4 (493 ms) as compared to Controls and adults in the Pain-Free group (297 and 365 ms respectively)

difference = 0.02–0.03), at greater rotations away from anatomical position (mean difference = 0.03–0.14), for plantar-view images versus dorsal-view images (mean difference = 0.10) and for the first versus second repetition within each block (mean difference = 0.01, $p \leq 0.001$ for all pairwise comparisons; see Table S3 for detailed post hoc comparisons of main effects). There were no significant main or interaction effects of *side* or *type*.

There were significant *group* * *angle* and *group* * *view* interactions, which were superseded by a significant *group* * *angle* * *view* interaction (Figure 4). While between-group differences were larger at more difficult angles in the plantar view as opposed to the dorsal view, between-group differences were only statistically significant for images presented from the plantar view, speaking to the consistency of Control accuracy when images were presented from the dorsal view. Adults with PLP were significantly less accurate than Controls when identifying dorsal-view images at 0°, 60° and 120° (mean differences = 0.02–0.03, SE = 0.008–0.013, $p = 0.011$–0.027), while Pain-Free adults were less accurate than Controls at 0° of rotation in the dorsal view (mean difference = 0.03, SE = 0.008, $p = 0.006$). No significant differences were observed between adults with PLP and Pain-Free peers at any angle or view ($p > 0.252$).

Regardless of view, rotation cost (i.e., decrement in proportion of correct responses as compared to that of the 0° orientation) was greatest for adults with PLP. Specifically, for dorsal-view images rotated 180°, the proportion of correct responses decreased by 0.11 among adults with PLP as compared to 0.09 and 0.07 among Controls and Pain-Free peers respectively. For plantar-view images rotated 120°, the proportion of correct responses decreased by 0.32 among adults with PLP, as compared to 0.24 and 0.27 among Controls and Pain-Free peers respectively.

3.4 | Post hoc analyses

Finally, given the high frequency of adults with PLP reporting concurrent residual limb pain (i.e., 63%), the abovementioned GEE models were repeated among the PLP group only, with the presence or absence of residual limb pain (i.e., 0 = no, 1 = yes) entered as a main effect and in place of the 'group' variable in all interaction effects. In these subset analyses, the main effects of residual limb pain were nonsignificant (accuracy model: Wald $\chi^2 = 1.37$, $p = 0.243$; response time model: Wald $\chi^2 = 0.05$, $p = 0.818$) and interaction terms were nonsignificant between residual limb pain and image side, view, foot type, or angle (Wald $\chi^2 = 0.01$–4.78; $p > 0.189$), indicating residual limb pain, in isolation, did not appear to affect left–right discrimination performance.

4 | DISCUSSION

In this novel investigation of body representation among adults with PLP, we hypothesized adults with PLP would demonstrate poorer performance when identifying amputated-side images during a left–right discrimination task as compared to pain-free peers with and without LLL. Overall, adults with PLP demonstrated slower response times and reduced accuracy as compared to Controls with intact limbs, suggesting the body schema may be impaired with PLP. Notably, however, performance was

