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Postprint copy deposited in [CURVE](#) March 2014

Original citation:

Hagenberg, A. and Carpenter, C. (2014) Mirror Visual Feedback for Phantom Pain: International Experience on Modalities and Adverse Effects Discussed by an Expert Panel: A Delphi Study *PM and R* (article in press).

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Original Research

Mirror Visual Feedback for Phantom Pain: International Experience on Modalities and Adverse Effects Discussed by an Expert Panel: A Delphi Study

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Background: Mirror visual feedback (mirror therapy) is practiced worldwide in very different ways to alleviate phantom pain; no study has compared these variations yet or researched the associated risk and harm.

Objectives: To establish use and justification of a generally accepted mirror visual feedback treatment plan after amputation; to explore the occurrence and handling of adverse effects; and to increase knowledge about contributing factors.

Methods: Experiential knowledge of 13 experienced practitioners from 6 countries and 5 professions was explored with a 3-round Delphi technique.

Results: Experience with the use of 5 different treatment plans was described, of which 1 has never been mentioned in the literature: an intense 1-off plan in which the illusion was carefully set up before the patient was left to the experience with no interference, resolving pain as well as adverse effects. In the 4 known treatment plans, the expectations of response time varied, which influenced the definition of responders/nonresponders; the set-ups, control, and use of material reflected the professional background of the practitioners. Contraindications also were defined according to the professional confidence to deal with the adverse effects. Adverse effects were reported, including emotional reactions, pain increase, sensory changes, freezing of the phantom limb, dizziness, and sweating. The attitude toward, and the handling of, adverse effects varied in patients as in practitioners according to their professional background. A tool to fine tune the experience was reported with covering of the limb during therapy. Full consensus was reached on several treatment modalities.

Conclusion: Analysis of the results suggests that the different treatment plans suit different patients and practitioners. Matching these could enhance effectiveness and compliance. Knowledge about adverse effects needs to inform treatment decisions. These findings triggered the development of a mirror visual feedback gateway to guide patients to the treatment plan for their needs, and to collect data from the practitioners to enhance neuroscientific understanding and inform practice.

PM R 2014;■:1-8

INTRODUCTION

Phantom pain is a well-known and frequently experienced problem after amputation, and often occurs in connection to altered or nonexistent movement abilities of the phantom limb [1-3]. In 1993, Ramachandran and Altschuler [3] first discovered mirror visual feedback (MVF) to be able to address the issue of phantom pain by creating an illusion with a mirror in a box that was placed in front of the patient in such a way that the missing limb could be seen as a reflection of the remaining limb. This visual input resulted in pain relief. Twenty years later, a number of professions use the principles of MVF (also known as mirror therapy or mirror box therapy) in treating chronic pain and learned nonuse. Research has been conducted on the effectiveness of MVF with the identified problem of heterogeneous study designs [4-6]. The treatment plans vary to a great extent (Table 1) [7-14]. These have never been compared and researched. Adverse effects are only rarely mentioned (Table 2) [7,10,11,14-16] and are by no means thoroughly researched [4]. Only

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Disclosure: nothing to disclose

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Disclosure: nothing to disclose

Presented at the scientific meeting of the Physiotherapy Research Society in Cardiff on April 9, 2013.

Submitted for publication June 16, 2013; accepted January 5, 2014.

