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Mirror therapy for phantom limb pain in moderate intellectual disability. A case report

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Abstract

Background: Phantom limb pain (PLP) is a common problem after limb amputation. There is mounting evidence supporting the use of mirror therapy (MT) in the treatment of individuals with PLP. However, there is no research studying the effects of MT on PLP in individuals with intellectual developmental disorders (IDD). The aim of this study was to increase our understanding of MT when used with adults with IDD and PLP through a case study approach.

Methods: Here, we describe the use of MT with a 53-year-old female with moderate IDD and PLP, related to her left leg being amputated after ulcer complications. The study followed an A-B-A-B design (baseline—treatment—withdrawal of treatment—re-introduction of treatment), lasting 2 years, which included a long-term follow-up.

Results: The data showed that the PLP sensation decreased after the MT treatment, with a raw change of 3.92 points and a 48% decrease in mean pain intensity ratings from pre- to post-treatment.

Conclusions: This is a unique case-report on the use of MT with an individual with IDD suffering from PLP. The findings show that MT helped to significantly reduce the intensity of the PLP in this patient.

Significance: This is a case-report that illustrates how mirror therapy can be applied to people with intellectual developmental disorders and phantom limb pain. The results showed that phantom limb pain decreased after the mirror therapy, with a raw change of 3,92 points and a percent change of 48%.

1 | INTRODUCTION

Phantom limb pain (PLP) is characterized by the persistent perception of painful phantom sensations that follow the loss of a limb (MeSH Browser, 2017; Ramachandran & Hirstein, 1998; Weinstein, 1998). The prevalence of PLP in individuals with amputations is uncertain (Limakatso et al., 2019). However, the incidence of PLP has been established to be 80% (Dijkstra et al., 2002; Richardson et al., 2006).

The pathophysiology of PLP is multifactorial, with peripheral and central mechanisms thought to be involved (Oscar Coppes & Sang, 2018). Nevertheless, mechanisms underlying the development of PLP are poorly understood (Jutzeler et al., 2015), and different theories involving the peripheral nerves, the spinal cord and the cortex have been suggested as processes leading to this phenomenon (Pirowska et al., 2014). Research has identified some variables as tentative risk factors for PLP (e.g. being female, having an upper limb amputation, presence of preamputation pain and time elapsed since amputation [Bosmans et al., 2010; Subedi & Grossberg, 2011]). However, these findings are under debate, as other studies showed that the occurrence of PLP is not associated with gender, age and the level or side of amputation (Nikolajsen & Jensen, 2001).

Several invasive (e.g. surgery, invasive neuromodulation) and non-invasive therapeutic approaches (e.g. pharmacological interventions, physical therapy) have been used to help cope with and adjust to PLP (Flor, 2008; Giummarra & Moseley, 2011; Knotkova et al., 2012; Richardson & Kulkarni, 2017; Tian, 2014).

Mirror therapy (MT) is a type of physical therapy that consists of a mirror box that provides a reflection of the intact limb, creating the illusion of observing the non-existent limb. The box allows the patient to experience vivid sensations of movement in the phantom limb when making mirror symmetric movements (Ramachandran & Rodgers-Ramachandran, 1996). There is mounting evidence supporting the use of MT in the treatment of individuals with PLP, as it helps reduce the duration and intensity of the pain episodes (Barbin et al., 2016; Campo-Prieto & Rodríguez-Fuentes, 2018; Chan et al., 2007; Deconinck et al., 2015; Griffin et al., 2017; Herrador Colmenero et al., 2018). Overall, studies about MT suggest that the ability to produce phantom limb movement or gain a sense of control is perhaps the most important factor for PLP alleviation, more than protocol, intensity, frequency or subject-dependent variables (Timms & Carus, 2015).

