REVIEWS

The effects of exergaming on individuals with limb loss: a systematic review

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Abstract

Losing a limb is a life-changing experience. Affected individuals (amputees) have reduced physical range of motion, poorer balance control, strength, and experience pain and fatigue. Exergaming is currently used in physical rehabilitation. There is currently no consensus on the efficacy of exergaming delivered to people with missing limbs. This systematic review aims to evaluate and summarize the current research on the effects of exergaming among individuals with missing limbs. Studies reporting on exergaming intervention delivered to individuals receiving prosthetic rehabilitation were included in the analysis. Ten electronic databases were searched. Twelve articles were identified. Data were extracted and assessed for quality.

Three main categories of exergaming interventions comprised custom made exergames, Nintendo Wii and the Computer Assisted Rehabilitation Environment (CAREN) system. All participants in the studies were adults, except for one study which evaluated exergaming in adolescents and children. Studies reported improvements in EMG muscle control, cognitive-motor ability, walking capacity, function, balance and reduced pain, and positive experiences amongst most participants. Results suggest that exergaming supports improvements in physical activity, balance, cognition, emotional states, quality of life and pain.

Exergaming interventions administered to people with missing limbs show heterogeneity in protocol, duration and gaming platform. Although there was evidence of improved outcomes in participants, the efficacy of exergaming is inconclusive due to varied differences in types of amputation, participant characteristics and assessed outcome measures. Nevertheless, reported enjoyment, acceptance and levels of motivation during exergaming appear to support the feasibility of exergaming for prosthetic training.

Keywords: amputees, exergaming, active video games, rehabilitation.

Introduction

Losing a limb is a life-changing experience and has negative impacts on the psychological and physical well-being of affected individuals (Senra et al., 2012). People with limb amputations experience decreased levels of physical activity and impaired balance (Gaunaurd et al., 2011; Ku et al., 2014). They may also experience phantom limb pain for the residual limb (Kooijman et al., 2000; Nikolajsen & Jensen, 2001). Thus, rehabilitation through exercise may encourage physical functioning following amputation through the restoration of muscle strength, endurance, power and physical flexibility (Vestering et al., 2005).

Using exergaming for therapeutic purposes is gaining interest (van Diest et al., 2013). One of the most recent interventions currently used in physical rehabilitation is exergaming (Barry et al., 2014; Robinson et al., 2015; Tough et al., 2018). Exergaming can be defined as physical exercise in a serious gaming environment enabled by

digital technology (e.g. Nintendo Wii Fit) (Oh & Yang, 2010). It has been recommended as an appropriate form of rehabilitation for several clinical groups, including cerebral palsy related disabilities in paediatric patients and age-related disabilities in older people (Goble et al., 2014). Karahan et al. (2016) reported significant improvements in pain, disease activity, functional capacity and quality of life in people with ankylosing spondylitis after exergaming. Another study that evaluated the effectiveness of exergaming on balance reported not only improved balance and gait amongst people with multiple sclerosis, but also significantly higher improvements in gait whilst dual tasking after exergaming (Kramer et al., 2014).

Despite potential health benefits of exergaming, there are differences in gaming pace and levels of cognitive complexity in certain exergames. For instance, people with Parkinson's disease have found difficulty in playing exergames that require fast physical movements (dos Santos Mendes et al., 2012). Therefore, using exergaming in rehabilitation must suit the therapeutic goal for which it

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was intended (Pirovano et al., 2016). Indeed, exergaming applications should support a wide range of physical exercises, allow the interventions to be personalized, involve the use of sensors that are comfortable to wear, and provide feedback to improve performance and encourage adherence without cognitive overload (Doyle et al., 2011).

Exergaming is a relatively new intervention in the rehabilitation of people with missing limbs (i.e. amputees) (Imam et al., 2018). To fill this knowledge gap, the authors conducted a systematic review of the literature to look at exergaming interventions and related physical health outcomes from exergaming to provide a broad overview of exergaming effects in people with missing limbs, and to inform evidence-based clinical practice.

Methods

Our systematic review is retrospectively registered with PROSPERO (1). The reporting of this review is consistent with PRISMA guidelines (Liberati et al., 2009). In this review, the terms patient and participant are synonymous.

Search strategy

A systematic search was conducted in eight electronic databases from April 2019 and updated in August 2019. The electronic databases were the following: CiNAHL, Google Scholar, Cochrane Library, MEDLINE, ScienceDirect, SPORTDiscus, PEDro, and Web of Science (science and social science citation index).

The titles, abstracts and keywords of publications resulting from the searches, where applicable, were searched with the following search terms: rehabilitation or 'limb loss' or amput* or telerehab* or physiotherapy or gam* or wii or digital or video gam* or prostheti* or 'virtual reality' or 'augmented'. The references of included publications were also checked.

Screening process

The first author (JD) screened the initial 3,773 publications and removed duplicates from the initial search. All titles and abstracts were screened independently by the two authors (JD and IC). Any disagreements over publications were resolved through a discussion until a consensus was reached.

Study selection

Participants, intervention, comparisons (if any), outcomes and study design were used to identify the inclusion and exclusion criteria for the study.

Participants

We formulated our inclusion criteria to include individuals of all ages with missing limbs either due to surgical removal or traumatic disarticulation by injury or by surgical amputation. People awaiting diagnosis of amputation or surgery including individuals with congenital absence of limbs were excluded.