Table 1. Treatment plans of mirror visual feedback in the literature

Author	Type of Study	Name of Treatment Plan	Structure	Frequency and Duration
Moseley, 2006 (9)	RCT	Graded Motor Imagery	Three phases: limb laterality recognition, imagined movements, mirror therapy	2 wk each phase, with hourly home exercise program
Grünert-Plüss et al, 2008 (7)	Review, protocol, and case series of 52 patients	St Gallen protocol	Looking first, then individual program	5-6 times a day, not more than 5-10 min
McCabe, 2011 (11)	Background and protocol	Bath MVF treatment protocol	Body schema, imagining movements first	5-6 times a day, not more than 5-10 min
MacLachlan et al, 2004 (8)	Case study	No specified	Fading out of therapist-mediated intervention over 3 wk	Practicing 2-4 times a day, 10 exercises, 10 repetitions each
Chan et al, 2007 (10)	RCT	Not specified	Movements, not specified	15 min/d for 8 wk
Darnall and Li, 2012 (14)		Self-delivered mirror therapy	7-min DVD and written instructions, diary	25 min/d for 8 wk
Mercier and Sirigu, 2009 (12)	8 Case studies	Visual virtual feedback	10 unilateral movements, 10 repetitions each	2 sessions/wk for 8 wk, lasting 30-60 min
Kawashima and Mita, 2009 (11)	Case study	Not specified	Synchronic and periodic wrist movements, smoothly and in as large a range as possible	1 h/wk for 3 mo

RCT = randomized controlled trial; MVF = mirror visual feed.

1 article based on retrospective evaluation of patient records specifically discusses the frequent adverse effects of MVF experienced during treatment, which resulted in withdrawal from MVF [16].

Thus, the questions about adverse effects of MVF, at which point in the treatment do they occur and why, and how to resolve these, remain unanswered in the current literature. Overall, there is considerable interest and clinical support for the use of MVF in treating phantom pain, and this treatment approach is practiced and taught in numerous countries in many different, possibly contradictory ways and without informing on risk and harm. This study sought to address the question, "How is MVF best practiced in treating phantom pain and what are the risks?" Secondary objectives included exploration of how MVF is used, the rationale underpinning clinical decision making, and the occurrence and management of adverse effects.

Research has been very limited, and we decided to perform a Delphi Study to further define the area. This study design can access the experiential knowledge of those who have seen and managed the patients to identify underlying principles. Three rounds of consecutive questionnaires are conducted with an expert panel of experienced practitioners.

Practitioners' experience represents 1 of the 3 components of evidence-based practice, namely, clinical expertise [17]. The outcome can then guide further research and practical application.

METHOD

The Delphi Method is defined as "a systematic and interactive research technique for obtaining the judgment of a panel of independent experts on a specific topic" [18]. It follows an iterative process of data collection, analysis, and feedback, and is particularly useful when there is a lack of empirical evidence or conflicting evidence [19]. A panel of experts are selected according to the criteria for expertise defined within the study context, and asked to participate in 2 or more rounds of structured questionnaires that progress to more specifically focused questions. After each round, the researcher provides an anonymous summary of the experts' input from the previous questionnaire, which also forms part of the subsequent questionnaire content [18,19]. The aim of the Delphi Method, through a process of initially open-ended questions to more specifically focused questions, is to decrease the variability of responses and to achieve a

Table 2. Adverse effects of mirror visual feedback in the literature

Study	Adverse Effects
Ramachandran and Rogers-Ramachandran, ¹⁵ 1996	Telescoping (perceived as beneficial, as pain disappeared together with phantom limb)
Chan et al, ¹⁰ 2007	Two brief grief reactions
Grünert-Plüss et al, ⁷ 2008	Pain increase possible
Casale et al, ¹⁶ 2009	Dizziness, irritation, uneasiness
Kawashima and Mita, ¹¹ 2009	Client vomited after an increasing feeling of nausea during the first session
Darnall and Li, ¹⁴ 2012	Boredom, increased depression, increase in phantom limb awareness, and phantom limb pain

predetermined level of group consensus [18,19]. A number of advantages contributed to the choice of the Delphi Method in this study: feeding back the experts' responses from previous rounds validates and enlarges the data, and rapid clarification can be obtained [20]. Also, the use of e-mail was convenient for the practitioners and could be assimilated more easily into their daily schedules [21].

The risk of bias is acknowledged. Practitioners were required to recall from memory effects that occurred in the past. In conducting a Delphi Study, the researcher may unconsciously impose his or her own interests and opinions on the emerging data [19]. However, our interest was to understand the matter in depth instead of seeking support for a specific protocol or assumption. This was reflected in the search of the widest possible range of treatment plans practiced and adverse effects experienced, with no limitation to profession or site. Every effort was made to limit bias through reflection on assumptions held about MVF, discussion at each phase with the second author (C.C.), and the use of a research journal. Practitioner bias was addressed through anonymity to eliminate competition between the various treatment plans, to provide continuous encouragement to report all observations, and to comment wherever they wished on the open-ended questionnaires.