However, there is no study on the effects of MT on PLP in individuals with IDD (Lonchampt et al., 2020). This population has chronic health conditions like the general population, but these individuals are more likely to experience health disparities (Folch et al., 2019; Hatton & Emerson, 2015; Krahn & Fox, 2014; McMorris et al., 2015) and poorer quality health care (Krahn et al., 2006; Perry et al., 2014; Williamson et al., 2017). Furthermore, they have been assumed to be insensitive or indifferent to pain and thus excluded from pain research (De Knegt & Scherder, 2011). However, studies have demonstrated that both adults (Walsh et al., 2011) and young people (Breau et al., 2003; Rattaz et al., 2013) with IDD experience chronic pain which significantly affects their quality of life (Oberlander & Symons, 2006). Despite this, chronic pain is still under-recognized and undertreated in individuals with IDD (Amor-Salamanca & Menchon, 2017; Findlay et al., 2014; McGuire et al., 2010; Symons et al., 2008), placing them in a situation of great vulnerability and higher risk for experiencing severe pain without adequate management (Baldridge & Andrasik, 2010).

Accordingly, even though PLP in people with IDD was first described 60 years ago (Simmel, 1959), there has been little interest in this matter. Given their increased risk for chronic pain (Symons et al., 2008), it is of fundamental importance to develop new strategies, or study available ones, to address the needs on pain management of this population.

Therefore, the aim of this study was to improve our understanding of MT when used with a patient with IDD with PLP.

2 | METHODS

2.1 | Case description

The patient was a 53-year-old woman with moderate IDD, assessed with WAIS-IV in 2019 (Full scale IQ = 40). She also had a diagnosis of epilepsy and residual schizophrenia (diagnosed in 2000), characterized by the presence of negative symptoms (e.g. affective flattening, social withdrawal, apathy, lethargy), while with positive symptoms such as hallucinations or delusions were in full remission. She lived in a residential facility for people with IDD since 1994 (age: 29 years old).

In 1984 (age: 19 years old), she suffered several injuries after accidentally falling from the 6th-floor of a building: L5–S1 fracture dislocation with flaccid paralysis beneath L4 in bilateral lower limbs, open fracture of left tibia and right foot calcaneal fracture (heel bone).

In 2001 (age: 36 years old), she was diagnosed with left foot calcaneal osteitis, and between 2016 and 2018 she underwent continuous surgical debridements and cures to treat calcaneal ulceration in the same foot. In 2018 (age: 53 years old), a supracondylar amputation of the left leg was performed due to ulcer complications. Other factors contributing to the overall pain experience were mild kyphosis, multiple fractures sequelae and a continued sitting position in a wheelchair.

The patient was taking tramadol hydrochloride 60 mg daily since the amputation in 2018. Regarding antipsychotic medication, she was taking levomepromazine and haloperidol (300 mg and 60 mg daily).

After the intervention, the patient was feeling phantom pain in the amputated leg, as well as pain in the preserved leg and hip. According to the caregiver staff, the patient often screamed in pain, complaining about continuous pain in both her legs. When asked about the pain, she usually expressed discomfort and described painful sensations in the amputated limb worsening in the morning.

Given this situation, the psychological and medical staff of the facility decided to implement a MT intervention to help reduce (or eliminate) the PLP problem.

2.2 | Measure

Pain intensity. We used the Revised Iowa Pain Thermometer scale (IPT-R) to assess pain intensity related to the phantom limb (Ware et al., 2015). The IPT-R is an 11-point verbal descriptor scale that uses a vertical thermometer with four pain descriptors, which are as follows: 0 ('No pain'); 1–3 ('Mild pain'); 4–6 ('Moderate pain') and 7–10 ('The most intense pain imaginable'). IPT-R pain intensity scores have demonstrated to provide valid and reliable information when used with patients with pain, regardless of age, gender, level of education and presence of cognitive impairment or communication difficulties (Herr, 2011; Herr et al., 2007; Taylor et al., 2005).

2.3 | Procedure

The study followed an A-B-A-B design (Baseline assessment of pain before the intervention—Pain assessment during the MT treatment—Pain assessment in the withdrawal of MT treatment—Pain assessment during the reintroduction of MT treatment; Figure 1).

Informed consent was obtained, and all procedures were performed in accordance with the standards of the 1964 Helsinki Declaration and its later amendments.