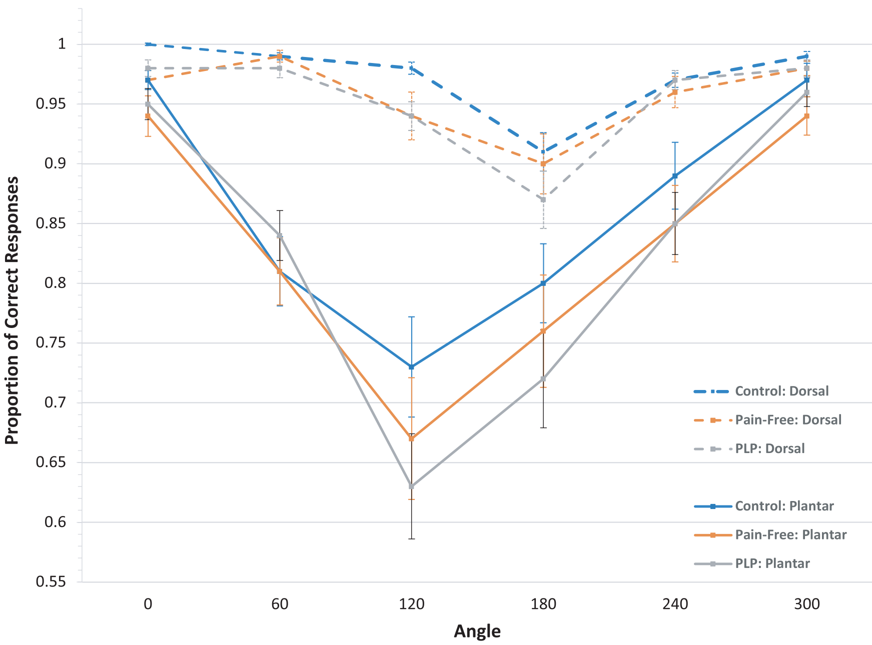

FIGURE 4 The proportion of correct responses among Control, Pain-Free and phantom limb pain (PLP) groups are presented by image angle and view. Between-group differences were only statistically significant for images presented from the dorsal view. Adults with PLP were significantly less accurate than Controls when identifying dorsal-view images at 0°, 60°, and 120°, while Pain-Free adults were less accurate than Controls at 0° of rotation in the dorsal view. No differences were observed at any angle or in any view between adults with PLP and Pain-Free peers

similar between PLP and Pain-Free groups, suggesting body schema impairments may be attributed to overarching, amputation-related somatosensory changes, rather than changes specific to PLP. Importantly, all groups appeared to utilize implicit motor imagery, as responses reflected biomechanical constraints of movement [i.e., poorer performance for images requiring greater limb rotation (Parsons, 1994)].

In previous studies (Coslett et al., 2010a, 2010b; Schwoebel et al., 2001), response times were substantially, and selectively, increased at larger degrees of hand and/or foot rotation, reflecting greater rotation costs among adults with chronic pain. This interaction between pain and rotation is attributed to the subliminal anticipation of pain when simulating awkward positions with the affected limb (Moseley, 2004b) and may capture specific deficits associated with painful movement (Coslett et al., 2010a, 2010b). Adults with PLP, however, did not demonstrate greater rotation costs as compared to Controls, and the *group * angle* interaction was nonsignificant when comparing only participants with PLP to Controls in a post hoc GEE ($p = 0.114$). Performance trends among adults with PLP were, instead, like those of 'pain Controls' (i.e., patients with chronic, non-lower-extremity pain) in a previous study of foot identification Coslett et al., 2010a). Thus, in this study, poorer performance may be attributed to general, pain-related changes in body representation, rather than changes specific to the region representing the phantom limb. Despite a lack of distinct rotation costs in comparison to controls, adults with PLP demonstrated augmented rotation costs as compared to pain-free peers and were significantly less accurate when identifying dorsal-view images, indicating a potential exacerbation of amputation-related body schema impairments in the presence of PLP.

Our hypothesis regarding poorer performance for amputated-side images was not supported among adults with PLP, further suggesting performance deficits observed may not be PLP-specific. Patients with chronic unilateral pain typically demonstrate poorer performance when identifying the affected side; however, the effect of side appears less robust than other image factors [e.g. angle, view (Breckenridge et al., 2019)]. Our findings parallel those reported by Fiorio et al. (2006) among adults with chronic focal hand dystonia and Kohler et al. (2019) among adults with complex regional pain syndrome (CRPS), where left–right discrimination was similar bilaterally, even among patients with chronic unilateral pain conditions.

Remarkably, adults with PLP were significantly slower when identifying human, as opposed to prosthetic, feet. While findings may suggest uniquely impaired foot identification among adults with PLP, findings were not confirmed in accuracy models; therefore, this effect may be attributed to other, unmeasured factors (e.g. differences in image features) and may require further investigation in future studies.

While prior work has tested only dorsal-view images to preferentially facilitate an egocentric (i.e., first-person) approach to mental rotation (Coslett et al., 2010a), we included both dorsal- and plantar-view images to evaluate view-specific performance differences. Between-group differences in performance were robust for dorsal-view images, confirming implicit motor imagery appears to be impeded by LLL. Inconsistences for plantar-view images, however, indicate adults with and without LLL may have been equally challenged by images presented from an allocentric perspective (i.e., third-person), when alternative strategies may have been used to determine laterality [e.g. rotating the object on the screen (Zacks et al., 2002)].