Rigorous selection of experts in the Delphi Method is fundamental [19]. The main inclusion criterion for this study was practical experience with patients in MVF after amputation. It was assumed that practitioners, who present this experience in public, such as in the literature, online forums, conferences, interest groups, or in teaching professional development courses, would fulfill this criterion. A strategy of purposive sampling was applied with the aim of covering the widest variety of treatment plans used and the widest variety of professions offering these:

1. Authors of the key literature were contacted.
2. A letter of invitation was posted on the Amputee Rehabilitation Network of the interactive site of the Chartered Society of Physiotherapists; amputation support groups (Amputierten-Initiative, Berlin and Empowering Amputees, USA) were contacted,
3. Internet search engines, such as Google (Google Inc, Mountain View, CA), and the video-sharing Web site YouTube (YouTube LLC, San Bruno, CA) were searched, and we identified and contacted these practitioners.
4. Twelve centers for artificial limb or pain services in the United Kingdom and Germany were contacted.

These strategies resulted in 8 participating practitioners with experience in MVF after amputation who agreed to participate. Subsequently, to increase the sample size, a snowball sampling approach was used; that is, the identified practitioners were asked to suggest further practitioners with experience in treating patients after amputation with MVF.

Fifteen further contacts were made; 5 were confident to have enough experience (on request, an aim of approximately 10 cases was given) or had limited experience but an unusual experience to report and agreed to participate. Of all the 36 MVF practitioners identified and contacted, 12 never replied, 3 gave a lack of time as a reason not to participate, 6 reported having no or very little experience in treating phantom pain and therefore did not think that they should participate as experts, and 2 never returned the first questionnaire despite reminders and therefore were not included. No practitioner who was experienced in treating phantom pain with MVF and wished to participate was refused participation.

The panel, therefore, consisted of 13 practitioners from 6 countries and 5 professions: medicine, psychology, physiotherapy, occupational therapy, and nursing. Two practitioners, who treated a relatively low number of patients with amputations and believed themselves unable to contribute with further data, withdrew after the first round. Eleven practitioners completed the study. Anonymity is a key component of the Delphi Method [19]. This was guaranteed to the practitioners. Informed consent was assumed when practitioners returned the first completed questionnaire. All the practitioners were assigned a code. Identifying features were removed from responses before being shared in subsequent rounds. The study received ethical approval from the Coventry University Research Ethics Committee. A pilot test of the first questionnaire was conducted, which involved 3 health professionals who were familiar with MVF but had not had enough experience to participate in the study. Minor changes were made to the first questionnaire based on the feedback obtained about the questionnaire design, the clarity of the questions, and content [22]. Because the Delphi Method requires data analysis and partial data presentation during each round, data collection and partial data presentation constituted an interwoven process [22].

In round 1, the Delphi approach customarily asks open questions to ascertain the nature, scope, and breadth of the topic being explored, which leads to very specific questions for clarification, cross-checking for verification, and further exploration of knowledge in subsequent rounds [19]. Questions in the first round focused on how treatment was organized in terms of length, duration, and frequency of sessions. Adverse effects were explored with respect to the nature, frequency, and severity of occurrence, management of the adverse effects, and how (or if) they were resolved; questions about experiences of nonresponse to MVF were included. The first round was concluded when all practitioners had returned the questionnaire. Each round had a deadline of 2-3 weeks for return of the completed questionnaire, and reminder e-mail messages were sent to the practitioners as needed. After each round, the responses were reviewed by 1 of us (A.H.), collated, and presented in tables.