Intervention

Exergames must have encouraged physical movement or physical activity in order to interact with the game. They should have demonstrated at least some of the following characteristics: interactivity, cognitive-physical purpose, presence of an opponent or incentive to win points, exploration of a virtual environment by physical movement (e.g. walking in a virtual environment where the terrain is uneven which serves as "obstacles" or playing exergames

by muscle activity) and the possibility of winning or losing.

Comparison

No comparative groups were required for inclusion.

Outcome measures

The outcomes were health related such as pain perception (i.e. phantom limb pain in amputees), balance, physical functioning and physical activity outcomes, including emotional states related to exergaming (i.e. motivation, acceptance of the exergaming intervention).

Study design

There were no limitations on trial design. However, reviews of the literature, articles from abstracts or summaries presented in a congress or conference were not included. Only articles available in English were included.

Data extraction

Data extraction was performed by one reviewer before being verified by the second reviewer. The following were extracted from selected studies: participant characteristics including type of amputation, details of intervention, equipment and setting, and clinical outcome measures. Two reviewers screened the articles independently. Any discrepancies were resolved through a discussion between the two reviewers. A third reviewer was consulted if there was a need for further resolve.

Risk of bias and quality of evidence

The Cochrane Collaboration Tool was used to assess risk of bias in randomized controlled trials (Higgins et al., 2011). The Cochrane Collaboration tool assesses biases as a judgement (high, low, or unclear) for individual elements from five domains (selection, performance, attrition, reporting, and other). The assessment of observational studies was conducted using the Quality Checklist for Healthcare Intervention Studies (Downs & Black, 1998). The Quality Checklist for Healthcare Intervention Studies comprises 27 questions covering five domains (study quality, external validity, study bias, confounding and selection bias and power). Any case reports, series or case studies were assessed using the IHE quality appraisal tool for case series studies (Guo et al., 2016; Moga et al., 2012). This quality appraisal tool is an 18-item questionnaire assessing the following: study objective, design, population, interventions, outcome measures, statistical analysis, results and conclusion, and competing interests and sources of support. Each article was assessed for risk of bias with the tools mentioned above by two reviewers working independently per study. Any disagreements were resolved through discussion between the reviewers.

Data analysis

A narrative synthesis of the findings from the selected studies was provided. The selected studies were described following their research design, sample population characteristics, exergaming intervention, timing of intervention delivery, setting and outcome measures.

Statistical analysis

A meta-analysis would have been conducted if the selected studies used the same type of intervention, sample population and reported similar outcome measures.

Results

Searches from eight electronic databases yielded a total of 3773 publications from which 236 were duplicates.

After excluding 3506 publications based on titles and abstracts, 31 publications were assessed in full-text, and 12 publications were included in the review (Fig. 1).

The 12 publications were published between 2010-2018 (2 in 2018, 3 in 2017, 1 in 2016, 2 in 2015, 1 in 2013, 2 in 2012 and 1 in 2010). The selected publications comprised 9 experimental intervention studies (2 RCTs (Imam et al., 2017; Prahm et al., 2018), single-subject study =1 (Prahm et al., 2017), case study = 2 (Ambron et al., 2018; Ortiz-Catalan et al., 2014), case reports = 3 (Chau et al., 2017; Miller et al., 2012; Sheehan et al., 2016) (Chau et al., 2017) and 4 feasibility studies (feasibility case series = 1 (Kruger, 2011), feasibility single-subject

study = 2 (Imam et al., 2013; Tousignant et al., 2015) and feasibility between-group study = 1 (Andrysek et al., 2012). The exergaming research was conducted in two different environments, respectively: research laboratories (4 university laboratories, 1 military medical laboratory) and clinical facilities (6 interventions were administered in hospitals, and one study also provided home-based rehabilitation interventions for the control group).

Study quality assessment

Two randomized controlled trials (Imam et al., 2017; Prahm et al., 2018) were included. When assessed for risk of bias using the Cochrane Collaboration's tool, no high risk of bias was detected for either study (Table I).

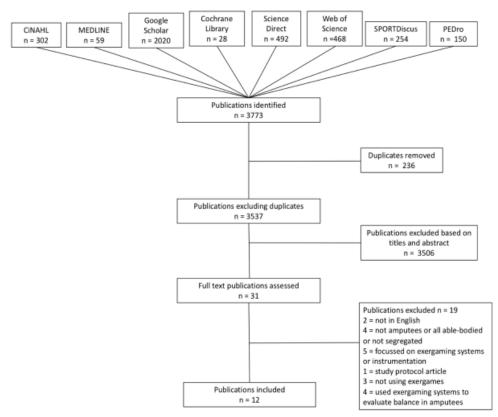


Fig. 1 – PRISMA flow diagram of the literature search strategy used.

Table I
Study quality assessment for controlled trials using Cochrane Collaboration's Tool.

	J 1 J	8		
Indicator	Study			
	Prahm et al. (2018)	Imam et al. (2017)		
Selection bias	Low - participants without limbs were randomised into two of three groups.	Low - participants without limbs were randomised into two groups.		
Allocation concealment	Unclear - randomisation concealment not specified.	Low risk randomisation concealment was specified.		
Performance bias	Unclear - blinding of participants not specified.	Unclear - blinding of participants not specified.		
Detection bias	Unclear - blinding was not specified.	Low - parallel evaluator - blind RCT.		
Attrition bias	Low - no missing outcome data.	Unclear - all outcome data was included despite 2 withdrawals and 1 lost to follow-up at T3.		
Reporting bias	Low - all outcomes reported in sufficient detail.	Low - primary and secondary outcomes were reported in sufficient detail.		
Other bias	Low - number of participants for a feasibility study.	Error in CONSORT flow diagram feasibility indicators were not sufficiently reported.		