Findings augment conflicting evidence of body schema impairments post-LLL. For example our findings support those of Palermo et al. (2018), where 14 male participants with LLL were less accurate than 11 controls during a left–right discrimination task, but contrast those of Stone et al. (2019) and Curtze et al. (2010) who found no differences in left–right discrimination among adults with LLL ($n = 19$ and $n = 18$ respectively) when compared to controls ($n = 33$ and $n = 18$ respectively). While we found significant differences between our moderately sized sample of controls ($n = 37$) and adults with LLL ($n = 68$), the left–right discrimination task may not be sensitive enough to detect body schema impairments among smaller, heterogeneous samples post-LLL. In support of this interpretation, Boccia et al. (2020) found functional reorganization within brain areas associated with body representation (e.g. anterior insula, supplementary motor area) among adults with LLL ($n = 9$) during a left–right discrimination task, despite their performance being similar to controls ($n = 11$).

Overall response time differences between controls and adults with PLP (i.e., 248 ms) were comparable to prior studies of temporomandibular joint pain [i.e., ~173 ms (Uritani et al., 2018)] and focal hand dystonia [i.e., ~300 ms (Fiorio et al., 2006)] but were less robust than those among adults with upper-limb loss (Nico et al., 2004; Reinersmann et al., 2010) and other forms of unilateral, chronic pain (Breckenridge et al., 2019). For example, adults with CRPS are up to 2000 ms slower than controls when identifying images of their affected limb (Moseley, 2004b). Discrepancies in response time deficits may be, in part, explained by consistent prosthesis use among adults in our sample. Maladaptive disuse of a painful body part may contribute to body schema deficits (Date et al., 2019); however, 88% of our participants with PLP reported using a prosthesis >8 h/day. Frequent prosthesis use is associated with reduced phantom limb sensations (Giummarra et al., 2010) and may diminish amputation-related changes in the body schema (Guo et al., 2017), potentially minimizing differences between adults with PLP and controls. Furthermore, discrepancies may be attributed to dissimilarities in pain severity and characteristics across samples. For example, in prior studies, individuals included in 'pain' groups largely reported moderate-to-severe pain [i.e., ~5-to-7 out of 10 (Breckenridge et al., 2019)], whereas our participants reported mild, yet bothersome, pain.

Unexpectedly, adults with PLP demonstrated the greatest response time improvements between Blocks 1 and 4. Thus, practice appears to ameliorate deficits to a greater degree among adults with PLP, potentially highlighting an important degree of body schema plasticity specific to PLP. Furthermore, while task performance was not selectively impaired in PLP, 80% of adults with pre-task PLP reported reduced PLP intensity during the task. Findings suggest implicit motor imagery practice may reduce PLP intensity, supporting recent efforts to utilize imagery-based treatment [e.g. graded motor imagery (Moseley, 2006), virtual reality (Ambron et al., 2018), mirror therapy (Chan et al., 2007)] as part of comprehensive, PLP treatment. Clinically, left–right discrimination tasks could be used to assess within-session improvements in both performance and PLP severity; patients with positive pain responses may especially benefit from further imagery-based training. Although 17% of participants with PLP reported increased PLP intensity during the task, increases did not exceed measurement error, indicating the benefits of utilizing imagery-based treatment for PLP may outweigh potential costs.

While imagery-based treatment shows promise in chronic pain management (Bowering et al., 2013; Moseley, 2006), our inclusion diagram indicates the rarity of imagery-based training in post-amputation care. Of the 150 adults with LLL screened for this study, only 5 (3%) were excluded secondary to prior participation in imagery-based treatment programmes, including mirror therapy. In clinical settings, hesitance to initiate imagery-based treatment may be attributed to limited evidence informing its use, and/or a lack of established protocols for training progression. Imagery-based treatment offers a promising alternative to invasive interventions (e.g. surgery) and can be inexpensively integrated across the post-amputation care continuum (i.e., acute, subacute, outpatient); therefore, future studies may consider investigating best practice techniques for administering imagery-based treatment post-LLL.