As a result of this process, the second round consisted of data presented for comment or rating on a 5-point Likert scale [23]: clarification questions and new questions that arose from the emerging information about MVF related to treatment outcomes, use of sensory materials or manual techniques, time frames, and other factors. In the second round, practitioner responses were added to the pooled data. Consensus in this study was defined as 100% agreement (agree or strongly agree) of those who responded to the particular statement within a table of 12 statements (Table 3). The third round focused on acquiring information and clarification about specific topics, such as experience with phantom limb shrinking and awareness, restoring a lost illusion, and the effect that covering a limb with a cloth during MVF has on the intensiveness of the experience.

Once questionnaires from the third round had been received, another document was produced by using a cross-sectional “code and retrieve” method to collate the data [24]. This means that the data from all questionnaires were labeled and brought together under various categories under recognition of their source, whereupon analysis was performed within the categories as well as across sections, for example, with regard to the professional background of the practitioners and the number of patients that they had seen. The data analysis was discussed with the second author (C.C.) after each round.

RESULTS

Equipment and the Illusion

The mirror equipment used comprised the following:

- simple, inexpensive mirrors from discount stores;
- mirror tiles from bathroom stores glued to the side of a cardboard box;
- large, plain, wall-style mirrors;
- long free-standing mirrors;

- wooden stands with acrylic glass mirror;
- commercially produced mirror boxes; and
- E-shaped constructions with a reversible mirror in the middle;
- projection system with a mirror.

Most therapists did not consider the nature of the equipment important, or the angle at which the mirror was set up, as long as a strong and vivid illusion could be achieved when the patient was in a relaxed position. The illusion could be spoiled by a patient's lapse of concentration, forgetting to move the phantom limb, or moving the 2 limbs at different speeds or through different ranges. Jewelry, watches, or tattoos spoiled the mirror illusion in some cases but not in others. An unusually high percentage (40%) of nonresponders to MVF was reported by a practitioner who defined 3 weeks as the response time.

Treatment Plans

The different treatment plans are shown in Figure 1 and can be described as follows:

- Remote MVF. Instruction via a leaflet and a DVD, which focused on patient education, with remote follow-up. This plan is appropriate for highly motivated and educated patients.
- Intense MVF. One lengthy session up to 3 hours in length. After the assessment and development of the specific treatment plan, the mirror illusion was set up. Once the process was underway, there was no interference from the practitioner, and the patient was left to explore the experience on his or her own. This approach often resulted in resolution of pain for several days, which could then often be permanently resolved after regular or as-needed self-administered sessions. Follow-up was conducted remotely. This approach has been effective for patients who had previously experienced no success with another MVF treatment plan.

Table 3. Consensus on statements about mirror visual feedback

Statement	% Agreement	% Disagreement	Consensus
Good education and preparation for mirror therapy is a key factor	100	0	Full
Individual set-up of mirror therapy is most important	100	0	Full
Face-to-face guidance is most important	100	0	Full
Reassessing is most important because some patients do not use the mirror correctly and therefore do not succeed	100	0	Full
The setting must be quiet	91	9	Most
Finding triggers that cause phantom pain is most important	80	20	Most
Sensory changes were seen as positive by the patients	90	10	Most
Mirror therapy requires a lot of concentration and emotional energy	73	27	Tendency
Time since amputation plays a role	66	33	Tendency
The repeated use of the mirror shrinks the phantom size	63	37	Ambiguous
Very analytical right-hemisphere–dominated patients have difficulties with mirror therapy	50	50	Ambiguous
It does not matter if a patient “believes” in the mirror illusion, the brain just works this way	40	60	Ambiguous