Two days prior to starting the study, the patient was taught how to report her pain intensity using the IPT-R scale. The scale was presented to her printed on A4 size paper, and some of her own painful past experiences were used as examples, to help her discriminate them as more or less painful on the verbal descriptor scale and the visual representation of the pain thermometer. In addition to this, the instructions on how to proceed with the rating method were repeated at the beginning of each session.

In October 2018, the IPT-R scale was administered to establish the patient's baseline pain levels for 23 days *(Baseline 1)*.

In phase B of the A-B-A-B design, the MT intervention *(Mirror therapy 1)* was conducted. Starting in late November 2018 until mid-February 2019, it consisted of 60 sessions of 10–12 min between 10:30 and 11:30 am, 5 days per week (from Monday to Friday). The structure for each session was set as follows: 1) Pain intensity assessment at the beginning of the session; 2) Exercises with the mirror box; and 3) Pain intensity assessment after the MT exercises.

During the MT sessions, the patient exercised her right leg and watched its movements in the mirror box under the direction of a qualified therapist, while the amputated limb was inside the box (see Figure 2). All sessions were carried out under the same conditions following a structured protocol, with three sets of 10 repetitions for each of the following exercises:

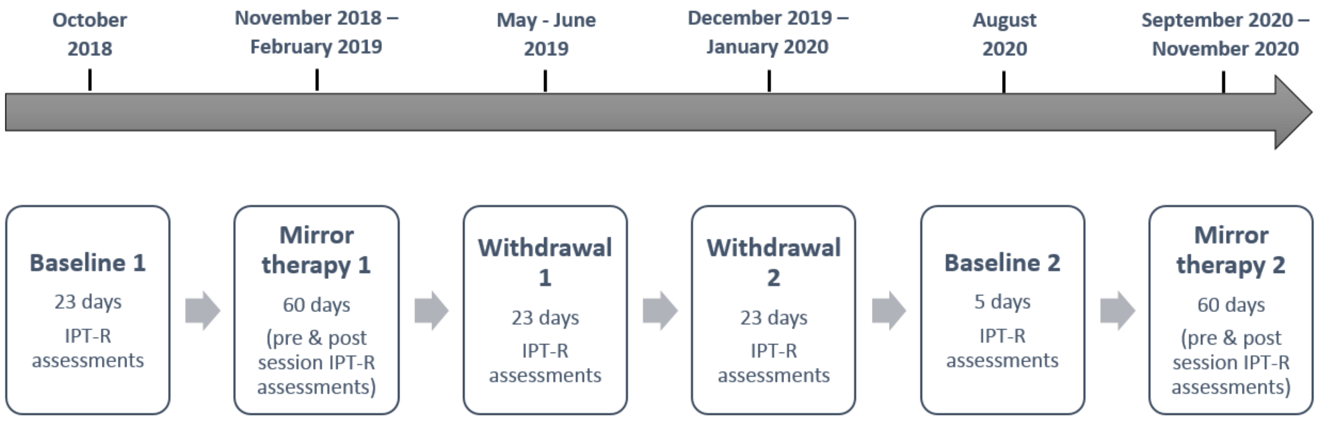

FIGURE 1 Phases of the study

FIGURE 2 Pictures of the mirror box used and a treatment session

- 1st set: Slowly move the right foot 10 times to the right
- 2nd set: Slowly move the right foot 10 times to the left
- 3rd set: Slowly raise the right foot about 30 degrees, 10 times

Three months after the intervention, pain intensity was measured again in two 8-week terms, between May and June 2019 *(Withdrawal 1)*, and between December 2019 and January 2020 *(Withdrawal 2)*.

At the end of August 2020, the patient started complaining again about feeling intense phantom limb pain. Pain intensity was measured to establish the second baseline pain level for 5 days *(Baseline 2)*, and after that, the MT was re-introduced for 60 more sessions between September and November 2020 *(Mirror therapy 2)*, under the same conditions explained above. Once again, pain intensity was measured before and after each MT session.

There were no changes in the patient's treatment plan other than the addition of the MT.