4.1 | Study limitations

While this study provides novel information regarding potential, underlying impairments in the body schema among adults with PLP post-LLL, limitations include its generalizability, as findings may only be generalized to adults with predominantly transtibial-level, traumatic amputations who experience relatively mild PLP. Furthermore, as this study was completed remotely, participants may not have completed the task in accordance with instructions (e.g. no limb movement, limbs covered to prevent visualization, prosthesis doffed). Participants reporting limb movement, however, were removed from analyses, and response time and accuracy rates reflect biomechanical constraints on movement, suggesting movement and visual input did not significantly affect study results. Findings validate the use of an online assessment of the body schema for future research and other clinical interventions (e.g. tele-rehabilitation programmes) when in-person assessments are not feasible. Additionally, the presence of non-painful phantom limb sensations appears to contribute to body representation impairments (Lyu et al., 2016); however, the impact of non-painful phantom limb sensations was not assessed in this study, given the small sample ($n = 7$) of adults without non-painful phantom limb sensations in the Pain-Free group. Similarly, while pain medication was not captured in this study, pain medication use may impact left–right discrimination (Pelletier et al., 2018). Future studies may investigate the unique impacts of non-painful phantom limb sensations and pain medication on body representation with PLP.

5 | CONCLUSION

Following LLL, adults with PLP appear to demonstrate impaired body representation, as evidenced by slower and less accurate left–right discrimination performance when compared to age-matched controls with intact limbs. Overall, however, performance appears similar between adults with PLP and pain-free peers with LLL, suggesting observed impairments may be associated with somatosensory reorganization associated with LLL, rather than PLP alone. Patterns of performance among adults with PLP suggest left–right discrimination improves with practice, and improvements in performance may be coupled with reductions in PLP intensity. Even in the absence of specific, PLP-related impairments in left–right discrimination, 80% of adults with pre-task PLP demonstrated improvements in PLP intensity during the task, which is compelling evidence for further, longitudinal investigation into the efficacy of imagery-based treatment interventions following LLL as a conservative management approach.

ACKNOWLEDGMENTS

The authors thank Drs. Carolin Curtze, PhD and Kayla Stone, PhD for supplying the stimuli for the foot identification task, as well as Samantha Stauffer, CPO, MSOP and the teams at Independence Prosthetics-Orthotics, Inc. and Real Life Prosthetics, LLC for their assistance with participant recruitment.

CONFLICT OF INTEREST

The authors have no conflicts of interest to report.

AUTHORS' CONTRIBUTIONS

Emma H. Beisheim-Ryan: conception and design; acquisition, analysis, and interpretation of data; drafting and revising of manuscript; final approval of manuscript. Ryan T. Pohlig: analysis and interpretation of data, revising of manuscript, final approval of manuscript. Jared Medina: interpretation of data, revising of manuscript, final approval of manuscript. Gregory E. Hicks: interpretation of data, revising of manuscript, final approval of manuscript. Jaclyn M. Sions: design, interpretation of data, revising of manuscript, final approval of manuscript.

REFERENCES

Ambron, E., Miller, A., Kuchenbecker, K. J., Buxbaum, L. J., & Coslett, H. B. (2018). Immersive low-cost virtual reality treatment for phantom limb pain: Evidence from two cases. *Frontiers in Neurology*, *9*, 67. https://doi.org/10.3389/fneur.2018.00067

Anwyl-Irvine, A. L., Massonnié, J., Flitton, A., Kirkham, N., & Evershed, J. K. (2020). Gorilla in our midst: An online behavioral experiment builder. *Behavior Research Methods*, *52*, 388–407. https://doi.org/10.3758/s13428-019-01237-x

Boccia, M., Di Vita, A., Palermo, L., Nemmi, F., Traballesi, M., Brunelli, S., De Giorgi, R., Galati, G., & Guariglia, C. (2020). Neural modifications in lower limb amputation: An fMRI study on action and non-action oriented body representations. *Brain Imaging and Behavior*, *14*, 416–425. https://doi.org/10.1007/s11682-019-00142-3

Bowering, K. J., O'Connell, N. E., Tabor, A., Catley, M. J., Leake, H. B., Moseley, G. L., & Stanton, T. R. (2013). The effects of graded motor imagery and its components on chronic pain: A systematic review and meta-analysis. *The Journal of Pain*, *14*, 3–13. https://doi.org/10.1016/j.jpain.2012.09.007

Breckenridge, J. D., Ginn, K. A., Wallwork, S. B., & McAuley, J. H. (2019). Do people with chronic musculoskeletal pain have impaired motor imagery? A meta-analytical systematic review of the left/right judgment task. *The Journal of Pain*, *20*, 119–132. https://doi.org/10.1016/j.jpain.2018.07.004