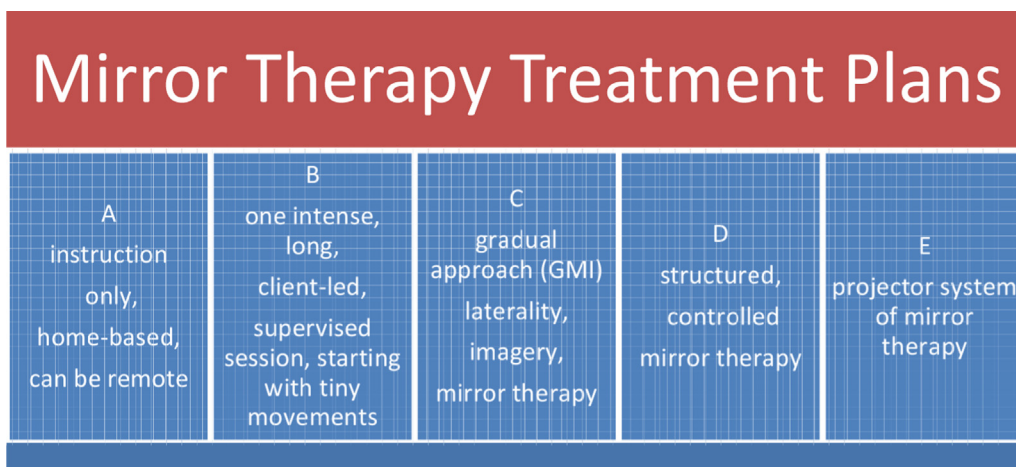


Figure 1. Treatment plans used by expert panel.

- C. Graded MVF. In this approach, MVF was preceded by up to 2 weeks of laterality training (distinguishing left from right with flashcards) and up to 2 weeks of imagery training of positions and movements of the phantom limb before MVF was introduced. This structure was based on graded motor imagery and was controlled and monitored by the practitioner, in usually 1 or 2 face-to-face sessions per week. It required self-administration of short sessions several times a day.
- D. Structured MVF. This approach involved highly structured sessions that used the mirror from the beginning. The various structures described here are summarized in [Figure 2](#).
- E. Prerecorded MVF. This approach used prerecorded movements of the sound limb or of another person's limb projected onto the mirror. This allowed unilateral movement but was restricted to the treatment setting.

Consensus

All the practitioners (100%) agreed that thorough patient education and preparation, individual set-up, face-to-face guidance, and reassessment are key factors in MVF after amputation. The majority of practitioners agreed on the need for a quiet setting and on the importance of individual triggers of patients' phantom pain. Other statements on observations were more ambiguous ([Table 3](#)). It was agreed that it took patients 1-10 minutes to become immersed in the illusion, and the session length was usually determined by the patient's ability to concentrate and to tolerate the program. The frequency of practicing was mostly limited by time constraints.

No consensus was reached on the optimal length of MVF sessions (the range was from less than 5 minutes to 120-180 minutes) or the frequency of sessions (the range was a single session to 6 or more times a day). A minority of practitioners

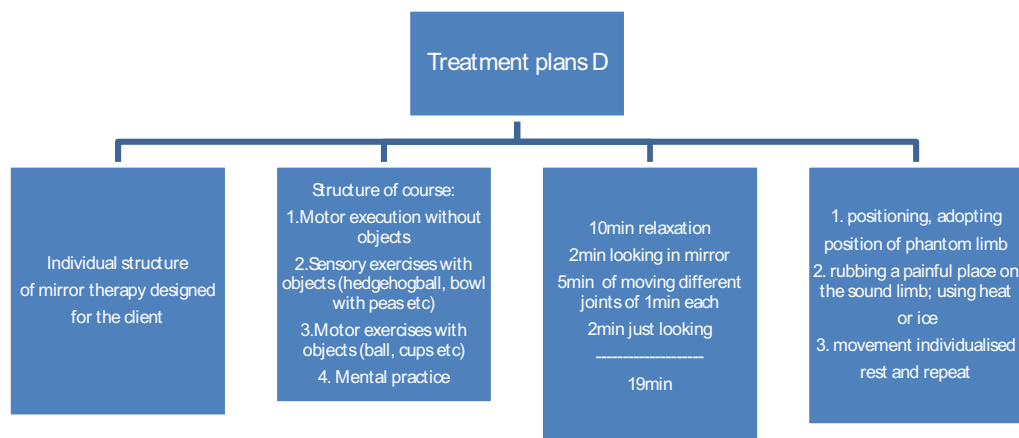


Figure 2. Structures used within treatment plan D (structured mirror visual feedback) of this study.