Six publications (Ambron et al., 2018; Chau et al., 2017; Ortiz-Catalan et al., 2014; Sheehan et al., 2016) were assessed by the IHE quality appraisal tool for case series studies (Guo et al., 2016; Moga et al., 2012). These selected case reports, case series and case studies fulfilled

most of the IHE checklist criteria (Table II).

The remaining four feasibility studies were assessed using the Quality Checklist for Healthcare Intervention Studies (Table III).

Table II

Quality assessment for case studies, case reports and case series using the IHE quality appraisal checklist

(Gou et al., 2016; Moga et al., 2012).

Q	Ambron et al. (2018)	Chau et al. (2017)	Sheehan et al. (2016)	Ortiz-Catalan et al. (2014)	Miller et al. (2012)	Kruger (2011)
Q1	Y	Y	Y	Y	Y	Y
Q2	Y	Y	Y	Y	Y	N
Q3	U	U	N	U	U	U
Q4	U	U	U	U	U	U
Q5	Y	Y	Y	Y	Y	Y
$Q6^1$	Y	Y	Y	Y	Y	Y
Q7	Y	Y	Y	Y	Y	Y
Q8	Y	Y	Y	Y	Y	Y
Q9	U	U	N	N	Y	N
Q10	Y	Y	Y	Y	Y	Pr
Q11	U	U	U	U	U	U
Q12	U	U	Y	Y	Y	Y
Q13	Y	Y	Y	Y	Y	Y
Q14	U	Y	Y	U	U	U
Q15	U	Pr	Y	Y	N	N
Q16	NA	NA	NA	NA	NA	NA
Q17	N	Y	Y	U	N	N
Q18	N	N	N	Y	N	Pr
Q19	Y^{a}	Y	Y^{a}	Y	Y	Pra
Q20	Pr	Y	Y	Pr	Pr	Pr

Y yes, N no, U unclear, Pr partially reported, I description of eligibility for the study described as inclusion criteria, NA not applicable, at the Conclusion section is absent but conclusive arguments have been discussed in the article.

Table III

Quality assessment of observational studies using the Quality Checklist
for Healthcare Intervention Studies (Downs & Black, 1998).

Q	Prahm et al. (2017)	Tousignant et al. (2015)	Imam et al. (2013)	Andrysek et al. (2012)
Q1	Y	Y	Y	Y
Q2	Y	Y	Y	Y
Q3	Y	Y	Y	Y
Q4	Y	Y	Y	Y
Q5	N	Y	Y	Y
Q6	Y	Y	Y	Y
Q7	Y	Y	Y	Y
Q8	N	N	N	Y
Q9	Y	Y	Y	Y
Q10	Y	N	Y	N
Q11	UD	UD	UD	UD
Q12	UD	UD	UD	UD
Q13	UD	Y	Y	Y
Q14	N	N	N	N
Q15	UD	UD	UD	UD
Q16	Y	Y	Y	Y
Q17	Y	Y	UD	Y
Q18	Y	Y	Y	Y
Q19	Y	Y	Y	Y
Q20	Y	Y	Y	Y
Q21	UD	Y	Y	Y
Q22	UD	Y	UD	UD
Q23	Y	N	N	N
Q24	N	N	N	N
Q25	N	N	N	Y
Q26	Y	Y	Y	Y
Q27	F	C	D	F
SUM	19	20	20	24

Y yes, N no, UD unable to determine, C = size of smallest intervention group is 3-4, D = size of smallest intervention group is 5-6, F = size of smallest intervention group > 8.

They showed relatively good study quality with the lowest Downs and Black score being 19 (fair) and the highest being 24 (good), in consonance with scores previously reported (Hooper et al., 2008).

Study populations

Participants

The studies included in the review enrolled a total

number of 105 participants from Canada (79) (Andrysek et al., 2012; Imam et al., 2017, 2013; Prahm et al., 2018; Tousignant et al., 2015), USA (18) (Ambron et al., 2018; Chau et al., 2017; Kruger, 2011; Miller et al., 2012; Sheehan et al., 2016), Austria (7) (Prahm et al., 2017) and Sweden (1) (Ortiz-Catalan et al., 2014) (Table IV).

Table IV Participant characteristics and study details of included studies.