3 | RESULTS

Patient's pain intensity scores showed a decreasing trend that was either maintained or improved with time. However, there is an exception to this in the Baseline 2 period, 1.5 years after the first MT intervention (Mirror Therapy 1). In the second period of MT (Mirror therapy 2), pain intensity scores dropped to their previous level, albeit somewhat lower than then.

Table 1 summarizes the information about pain intensity scores along the phases in the study. Table 2 displays the raw changes and percent changes in the IPT-R scores between the different phases of the study. Figure 3 shows an overview of all pain intensity measurements during the different phases of the study.

TABLE 1 Patient's pain intensity scores along the different phases of the study

	Pain intensity scores	
Phase study	**Mean**	**Standard deviation**
Baseline 1	8.17	1.25
Mirror therapy 1 Pre-session	7.24	1.40
Mirror therapy 1 Post-session	5.76	1.20
Withdrawal 1	5.87	0.77
Withdrawal 2	6.02	0.64
Baseline 2	8.20	0.57
Mirror therapy 2 Pre-session	5.33	0.92
Mirror therapy 2 Post-session	4.25	0.76

The data showed that patient's pain intensity scores decreased following MT. As can be seen in Table 2, there was a raw change of 2.41 points in pain intensity scores from Baseline 1 to Mirror Therapy 1 Post-session, which is a 30% change. From Baseline 2 and Mirror Therapy 2 Post-session, the raw change was 3.95, a 48% change. Importantly, data showed a raw change of 3.92 points (48% change) in the mean pain intensity scores between the first and last phases of the study (i.e. Baseline 1 and Mirror therapy 2 Post-session; see Table 2 for additional information on raw and percentage of changes).

Staff entries in the patient's clinical record during the intervention were reviewed to identify complaints about pain and pain-related behavioural disturbances in order to corroborate the clinical implications of the reported pain reduction. During the Baseline 1 period, six staff entries referred as 'cries of pain, yelling and banging on objects'. Four pain-related entries were also identified in August 2020, resulting in the re-introduction of the MT. No pain-related entries were written during Mirror

Comparison	IPT-R mean raw change	IPT-R mean percentage change (%)
Baseline 1 & Mirror therapy 1 Post-session	2.41	30
Mirror therapy 1 Pre-session & Post-session	1.48	20
Baseline 1 & Withdrawal 1	2.30	28
Baseline 1 & Withdrawal 2	2.15	26
Withdrawal 2 & Baseline 2	2.02	27
Baseline 2 & Mirror therapy 2 Post-session	3.95	48
Mirror therapy 1 Pre-session & Post-session	1.08	20
Baseline 1 & Mirror therapy 2 Post-session	3.92	48

TABLE 2 Pain intensity mean raw change and percentage change scores between the different phases of the study