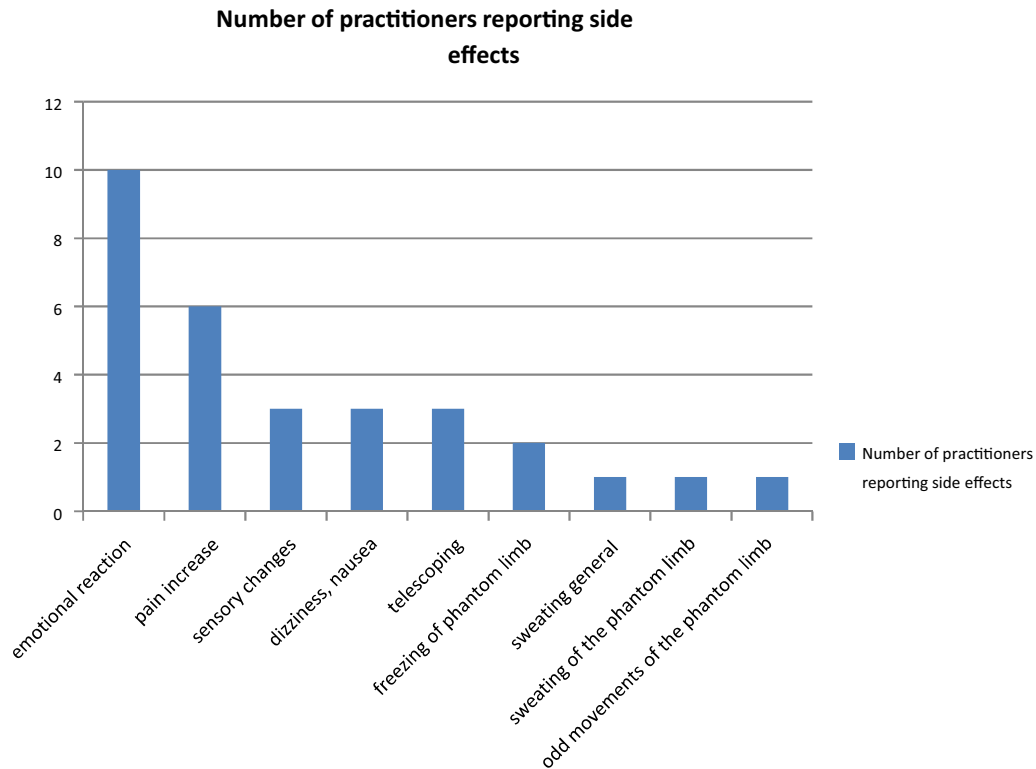


Figure 3. The number of practitioners who reported adverse effects that occurred with mirror visual feedback.

believed strongly that practicing for a short time and often was more beneficial than longer and less-frequent sessions. There was no consensus on the causes of nonresponse, but suggestions included a lack of remapping phenomena (referred sensations to locations on other body parts, eg, touch on the ipsilateral cheek, neck, upper arm, or genitals felt on the phantom limb), effort sensation (felt level of effort when moving phantom limb), sensory referral (touch of the same location in another person felt on the phantom limb) of tactile sensation (when touching an object), telescoping (perceived shift of position of phantom limb into its proximal parts or stump), and poor technique.

Adverse Effects

A variety of adverse effects were identified (Fig 3); these comprised emotional reactions, pain increase, sensory changes, dizziness, nausea, telescoping, freezing of the phantom limb, overall sweating, sweating of the phantom limb, and spontaneous movements of the phantom limb. Emotional reactions, described by 85% of the practitioners, were associated with suddenly “seeing” the limb and sometimes with “taking it away again.” These reactions, observed in the first and second sessions only, ranged in severity from “amazement” to, in rare cases, significant depression. Physiotherapists and occupational therapists generally chose to stop the MVF if emotional reactions occurred, and some

classified them as a contraindication for MVF. Remarkably, those practitioners with psychological training saw a necessity in accepting emotional reactions as long as posttraumatic stress disorder was treated before commencing MVF.