Author (date)	Study design	Population: n enrolled, n completed/relevant (gender), mean age (range), location	Amputation	Timing of intervention delivery post-surgery (n)	Location
Ambron et al. (2018)	Case-study	2, 2 (gender NS), age NS, USA	1 right transtibial, 1 left transtibial	11 months (1), 7 months (1)	University laboratory
Prahm et al. (2018)	Randomised controlled trial	14 amputees (gender NS), mean age NS, Canada -Control group = 10 (able-bodied) -Experimental (Group A, game- based rehabilitation) = 7 (transradial or transhumeral) -Experimental (Group B, Myoboy, standard rehabilitation tool) = 7 (transradial or transhumeral)	14 transradial or transhumeral	NS	Hospital
Prahm et al. (2017)	Single-subject study	7, 7 (gender NS), age NS, Austria	7 transradial or transhumeral	NS	University laboratory
Imam et al. (2017)	Randomised controlled trial	28, 23 (gender NS), 62 ^{mdn} (50-78), Canada -Control group = 14 (2 withdrawals before commencing) -Experimental = 14 (2 withdrawals before commencing, 1 drop-out)	Unilateral transtibial or transfemoral	Within 12 months (28)	Hospital
Chau et al. (2017)	Case report	1, 1 (1 male), 49, USA	1 right wrist disarticulation	5 months (1)	University laboratory
Sheehan et al. (2016)	Case report	1, 1 (1 male), 43, USA	1 right transfemoral	7 years (1)	Military medical laboratory
Ortiz-Catalan et al. (2014)	Case study	1, 1 (male), 72, Sweden	1 transradial	48 years (1)	Hospital
Tousignant et al. (2015)	Feasibility pilot single-subject study	3, 3 (gender NS), mean age NS, Canada	2 left transtibial, 1 left transfemoral	NS	Hospital
Imam et al. (2013)	Feasibility single- subject case study	8, 6 (5 male), 48.5 ^m (45 - 59), Canada	4 transtibial, 2 transfemoral	≤ 12 months (6)	Hospital
Miller et al. (2012)	Case report	2, 2 (2 male), 60 (NS), USA	2 transfemoral	32 months, 9 years	University laboratory
Andrysek et al. (2012)	Feasibility pilot between-group study	16 children and adolescents; control group (age-matched, non-amputee) = 10 (5 male), 10.7 (range NS) (mean age NS), experimental group = 6 (2 male); 11.8 (8 - 18), Canada	3 transfemoral, 3 Van Ness	Within 36 months (6)	Hospital (experimental group), participants' homes (control group)
Kruger (2011)	Feasibility case series	11, 11 (11 male), 28.9 (range NS), USA	2 right transtibial, 4 left transtibial, 1 right transfemoral, 1 left transtibial with right midfoot, 1 left transtibial with right knee disarticulation, 2 bilateral transfemoral	NS	Military medical laboratory

NS Not specified, mdn median age, m mean

Table V Data summary of studies using exergames.

				Data summary of studies using exergames.
Author (Date)	System	Game	Intervention	Outcome measures (Method of assessment (significant findings))
Ambron et al. (2018)	Computerised video games	Quest for Fire, Web Browser, Chess and Checkers	2 sessions for the first individual, 4 sessions over six weeks for the second individual.	Pain intensity (VAS), usability (System Usability Scale)
Prahm et al. (2018)	Computerised video games via muscle control	2-dimensional labyrinth, racing game and rhythm game	Single session.	Muscle activity via EMG signals: maximum voluntary contraction (increased, $p=.02$ in groups and C), proportional precision control (in groups A and C, $p<.01$ for all target intensities, $p<.01$ for reaching low goal activation) in Group B, electrode separation (less activation in opposing electrode in all groups, $p01$ 46; groups A and C, decrease in involuntary activation of opposing electrode for low intensity levels, $p=.02$ 04) and muscle endurance (correlation between groups A and C, $p<.01$) User experience (IMI: enjoyment of games more than Myoboy ($p<.01$) User evaluation of game (custom questionnaire).
Prahm et al. (2017)	Computerised video games via muscle control	Pospos, Super Tux Kart, Step Mania 5	Single session.	-Muscle activity via EMG signals (precision control (p < .01), (electrode separation (significantly less activated during low goal activation levels), (endurance control (p< .01)). -User experience from playing the games (IMI), using the MyoBoy (IMI (lower enjoyment (p = .02)), enjoyment of EMG assessments (custom questionnaire) -User evaluation of game (custom questionnaire).
Imam et al. (2017)	Nintendo WiiFit™	- Experimental: Wii.n. Walk (WBB yoga, balance games, strength training and aerobics games) - Control: Wii Big Brain Academy TM	3 sessions/week for 4 weeks, 40 min/session	Walking capacity (2MWT), functional tasks (SPPB), physical activity (PASE), balance confidence (ABC), step activity (SAM), cognitive-motor (WWT), locomotor activity (LCI-5), feasibility parameters.
Chau et al. (2017)	Computerised video games	Interactive kitchen, Autoshield, Eleven: Table Tennis	5 sessions over several weeks, 45 min/session	Pain (VAS (55% decrease (p = .0143)), pain (SF-MPQ (60% decrease (p = .023)), pain (<i>WB FACES</i> (90% decrease (p = .0024))
Sheehan et al. (2016)	CAREN	Treadmill with virtual terrains (using randomised perturbations) - walking outdoors, hiking and playing golf	2 sessions/week for 4 weeks, 30 min/session	Walking speed (increase), improvement in functional stepping times, step width variability, stepping stability and margin of stability.
Ortiz- Catalan. (2014)	Computerised video games	Racing game (Trackmania Nations Forever)	Once a week for 13 weeks, followed by twice a week for 5 weeks.	Pain perception (McGill pain questionnaire), pain intensity (VAS). EMG signals: wrist pro/supination and elbow flexion/extension. Motion test: physical movements (Custom questionnaire).
Tousignant et al. (2015)	Nintendo Wii	WBB and WFg	5 sessions per week for 8 weeks	Walking (L test), Function and balance (AMPPRO), satisfaction (HCSQ), motivation (VAS), quality of life (TAPES).
Imam et al. (2013)	Nintendo WiiFit TM	Yoga, balance games, strength training and aerobics games	5 sessions/week for 2 weeks (10 sessions) and a maximum of 6 weeks (30 sessions), 30 min/session	Walking capacity (2MWT (increase ^{2SD} in 5 patients)), functional tasks (SPPB (improvement in 4 patients)), a functional mobility (L test (improvement in 2 patients)), balance confidence (ABC (improvement ^{2SD} in 3 patients)), pain and fatigue (NRS), acceptability (SFQ-M).
Miller et al. (2012)	Nintendo WiiFit TM and BWS gait training	Wii games and aerobic balance training	2 sessions/week for 6 weeks, 20 min/session each of WiiFit TM and gait training	OUES, movement, dynamic balance, ABC, gait (GAlTRite)
Andrysek et al. (2012)	Nintendo WiiFit™	Table Tilt and Tightrope Walk	4 sessions/week for 4 weeks, 20 min/session	Dynamic balance (COP), function and mobility (CB&M), feasibility (custom questionnaire and user logbook).
Kruger (2011)	CAREN	Continuous road, road with overhead targets	1 or 2 sessions/week for 4 to 8 weeks, 30 min/session	Walking speed (increase in self-selected velocities).