Chan, B. L., Witt, R., Charrow, A. P., Magee, A., Howard, R., Pasquina, P. F., Heilman, K. M., & Tsao, J. W. (2007). Mirror therapy for phantom limb pain. *New England Journal of Medicine*, *357*, 2206–2207. https://doi.org/10.1056/NEJMc071927

Chiarotto, A., Maxwell, L. J., Ostelo, R. W., Boers, M., Tugwell, P., & Terwee, C. B. (2019). Measurement properties of Visual Analogue Scale, Numeric Rating Scale, and Pain Severity Subscale of the Brief Pain Inventory in patients with low back pain: A systematic review. *The Journal of Pain*, *20*, 245–263. https://doi.org/10.1016/j.jpain.2018.07.009

Childs, J. D., Piva, S. R., & Fritz, J. M. (2005). Responsiveness of the Numeric Pain Rating Scale in patients with low back pain. *Spine*, *30*(11), 1331–1334. https://doi.org/10.1097/01.brs.0000164099.92112.29

Coslett, H. B., Medina, J., Kliot, D., & Burkey, A. R. (2010a). Mental motor imagery and chronic pain: The foot laterality task. *Journal of the International Neuropsychological Society*, *16*, 603–612. https://doi.org/10.1017/S1355617710000299

Coslett, H. B., Medina, J., Kliot, D., & Burkey, A. R. (2010b). Mental motor imagery indexes pain: The hand laterality task. *European Journal of Pain*, *14*, 1007–1013. https://doi.org/10.1016/j.ejpain.2010.04.001

Curtze, C., Otten, B., & Postema, K. (2010). Effects of lower limb amputation on the mental rotation of feet. *Experimental Brain Research*, *201*, 527–534. https://doi.org/10.1007/s00221-009-2067-z

Date, S., Kurumadani, H., Yoshimura, M., Fukae, A., Onishi, K., Hayashi, J., Shinomiya, R., & Sunagawa, T. (2019). Long-term disuse of the hand affects motor imagery ability in patients with complete brachial plexus palsy. *NeuroReport*, *30*, 452–456. https://doi.org/10.1097/WNR.0000000000001229

Ehde, D. M., Nitsch, K. P., & Smiley, J. P. (2015). Measurement characteristics and clinical utility of the Brief Pain Inventory-Short Form for individuals with multiple sclerosis. *Rehabilitation Psychology*, *60*, 365–366. https://doi.org/10.1037/rep0000065

Ephraim, P. L., Wegener, S. T., MacKenzie, E. J., Dillingham, T. R., & Pezzin, L. E. (2005). Phantom pain, residual limb pain, and back pain in amputees: Results of a national survey. *Archives of Physical Medicine and Rehabilitation*, *86*, 1910–1919. https://doi.org/10.1016/j.apmr.2005.03.031

Fiorio, M., Tinazzi, M., & Aglioti, S. M. (2006). Selective impairment of hand mental rotation in patients with focal hand dystonia. *Brain*, *129*, 47–54. https://doi.org/10.1093/brain/awh630

Giummarra, M. J., Georgiou-Karistianis, N., Nicholls, M. E., Gibson, S. J., Chou, M., & Bradshaw, J. L. (2010). Corporeal awareness and proprioceptive sense of the phantom. *British Journal of Psychology*, *101*, 791–808. https://doi.org/10.1348/000712610X492558

Giummarra, M. J., Gibson, S. J., Georgiou-Karistianis, N., & Bradshaw, J. L. (2007). Central mechanisms in phantom limb perception: The past, present and future. *Brain Research Reviews*, *54*, 219–232. https://doi.org/10.1016/j.brainresrev.2007.01.009

Goller, A. I. (2012). *Perceptual abnormalities in amputees: Phantom pain, mirror-touch synaesthesia, and referred tactile sensations* (Doctoral thesis). Psychology, University of Sussex.