When pain increase occurred, it was, according to two-thirds of the practitioners, limited to the duration of the MVF session. Individual practitioners, however, reported delayed onset briefly after the session or pain increase that lasted after the session and that spread in location. Most practitioners advised patients not to tolerate pain increase. Residual limb spasms associated with irritated neuromata and increased or excessive activity were mostly held responsible and were resolved with relaxation techniques, looking without moving, minimal session time or slower movements without moving the stump muscles, a quiet setting, and comfortable positioning. Manual techniques also were applied to either the stump or to the unaffected limb. Pain increase was regarded as a reason to stop MVF for the moment and alter posture, relax the residual limb, or reduce intensity or duration of the session. Yet, a minority regarded effort-related pain increase for the duration of the session as the norm. Sensory changes in the phantom limb were common and were generally perceived as enjoyable.

Locking or freezing of a phantom limb is a phenomenon that can typically be resolved with MVF. Interestingly, 2 practitioners witnessed this as an adverse effect from MVF treatment. In 1 case, the locked position was not resolvable

with movement imagery, and the patient left in distress. Another practitioner described the same effect as a worrying and dramatic experience that was resolved by the patient himself during the long session. On follow-up, the patient rated it as one of the most exciting experiences of his life with complete and permanent pain resolution as a result. Dizziness and nausea were reported as occasional mild effects except in 1 patient who subsequently withdrew from MVF. Subtle eye problems were made accountable, and it was recommended to cover the affected limb during MVF as a means of refining the MVF experience. Sweating in general, spontaneous movements, or “odd” behaviors of the phantom limb occurred as transient phenomena. Boredom during therapy was associated with monotonous exercise programs and sleep deprivation; it was addressed with exercise variety and progression, short breaks, incorporating music and meaningful functional tasks, and shorter sessions.

DISCUSSION

Defining nonresponse after 3 weeks seemed too early when others reported improvement after 5 months, which confirmed an assumption of the possible need of longer practice [3]. In some cases, mirror therapy was successful despite patient skepticism whereas in other cases, patient expectation did not produce a positive outcome, which questions the assumed prerequisite that one has to believe in the illusion [13]. The variety of treatment approaches and attitudes (outlined earlier) reflects the practitioners’ professional experience and confidence, and supports the suggestion by Ramachandran and Altschuler [3] that different treatments may suit different patients.

The low incidence of dizziness and nausea reported in the current study is in contrast to the high incidence reported in the study by Casale et al [16], a note evaluation with a high withdrawal rate from MVF treatment [16]. The researchers assumed a conflict of body schema (image of normal limb vs prosthesis) to be responsible for the high incidence of irritation in their patients. Whether boredom of flexing and extending a limb for 30 minutes or subtle eye problems in a mostly elderly inpatient cohort may have played a role remains to be seen in further research. As a precaution, careful management is advised, especially because the literature mentioned vomiting as a possible outcome [11]. Whether the use of clothing can help to prevent these exacerbations needs to be investigated.

CONCLUSION

Important insights have been gained about the practice and adverse effects of MVF as a result of using the Delphi Method to access experts’ knowledge. Five different treatment approaches were described, 1 of which (intense MVF) has not been described in the literature. This approach, being longer and more intense, was successful in cases of patients

who had previously failed with other MVF treatment. This indicates an area for further research. Each treatment plan has specific advantages related to the professional background of the practitioner and individual patient needs; therefore, the best practice seems to be to match these.

Adverse effects have been identified and described here, and, although they were generally mild, some severe adverse effects occurred. These were freezing of the phantom limb and severe nausea. Practitioners and patients interested in using MVF should be informed of these risks. Tools to address the adverse effects have been described by practitioners. The findings of this study will contribute to the development of an MVF gateway project that will provide guidance for patients in choosing the treatment plan that best meets their needs and will collect anonymous treatment data to enhance understanding of the underlying principles.

ACKNOWLEDGMENTS

We thank Jessica Aldis and Richard Catlow, the colleagues who pilot tested the first questionnaire, and the participating practitioners for their time and commitment to sharing their experiential knowledge.

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