^{2SD} 2 standard deviation band method of statistical significance, *2MWT* 2 Minute Walk Test, *SPPB* Short Physical Performance Battery, ABC Activities-Specific Balance Confidence scale, *AMPPRO* Amputee Mobility Predictor, *BWS* body weight support, *CAREN* Computer Assisted Rehabilitation Environment, *COP* centre of pressure, *CB&M* Community Balance and Mobility scale, *EMG* electromyography, *HCSQ* Health Care Satisfaction Questionnaire, *IMI* Intrinsic Motivation Inventory, *L* L-test of functional mobility, NRS numeric rating scale, ML mediolateral, *MyoBoy* assessment and training system, *OUES* oxygen uptake efficiency slope, *PASE* Physical Activity Scale for the Elderly, *LCI-5* Locomotor Capability Index in Amputees, SAM Modus Health Stepwatch™ Activity Monitor, *SF-MPQ* Short-form McGill Pain Questionnaire, SFQ-M Short Feedback Questionnaire-modified, *TAPES* Trinity Amputation and Prosthesis Experience Scale, VAS visual analogue scale, *WBB* Wii Balance Board, *WB FACES* Wong-Baker Faces pain scores, *WFg* Wii Fit game.

Out of these, 88 were amputees and 17 were ablebodied. The participant population comprised individuals presenting either one extremity amputation or double amputation. One extremity amputees included: 55 lower limb amputees (14 transtibial, 28 transtibial or transfemoral, 10 transfemoral, 3 Van Ness) and 23 upper limb amputees (21 transradial or transhumeral, 1 wrist, 1 transradial). Double amputees included 4 individuals presenting: 1 left transtibial with right midfoot, 1 left transtibial with right knee disarticulation, 2 bilateral transfemoral. With regard to attrition, 1 dropout (at follow-up after pre- and post-testing) and 4 withdrawals were reported across these studies (Imam et al., 2017). A dropout refers to a participant who voluntarily withdraws his participation from a study, whereas a withdrawal refers to a well-weighed decision by research administrators to terminate participation of an individual, respectively. The reviewed studies included 16 children and adolescents, and 89 adults, within the age range of 8-78 years. The gender distribution was 28 male, 8 female and 70 non-specified. Only one study recorded participants' level of education (high school 32%, college 42.9%, university 25%), employment status (32% employed) and cognitive functioning (mean 29 scored from MMSE, range 23-30). In addition, they also recorded socket comfort for their participants (8 median score, range 4-10) (Imam et al., 2017).

Study interventions

Five of the included interventions used computerized video games for their exergaming intervention (Ambron et al., 2018; Chau et al., 2017; Ortiz-Catalan et al., 2014; Prahm et al., 2018; Prahm et al., 2017) (Table V).

The Nintendo Wii was used by five studies (Andrysek et al., 2012; Imam et al., 2017; Imam et al., 2013; Miller et al., 2012; Tousignant et al., 2015) and the remaining two studies used CAREN (Kruger, 2011; Sheehan et al., 2016). The reported duration ranged from 20 to 45 minutes per session. Not all durations were reported as sessions depending on each individual's adherence and motivation to persist. The duration of interventions ranged from one day to 8 weeks. Two studies included a comparison group of able-bodied individuals (Andrysek et al., 2012; Prahm et al., 2018), whereas one also included a comparison group of amputees (Imam et al., 2017). Andrysek et al. (Andrysek et al., 2012) presented the only study to use home-based exergaming for the experimental group (children with amputations). The study by Imam et al. (2017) used the Nintendo WiiFitTM and Wii Big Brain AcademyTM, played with a handheld remote control. Their exergaming intervention was designed to receive training at the hospital before undertaking unsupervised homebased exergaming. Prahm et al. (2018) used computerized video games played by muscle control and the Myoboy, a standard rehabilitation tool designed for muscle activity and prosthetic training. Collectively, intervention delivery occurred within 5 months to 48 years post-surgery. Four studies took place within twelve months post-surgery (Ambron et al., 2018; Chau et al., 2017; Imam et al., 2017; Imam et al. 2013). Andrysek et al. (2018) carried out their study within 36 months post-surgery. In the study by Ortiz-Catalan et al. (2014), their participant took part in the exergaming intervention 48 years post-surgery. Betweengroup comparisons were carried out in three studies (Prahm et al., 2018; Imam et al., 2017; Andrysek et al., 2017). Seven of the interventions were delivered by research staff (Ambron et al., 2018; Prahm et al., 2018; 2017; Imam et al., 2017; Chau et al., 2017; Ortiz-Catalan, 2014; Miller et al., 2012), three were delivered by physiotherapists (Sheehan et al., 2016; Kruger et al., 2011; Imam et al., 2013) and one study employed a physiotherapist and occupational therapist (Tousignant et al., 2014).