Griswold, M. E., Swihart, B. J., Caffo, B. S., & Zeger, S. L. (2013). Practical marginalized multilevel models. *Stat*, *2*. https://doi.org/10.1002/sta4.22

Guo, X., Lin, Z., Lyu, Y., Bekrater-Bodmann, R., Flor, H., & Tong, S. (2017). The effect of prosthesis use on hand mental rotation after unilateral upper-limb amputation. *IEEE Transactions on Neural Systems and Rehabilitation Engineering*, *25*, 2046–2053. https://doi.org/10.1109/TNSRE.2017.2702117

Hand, B. N., Velozo, C. A., & Krause, J. S. (2018). Measuring the interference of pain on daily life in persons with spinal cord injury: A Rasch-validated subset of items from the Brief Pain Inventory interference scale. *Australian Occupational Therapy Journal*, *65*, 405–411. https://doi.org/10.1111/1440-1630.12493

Hanley, M. A., Ehde, D. M., Campbell, K. M., Osborn, B., & Smith, D. G. (2006). Self-reported treatments used for lower-limb phantom pain: Descriptive findings. *Archives of Physical Medicine and Rehabilitation*, *87*, 270–277. https://doi.org/10.1016/j.apmr.2005.04.025

Harding, J. L., Andes, L. J., Rolka, D. B., Imperatore, G., Gregg, E. W., Li, Y., & Albright, A. (2020). National and state-level trends in nontraumatic lower-extremity amputation among U.S. Medicare beneficiaries with diabetes, 2000–2017. *Diabetes Care*, *43*, 2453–2459. https://doi.org/10.2337/dc20-0586

Harris, P. A., Taylor, R., Minor, B. L., Elliott, V., Fernandez, M., O'Neal, L., McLeod, L., Delacqua, G., Delacqua, F., Kirby, J., Duda, S. N. & REDCap Consortium. (2019). The REDCap consortium: Building an international community of software platform partners. *Journal of Biomedical Informatics*, *95*, 103208. https://doi.org/10.1016/j.jbi.2019.103208

Harris, P. A., Taylor, R., Thielke, R., Payne, J., Gonzalez, N., & Conde, J. G. (2009). Research electronic data capture (REDCap)–a metadata-driven methodology and workflow process for providing translational research informatics support. *Journal of Biomedical Informatics*, *42*, 377–381. https://doi.org/10.1016/j.jbi.2008.08.010

Heagerty, P. J., & Zeger, S. L. (2000). Marginalized multilevel models and likelihood inference. *Statistical Science*, *15*(1), 1–26.

Hubbard, A. E., Ahern, J., Fleischer, N. L., Van der Laan, M., Lippman, S. A., Jewell, N., Bruckner, T., & Satariano, W. A. (2010). To GEE or not to GEE: Comparing population average and mixed models for estimating the associations between neighborhood risk factors and health. *Epidemiology*, *21*, 467–474. https://doi.org/10.1097/EDE.0b013e3181caeb90

Kohler, M., Strauss, S., Horn, U., Langner, I., Usichenko, T., Neumann, N., & Lotze, M. (2019). Differences in neuronal representation of mental rotation in patients with complex regional pain syndrome and healthy controls. *J Pain*, *20*, 898–907. https://doi.org/10.1016/j.jpain.2019.01.330

Lee, D., Choi, S. H., Noh, E., Lee, W. J., Jang, J. H., Moon, J. Y., & Kang, D. H. (2021). Impaired performance in mental rotation of hands and feet and its association with social cognition in patients with complex regional pain syndrome. *Pain Medicine*, *6*, 1411–1419. https://doi.org/10.1093/pm/pnaa482

Liang, K., & Zeger, S. (1986). Longitudinal data analysis using generalized linear models. *Biometrika*, *73*(1), 13–22. https://doi.org/10.1093/biomet/73.1.13

Limakatso, K., Bedwell, G. J., Madden, V. J., & Parker, R. (2020). The prevalence and risk factors for phantom limb pain in people with amputations: A systematic review and meta-analysis. *PLoS One*, *15*(10), e0240431. https://doi.org/10.1371/journal.pone.0240431

Louw, A., Puentedura, E. J., Reese, D., Parker, P., Miller, T., & Mintken, P. E. (2017). Immediate effects of mirror therapy in patients with shoulder pain and decreased range of motion. *Archives of Physical Medicine and Rehabilitation*, *98*, 1941–1947. https://doi.org/10.1016/j.apmr.2017.03.031