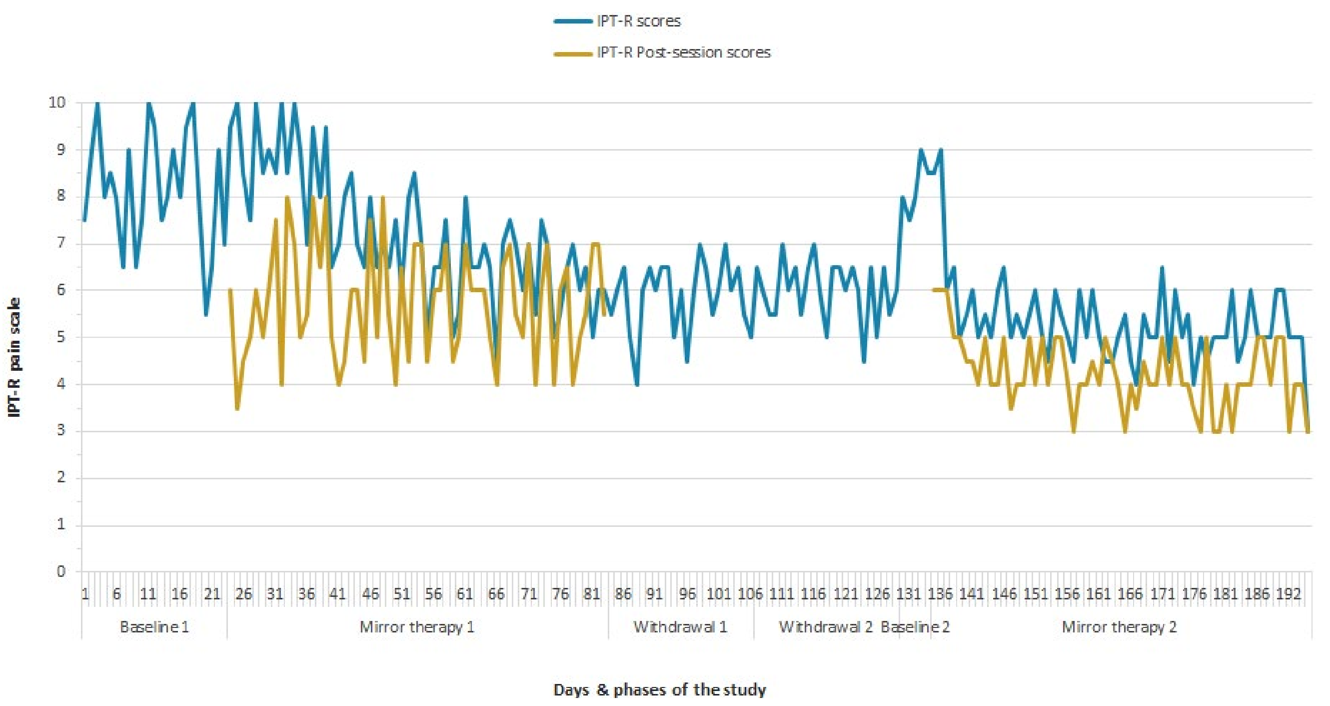

FIGURE 3 Patient's pain intensity reports (IPT-R) across the study. The blue line shows pain intensity scores during the different phases of the study (including pre-session assessments right before the MT exercises during Mirror therapy 1 & 2 periods). The yellow line shows post-session pain intensity scores during Mirror therapy 1 & 2 periods

therapy 1, Withdrawal 1, Withdrawal 2 and Mirror therapy 2 phases.

4 | DISCUSSION

The aim of this study was to improve our understanding of MT when used with a patient with IDD with PLP.

The findings suggest that the MT was effective in achieving a clinically meaningful reduction in the patient's pain intensity scores. Further validity for the positive effects of MT on the patient's pain intensity is provided by a reduction in the number of pain-related entries in the patient's clinical record.

Research has shown that a 30% decrease in pain intensity scores can be considered a clinically meaningful change (Younger et al., 2009). Clinical significance is the practical importance of an intervention effect—whether the change makes a real difference to subject lives, and represents an important factor in the evaluation of intervention effects, including prevention, education and rehabilitation as well (Kazdin, 1999; Ranganathan et al., 2015). In relation to this, Farrar and colleagues (Farrar et al., 2001) showed that a 28% change was associated with a 'very much improved' response from patients when reporting on their global impression of change (Farrar et al., 2001; Younger et al., 2009). Interestingly, a recent study (Foell et al., 2011) reported that an average decrease of 27% in PLP after MT, was associated with a reversal of dysfunctional cortical organization in primary somatosensory cortex, based on fMRI data analyses, thus indicating that maladaptive changes in cortical organization are reversed during MT.

It is also important to note that the average pain intensity scores on the second period of MT was 4.25, and this could indicate that there is residual somatic pain. As stated earlier, there were other factors contributing to the overall pain intensity on which the MT has no effects, such as mild kyphosis, multiple fractures sequelae and continued sitting position in a wheelchair.