Outcome measures from exergaming interventions

Exergaming interventions were used to assess the following measures: pain (Ambron et al., 2018; Chau et al., 2017; Imam et al., 2013; Ortiz-Catalan et al., 2014), fatigue (Imam et al., 2013), physical functioning (Imam et al., 2017; Imam et al., 2013; Kruger, 2011; Sheehan et al., 2016; Tousignant et al., 2015), muscle control (Prahm et al. 2018; Prahm et al. 2017), feasibility (Andrysek et al., 2012; Imam et al., 2017), acceptability (Imam et al., 2013), quality of life (Tousignant et al., 2015) and user experience (Prahm et al., 2018; Prahm et al., 2017).

Pain and fatigue

The Visual Analogue Scale (VAS) was used to assess pain (Ambron et al., 2018; Chau et al., 2017; Ortiz-Catalan et al., 2014). Imam et al. (2013) used the Numerical Rating Scale (NRS), whereas Chau et al. (2017) used three pain rating scales (the Visual Analogue Scale (VAS), the shortform McGill Pain Questionnaire (SF-MPQ), and Wong-Baker FACES pain rating scale) to assess pain before and after the exergaming intervention. One study recorded fatigue scores by using a Short Feedback Questionnaire (SFQ-M) (Imam et al., 2013).

Physical functioning and mobility

The assessed outcomes were walking and step activity using the following: the 2 Minute Walk Test (2MWT) (Imam et al., 2017; Imam et al., 2013), L test (Imam et al., 2013; Tousignant et al., 2015) and computerized treadmill in combination with the Vicon motion capture system (Kruger, 2011; Sheehan et al., 2016). Imam et al. (2017) assessed the number of steps taken each day for a week using the Modus Health StepwatchTM Activity Monitor (SAM), mounted on the prosthetic ankle. They also assessed self-reported physical activity by using the Physical Activity Scale for the Elderly (PASE). Cognitivemotor interaction was assessed using the Walking While Talking Test (WWT) (Imam et al., 2017), and locomotor activity was assessed using the Locomotor Capabilities Index in Amputees (LCI-5) (Imam et al., 2017). Tousignant et al. (2015) assessed functional mobility with a prosthesis using the Amputee Mobility Predictor (AMPPRO) questionnaire. Outcome measures for muscle control were levels of EMG control, fine muscle activation and electrode separation assessed by using recorded electromyographic (EMG) biofeedback via myoelectric signals (Prahm et al., 2018; Prahm et al., 2017). Miller et al. (2012) was the only study to assess aerobic capacity whilst walking in older people with amputations.

The assessed outcomes for balance were balance confidence using a self-administered subjective questionnaire called the Activities-Specific Balance Confidence (ABC) scale (Imam et al., 2017; Imam et al., 2013; Miller et al., 2012), centre of pressure (COP)

displacements during quiet standing using the Nintendo Wii balance board (Andrysek et al., 2012), dynamic balance using the Biodex system (Miller et al., 2012) and functional balance using the Community Balance and Mobility Scale (CB&M) (Andrysek et al., 2012).

Feasibility and acceptability

Feasibility of the exergaming intervention was assessed using a customized questionnaire and a recorded logbook (Andrysek et al., 2012), whereas another study collectively assessed feasibility by considering outcome measures of safety and report of any adverse events from the exergaming intervention, post-intervention fatigue, pain levels, adherence and user acceptability of the exergaming intervention (Imam et al., 2013). User evaluation and acceptability of the games were assessed using a custommade questionnaire (Prahm et al., 2017), System Usability Scale (Prahm et al., 2017) and the Short Feedback Questionnaire-modified (SFQ-M) (Imam et al., 2013).

Quality of life, motivation and user evaluation

One study assessed quality of life amongst amputees using the Trinity Amputation and Prosthesis Experience Scales (TAPES) (Tousignant et al., 2015). Motivation was assessed by using the Intrinsic Motivation Inventory (IMI) questionnaire (Prahm et al., 2018; Prahm et al., 2017), while another study used a custom made questionnaire to evaluate motivation by rating on a Visual Analogue Scale (VAS) and assessed patient satisfaction with health care services using the Health Care Satisfaction Questionnaire (HCSQ) (Tousignant et al., 2015).

Effects of the intervention

Two randomised controlled trials were included in this review. Imam et al. (2017) tested the effects of exergaming on walking capacity using the Nintento WiifFitTM for 12 sessions (over 4 weeks) compared with cognitive games using the Big Brain Academy DegreeTM in older people with missing limbs. Their clinical outcome results were based on intention to treat analyses. Although there were no significant changes in the other outcomes, their results on walking capacity at post-intervention and 3-week retention were comparable to those of an RCT with younger individuals (Rau et al., 2007). Improvements were observed in walking capacity and cognitive-motor tasks in favour of the exergaming intervention (Wii.n.Walk). The overall adherence to the exergaming intervention was high, although in-home adherence was slightly lower than inclinic adherence. Their patients preferred supervised group training and welcomed the convenience and accessibility of home-based exergaming.