Louw, A., Schmidt, S. G., Louw, C., & Puentedura, E. J. (2015). Moving without moving: Immediate management following lumbar spine surgery using a graded motor imagery approach: A case report. *Physiotherapy Theory and Practice*, *31*, 509–517. https://doi.org/10.3109/09593985.2015.1060656

Lustenhouwer, R., Cameron, I. G. M., van Alfen, N., Oorsprong, T. D., Toni, I., van Engelen, B. G. M., Groothuis, J. T., & Helmich, R. C. (2020). Altered sensorimotor representations after recovery from peripheral nerve damage in neuralgic amyotrophy. *Cortex*, *127*, 180–190. https://doi.org/10.1016/j.cortex.2020.02.011

Lyu, Y., Guo, X., Bekrater-Bodmann, R., Flor, H., & Tong, S. (2016). Phantom limb perception interferes with motor imagery after unilateral upper-limb amputation. *Scientific Reports*, *6*, 21100. https://doi.org/10.1038/srep21100

Medina, J., & Coslett, H. B. (2010). From maps to form to space: Touch and the body schema. *Neuropsychologia*, *48*, 645–654. https://doi.org/10.1016/j.neuropsychologia.2009.08.017

Mendez-Rebolledo, G., Gatica-Rojas, V., Torres-Cueco, R., Albornoz-Verdugo, M., & Guzman-Munoz, E. (2017). Update on the effects of graded motor imagery and mirror therapy on complex regional pain syndrome type 1: A systematic review. *Journal of Back and Musculoskeletal Rehabilitation*, *30*, 441–449. https://doi.org/10.3233/BMR-150500

Mendoza, T., Mayne, T., Rublee, D., & Cleeland, C. (2006). Reliability and validity of a modified Brief Pain Inventory short form in patients with osteoarthritis. *European Journal of Pain*, *10*, 353–361. https://doi.org/10.1016/j.ejpain.2005.06.002

Moseley, G. L. (2004a). Graded motor imagery is effective for long-standing complex regional pain syndrome: A randomised controlled trial. *Pain*, *108*, 192–198. https://doi.org/10.1016/j.pain.2004.01.006

Moseley, G. L. (2004b). Why do people with complex regional pain syndrome take longer to recognize their affected hand? *Neurology*, *62*, 2182–2186. https://doi.org/10.1212/01.WNL.0000130156.05828.43

Moseley, G. L. (2006). Graded motor imagery for pathologic pain: A randomized controlled trial. *Neurology*, *67*, 2129–2134. https://doi.org/10.1212/01.wnl.0000249112.56935.32

Moseley, G. L., Gallace, A., & Spence, C. (2012). Bodily illusions in health and disease: Physiological and clinical perspectives and the concept of a cortical 'body matrix'. *Neuroscience and Biobehavioral Reviews*, *36*, 34–46. https://doi.org/10.1016/j.neubiorev.2011.03.013

Nico, D., Daprati, E., Rigal, F., Parsons, L., & Sirigu, A. (2004). Left and right hand recognition in upper limb amputees. *Brain*, *127*, 120–132. https://doi.org/10.1093/brain/awh006

Owings, M. F., & Kozak, L. J. (1998). Ambulatory and inpatient procedures in the United States, 1996. *Vital Health Statistics*, *13*, 1–119.

Palermo, L., Di Vita, A., Boccia, M., Nemmi, F., Brunelli, S., Traballesi, M., De Giorgi, R., Galati, G., & Guariglia, C. (2018). Action and non-action oriented body representations: Insight from behavioural and grey matter modifications in individuals with lower limb amputation. *BioMed Research International*, *2018*, 1529730. https://doi.org/10.1155/2018/1529730

Parsons, L. M. (1987). Imagined spatial transformations of one's hands and feet. *Cognitive Psychology*, *19*, 178–241. https://doi.org/10.1016/0010-0285(87)90011-9

Parsons, L. M. (1994). Temporal and kinematic properties of motor behavior reflected in mentally simulated action. *Journal of Experimental Psychology: Human Perception and Performance*, *20*, 709–730. https://doi.org/10.1037/0096-1523.20.4.709

Parsons, L. M., & Shimojo, S. (1987). Perceived spatial organization of cutaneous patterns on surfaces of the human body in various positions. *Journal of Experimental Psychology: Human Perception and Performance*, *13*, 488–504. https://doi.org/10.1037/0096-1523.13.3.488