Studies on the effects of MT have not reported long-term follow-ups (Campo-Prieto & Rodríguez-Fuentes, 2018), but some have informed on short term follow-ups (i.e. 1–6 months) showing the benefits of MT, particularly the reduction in pain intensity (Timms & Carus, 2015). This study is unique as it reports on a very long-term follow-up. During this two-year research period the patient experienced an increase in PLP 1.5 years after the first MT intervention. That increase in PLP led to a second period of MT intervention which resulted in a significant decrease in pain intensity scores, as in the first period. This finding supports the suggestion for MT to become a long-term intervention in the treatment of individuals with PLP, because PLP tends to slowly return over time (Foell et al., 2014; Moseley, 2006; Timms & Carus, 2015) and the treatment effects are associated with cortical reorganization and enhanced by regular and frequent use of MT (Foell et al., 2011; Timms & Carus, 2015). Accordingly, current clinical guidelines recommend that the total duration of the MT treatment should depend on how long improvements in pain are perceived by the patient with PLP and the therapist, or on which extend the treatment is beneficial or necessary to achieve sustainable effects (Rothgangel et al., 2015, 2016).

Besides the above-mentioned benefits, MT is a valid, simple, easy-to-use and low-cost therapeutic technique that can be widely used to treat PLP (Campo-Prieto & Rodríguez-Fuentes, 2018; Kiabi et al., 2013). Despite this, as far as we know, there is no single research about MT treatment in people with PLP and IDD, so the present case-study is the first of this kind.

The lack of previous studies about MT in IDD has been attributed to a presumed lack of effectiveness of this technique due to their cognitive limitations (Rothgangel et al., 2015, 2016). However, this study shows the effectiveness of MT in an adult with moderate IDD and PLP. It is important, however, to adapt the procedure to the personal characteristics of the patient. For example, some cognitive and communicative abilities are important to ensure that the patient will be able to focus at least for ten minutes on the mirror and follow therapist instructions (Rothgangel et al., 2015, 2016). Thus, for some, the MT intervention will require specific preparations It is also important to take into consideration staff observations as a complementary method to corroborate the patient's self-reported pain.

It should be noted that pain assessment for persons with IDD may be challenging due to difficulties in self-reporting of pain, related to limitations in communication skills and cognitive ability (Defrin & McGuire, 2019; Doody and E. Bailey, 2017). However, there are alternative means to assess the pain in this population group. The use of validated tools (Zabalia, 2016), can be completed with health assessment and observation of behaviours (Doody and E. Bailey, 2017).

Following this path, more work is needed to explore MT in people with PLP and different levels of cognitive limitations, such as borderline intellectual functioning, and mild and moderate IDD. It would also be interesting to know whether the persons with severe and profound levels of IDD can benefit from this intervention.

Further studies are also required to evaluate the impact of the MT intervention on other variables such as mobility improvement, enhancement and maintenance of wellbeing and mood, attenuation of disruptive behaviours and adjustment or reduction of long-term opioid use in persons with IDD.

This is a single-case study, and the findings are specific to the patient that received the treatment. However, single-case study is a well-known and widely used strategy (Manolov & Moeyaert, 2017), in the development and implementation of evidence-based practice (Byiers et al., 2012). It can be used to design personalized interventions to promote health outcomes and empower individuals to take an active role in their own health (McDonald et al., 2020). Furthermore, the ABAB design is one of the most straightforward and strongest treatment effect demonstration, as they have the benefit of an additional demonstration of experimental control with the reimplementation of the intervention (Byiers et al., 2012).

To summarize, this study adds new and important clinical information, as it is a unique study about the effects of MT to help manage pain intensity in an individual with IDD and PLP. Moreover, it is the one reporting on the longest follow-up period. The findings show that MT was effective in achieving a clinically meaningful reduction in the patient's phantom limb pain intensity scores, and highlight the importance of establishing an adequate management of pain in this population to improve their quality of life. However, additional longitudinal studies, with representative samples, are needed to confirm the validity of the findings.

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CONFLICTS OF INTEREST

The authors declare no financial or other relationships that might lead to a conflict of interest related to this study.

AUTHORS' CONTRIBUTIONS

The authors confirm contribution to the paper as follows: study conception and design: D.G & R.M.L.; data collection: D.G.; analysis and interpretation of results: D.G. & A.F.; draft manuscript preparation: A.F.; critical revision of the article and final manuscript draft: L.S.C., J.M., R.M.L. and A.F. All authors discussed the results, commented on the manuscript and approved the final version of the manuscript.

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