Prahm et al. (2018) assessed short-term effects of exergaming on EMG muscle control in two patient groups and one control group comprising able-bodied participants. One of the patient groups served as a control, performing random EMG activations, whereas the experimental and able-bodied group played exergames (computerized video games). They found significantly increased maximum voluntary contractions in the groups that played the exergames, indicating stronger muscle contraction and improved muscle control. Improved proportional precision control was also observed in these groups for all EMG target intensities. The patient control group, however, showed significant improvement for the middle intensity

target. Although there was overall improvement in muscle separation in almost every instance, these results were not always significant. Only the groups that played exergames showed significant decreases of involuntary activation of the opposing electrode for the first to third measurements for low goal intensity levels. Improved endurance and muscle isolation was also found in favour of exergaming. Their patients significantly enjoyed playing the exergames and perceived the MyoBoy to be a useful EMG training tool. In terms of exergame evaluation, they preferred rhythm and racing games. Racing games scored slightly higher motivational scores.

Three of the twelve studies evaluated whether pain improved after an exergaming intervention (Ambron et al., 2018; Chau et al., 2017; Ortiz-Catalan et al., 2014). All three reported reductions in pain intensity. Ambron et al. (2018) found lower pain intensity ratings at post-intervention, but were not able to establish the association between pain and level of fatigue. The patient in the study of Chau et al. (2017) reported significant pain relief taking effect approximately 24 hours after each exergaming session. There was also a decrease in pain, lasting progressively longer for several days after each exergaming session. Follow-up feedback on pain one week post-intervention reported continued pain relief over five days after the last exergaming session and an overall decrease in baseline pain levels. At six weeks follow-up, the patient reported that the pain was still present but generally decreased in severity and was much more tolerable. This indicates longer lasting benefits retained after exergaming. The results of Ortiz-Catalan et al. (2014) were especially interesting where the patient experienced an increment of pain at the beginning of the exergaming intervention, followed by reduced pain intensity after 4 weeks and pain-free periods after 10 weeks, which then developed into completely pain-free periods a couple of sessions later. Although pain was not their primary clinical outcome, Imam et al. (2013) reported post-intervention pain and fatigue scores which ranged less than 6 on a scale of 0 to 10 (0 = no pain; 10 = extremepain and 0 = no fatigue, 10 = extreme fatigue). They also reported high adherence (80%) to their reported median scores for pain and fatigue, suggesting beneficial effects on phantom limb pain from exergaming in amputees.

The studies that used CAREN to evaluate clinical outcomes found improvements in walking, gait, physical functioning and balance, including progression of level walking to more challenging terrain (Kruger, 2011; Sheehan et al., 2016). One of the studies demonstrated evidence of retaining benefits in gait at least 5 weeks after the final exergaming session (Sheehan et al., 2016). The other reviewed studies found improvements favouring the exergaming group in some of the outcomes assessed, such as better muscle control (Prahm et al., 2017), dynamic balance (Miller et al., 2012) and balance confidence (Imam et al., 2017; Imam et al., 2013; Miller et al., 2012). One study (Andrysek et al., 2012) showed differences in functional balance and mobility between patient groups, where patients with transfemoral amputations scored lower than those of the Van Ness group despite overall improvement in functional balance and mobility (CB&M) scores between baseline, at post-intervention and followup. Another study found high levels of motivation after exergaming amongst patients (Tousignant et al., 2015). Study participants demonstrated positive responses in terms of acceptability of exergaming (Imam et al., 2013).

Discussions

This is the first systematic review to evaluate and summarize current literature concerning the effects of exergaming on individuals with missing limbs. The interventions we found through this review showed variability from one another in terms of clinical and methodological diversity. Hence, it is difficult to conclude which method of delivery would prove to be the most advantageous.

Exergaming interventions in the reviewed studies had different therapeutic targets and varied in terms of participants, duration, gaming design and strategies, whether it was to improve balance and stability responses through repeated practice (Sheehan et al., 2016), to provide treatment for phantom limb pain (Ambron et al., 2018) or to improve muscle control (Prahm et al., 2018). The Nintendo WiiFitTM was the most used intervention in studies involving people with lower extremity amputations, whereas computerized video games were used in the studies involving people with upper extremity amputations. Only one study using computerized video games involved two individuals with lower extremity amputations, whereby one patient (with left transtibial amputation) reported reduced pain severity after exergaming and a progressive decrease in phantom limb pain across the exergaming sessions (Ambron et al., 2018). This suggests the suitability of exergaming interventions across different types of amputations in individuals.

In terms of clinical benefit, exergaming was seen to improve mobility and balance (Andrysek et al., 2012; Imam et al., 2017; Imam et al., 2013; Kruger, 2011; Miller et al., 2012; Sheehan et al., 2016; Tousignant et al., 2015) when assessed through the current review, showing alignment with previous exergaming studies involving able-bodied clinical groups (Hung et al., 2014; Karahan et al., 2016; Robinson et al., 2015). The studies that used CAREN found improved outcomes in their participants individually, particularly in walking and balance (Kruger, 2011; Sheehan et al., 2016).

Pain was assessed in three studies in this review (Ambron et al., 2018; Chau et al., 2017; Ortiz-Catalan et al., 2014), showing improvements following the exergaming intervention. These findings are consistent with those of Pekyavas & Ergun (2017), who compared the Wii with a home exercise programme provided to patients with subacromial impingement syndrome (SAIS). They found that the exergaming group demonstrated significantly better improvements in range of movement in the shoulder and scapular rotation and retraction compared to the home exercise group, despite improvements in pain in both groups after exergaming.