Pelletier, R., Bourbonnais, D., Higgins, J., Mireault, M., Danino, M. A., & Harris, P. G. (2018). Left right judgement task and sensory, motor, and cognitive assessment in participants with wrist/hand pain. *Rehabilitation Research and Practice*, *2018*, 1530245. https://doi.org/10.1155/2018/1530245

Penfield, W., & Boldrey, E. (1937). Somatic motor and sensory representation in the cerebral cortex of man as studied by electrical stimulation. *Brain*, *60*, 389–443. https://doi.org/10.1093/brain/60.4.389

Pirowska, A., Wloch, T., Nowobilski, R., Plaszewski, M., Hocini, A., & Ménager, D. (2014). Phantom phenomena and body scheme after limb amputation: A literature review. *Neurologia i Neurochirurgia Polska*, *48*, 52–59. https://doi.org/10.1016/j.pjnns.2013.03.002

Pitron, V., Alsmith, A., & de Vignemont, F. (2018). How do the body schema and the body image interact? *Consciousness and Cognition*, *65*, 352–358. https://doi.org/10.1016/j.concog.2018.08.007

Poquet, N., & Lin, C. (2016). The Brief Pain Inventory (BPI). *Journal of Physiotherapy*, *62*, 52. https://doi.org/10.1016/j.jphys.2015.07.001

Ramachandran, V. S., Brang, D., & McGeoch, P. D. (2010). Dynamic reorganization of referred sensations by movements of phantom limbs. *NeuroReport*, *21*, 727–730. https://doi.org/10.1097/WNR.0b013e32833be9ab

Ramachandran, V., Chunharas, C., Marcus, Z., Furnish, T., & Lin, A. (2018). Relief from intractable phantom pain by combining psilocybin and mirror visual-feedback (MVF). *Neurocase*, *24*, 105–110. https://doi.org/10.1080/13554794.2018.1468469

Reinersmann, A., Haarmeyer, G. S., Blankenburg, M., Frettloh, J., Krumova, E. K., Ocklenburg, S., & Maier, C. (2010). Left is where the L is right. Significantly delayed reaction time in limb laterality recognition in both CRPS and phantom limb pain patients. *Neuroscience Letters*, *486*, 240–245. https://doi.org/10.1016/j.neulet.2010.09.062

Saimpont, A., Pozzo, T., & Papaxanthis, C. (2009). Aging affects the mental rotation of left and right hands. *PLoS One*, *4*, e6714. https://doi.org/10.1371/journal.pone.0006714

Schwoebel, J., Friedman, R., Duda, N., & Coslett, H. B. (2001). Pain and the body schema: Evidence for peripheral effects on mental representations of movement. *Brain*, *124*, 2098–2104. https://doi.org/10.1093/brain/124.10.2098

Steenwinkel, F. A., Ter Meulen, B. C., & Ramachandran, V. S. (2019). Teaching video neuroimages: Mirror therapy against phantom pain. *Neurology*, *92*, e1002.

Stone, K. D., Dijkerman, H. C., Bekrater-Bodmann, R., & Keizer, A. (2019). Mental rotation of feet in individuals with Body Integrity Identity Disorder, lower-limb amputees, and normally-limbed controls. *PLoS One*, *14*, e0221105. https://doi.org/10.1371/journal.pone.0221105

Uritani, D., Nishida, T., Sakaguchi, N., Kawakami, T., Jones, L. E., & Kirita, T. (2018). Difference in response to a motor imagery task: A comparison between individuals with and without painful temporomandibular disorders. *Pain Research and Management*, *2018*, 6810412. https://doi.org/10.1155/2018/6810412

Zacks, J. M., Mires, J., Tversky, B., & Hazeltine, E. (2002). Mental spatial transformations of objects and perspective. *Spatial Cognition & Computation*, *2*, 315–332.

SUPPORTING INFORMATION

Additional supporting information may be found in the online version of the article at the publisher's website.

How to cite this article: Beisheim-Ryan, E. H., Pohlig, R. T., Medina, J., Hicks, G. E., & Sions, J. M. (2022). Body representation among adults with phantom limb pain: Results from a foot identification task. *European Journal of Pain*, 26, 255–269. https://doi.org/10.1002/ejp.1860