The current review was unable to find strong evidence of long-term benefits from exergaming. However, from the studies assessed, exergaming interventions appear to confer at least short-term benefits to people with amputations, where one study demonstrated evidence in the retainment of improved gait at least five weeks after the last exergaming session (Sheehan et al., 2016). This is similar to a study by Sims et al. (2013), which evaluated the effects of exergaming on static postural control in able-bodied people with a history of lower limb injury. In addition to improved static postural control after exergaming, they found significant improvement in self-reported function at four weeks post-intervention.

Patient motivation and adherence to rehabilitation encourage recovery and improved health outcomes in patients (Maclean & Pound, 2000). Findings from the studies reviewed showed increased motivation amongst patients after exergaming (Imam et al., 2017; Prahm et al., 2018; Prahm et al., 2017). For instance, rhythm and racing games were perceived to be more enjoyable than dexterity games, and motivation scores were rated higher in racing games when compared to rhythm games (Imam et al., 2017). The single participant in the study of Sheehan et al. (2016) attributed the benefits to exergaming; he believed that interacting with the exergaming intervention had challenged him to focus on the surroundings and to make necessary gait and posture changes in order to play the exergames. The participants in the study of Miller et al. (2012) found exergaming to be challenging and enjoyable. Participants in the study by Tousignant et al. (2015) demonstrated high motivation and adherence to the exergaming intervention and were satisfied with the service provided. They also scored highly on the Health Care Satisfaction Questionnaire (97%, 100% and 84%, respectively).

Because the included studies showed wide heterogeneity, it is difficult to draw firm conclusions in the delivery of interventions and the clinical outcome measures assessed within the selected studies. Nevertheless, exergaming interventions appear to be feasible and favourably received by individuals with missing limbs. A high degree of adherence and a low level of dropouts from the reviewed studies indicated high acceptance regarding the proposed exergaming interventions, including both immersive and non-immersive virtual reality designs. Of all the included studies, there was one which had the only lost to follow-up participant (Imam et al., 2017). The participant developed complications with preexisting lung disease, unrelated to the study. In spite of this, adherence to the study was 83.4% (Imam et al., 2017).

With regard to feasibility, the exergaming sessions were well accepted and received positive feedback from participants (Chau et al., 2017). Participants in the study of Andrysek et al. (2102) perceived the exergames to be fun and easy to play. Furthermore, participants in Prahm et al.'s study (2018) significantly enjoyed exergaming, even though the required physical movements put more pressure on them. Participants in the study by Imam et al. (2017) were willing to exert more physical effort to play the exergames in comparison to using the MyoBoy. They perceived the exergaming intervention to be useful for improving their walking abilities and intended to continue using the equipment at home on a regular basis (Imam et al., 2017). Usability of exergaming interventions also received favourable ratings in the study of Ambron et al. (2018), where the majority of ratings via the System Usability Scale

questionnaire fell within the acceptable range of above 50 out of 100, where scores of 70+ mean good prospective usability for an information technology-based application in development (Sauro, 2011). Nevertheless, there was also report of low ratings in usability for one of the exergames called Quest for Fire by one participant, reflecting the frustration encountered whilst learning to move the avatar around the labyrinth (Ambron et al., 2018). With regard to safety, 4 near-fall incidents while exergaming with lower limb prostheses were recorded by Andrysek et al. (2012). Nonetheless, there was no report of adverse effects related to the exergaming interventions.

With respect to quality of life, one of the domains of life classified by the International Classification of Functioning, Disability and Health (ICF) is mobility (2). In fact, the benefits of exergaming derived from the variety of exergames and complex challenges presented to the user are not limited to improved functional parameters, but instead, also encompass domains directly influencing the quality of life of the participants. For instance, reducing pain at phantom limb level, improving prosthesis control, self-confidence and outdoor environment ambulation management, as related by participants in the reviewed studies (Ambron et al., 2018; Sheehan et al., 2016). One participant in the study of Ambron et al. (2018) reported dramatic improvements in his physical activity over the course of the exergaming intervention. After two exergaming sessions, he successfully walked to the local grocery store using a lower-limb prosthesis for the first time after amputation. The participant attributed his improved physical activity to exergaming training. Feedback from the participant in Chau et al.'s study (2017) was also promising. He stated that playing the exergames made him forget the pain, move as if the pain was not there and he felt normal. His remark "I feel like my hand is back" is an especially important response to exergaming as this reflects the potential therapeutic benefit from exergaming on physical recovery and movements on a residual limb.

The current review is not without limitations. The selected studies showed great heterogeneity. The study protocols differed in terms of exergaming intervention and length of therapy sessions. Furthermore, the exergaming interventions from the reviewed studies differed in frequency, duration, gaming elements, and physical and cognitive user tasks. The actual power of the studies is also limited by the low number of participants enrolled. Outcome measures also differed in assessment methods. Due to the scarcity of literature for exergaming in people with amputations, more research should be conducted to explore common clinical outcomes from exergaming interventions, suitable for individuals with different types of amputations. Future research should also assess longer follow-ups post-intervention in order to evaluate the effects of exergaming over time.

Conclusions

Upon completion of the current review, we concluded that:

1. There was a wide variability in the studies assessed. Due to the heterogeneity of the included studies, we were unable to conclude about the effectiveness of exergaming.

2. However, there was evidence of improved health outcomes after exergaming, feasibility and acceptance of the exergaming interventions to suggest that exergaming may be potentially therapeutic for people with missing limbs.

Conflicts of interests

The authors have none to declare.

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