

# Multimodal sensory feedback in augmented reality for gait rehabilitation in children with cerebral palsy

Retours sensoriels multimodaux en réalité augmentée pour la rééducation de la marche des enfants atteints de paralysie cérébrale

## Thèse de doctorat de l'Université Paris-Saclay

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C'est pour savoir où je vais que je marche.

Goe the

A toi, qui aurait fait chanter Yuki.



# Abstract

The global aim of our work is to improve gait rehabilitation for children with cerebral palsy after single-event multi-level surgery of the lower limbs. Cerebral palsy describes a set of movement and posture disorders causing activity limitations and participation restrictions. It is attributable to a non-progressive brain lesion during fetal development or the first year of life. During childhood, musculoskeletal growth inequalities lead to gait abnormalities that severely impair the quality of life of these children. During adolescence, a surgical intervention allows restoring these bone/tendon/muscle deformities in the same operating time. First, we conducted a literature review on postoperative rehabilitation because there was a lack of standardized presentation (Article 1). We highlighted that this rehabilitation could be divided into five key steps. In the fourth step, intensified gait training is recommended to optimize postoperative walking capacity. The question of how to maintain the motivation of young people during such a long period of rehabilitation and how to provide effective gait rehabilitation was raised.

To this end, we have chosen to investigate the potential of active video games. Over the last decade, interest in active video games used in motor rehabilitation has continued to grow. The literature reports that not all games used with therapeutic purposes clinically tested are effective, far from it. Today there is a vast mixture, sources of misunderstandings, particularly the use of commercial games diverted to be used in therapy. These are not always adapted to children with motor disabilities and are especially not developed considering the essential ingredients of motor learning and motor rehabilitation. Moreover, Active Video Games can be displayed using different devices, from virtual reality to augmented reality. This choice is crucial.

In our context, augmented reality has been preferred to deliver our active video game so that children feel free to walk in their environment using their usual walking aids without restriction of movement. Augmented reality is a technology that combines virtual elements generated by a computer to modify or augment the real environment. These virtual objects are supplemented by multimodal perceptual information, notably visual, auditory, or haptic. Thus, the individual perceives and interacts with a virtual object superimposed on his real environment in real-time.

The field of application is extensive, ranging from the industrial context to optimize the production to the surgical area by facilitating access to medical information for the surgeon. In this thesis, we aimed to foster a novel field of research and use for augmented reality, motor rehabilitation and particularly gait rehabilitation in pediatric neurology.

Indeed, in the area of gait rehabilitation, there is currently no research, fundamental or clinical, on the use of active video games in augmented reality. New approaches on health technolo-

gies are encouraged to support health professionals, provide patients with personalized and efficient healthcare and information, and stimulate health promotion and disease prevention in the general population. These technologies could bring some fun into rehabilitation care, which is essential for kids. Their use in quasi-autonomy would also allow therapists to offer more rehabilitation time.

The tool used to deliver our active video game was the Microsoft Hololens® augmented reality headset. The entire research team validated this choice according to precise specifications. We conducted two studies. First, we assessed the reliability of the headset tracking in comparison with a reference motion analysis tracking system (Article 2). Second, we described and validated a new algorithm, HoloStep, calculating in real-time gait spatiotemporal parameters with the only position of the Hololens (Article 3). In parallel to these technological considerations, we focused our fundamental research on the impact of feedback delivered in augmented reality. Because feedback in AR can be displayed to the user through different characteristics, we have developed a theoretical model of feedback in augmented reality for rehabilitation. This theoretical work was necessary before designing feedback and testing specific characteristics of them in healthy adults (Article 4) and children with CP (Article 5 & Article 6). Some of the observations from this work have led to new research questions, notably the characterization of patients as "non-responders" to feedback. Finally, we analyzed the literature to create the most adapted gait rehabilitation protocol, using good ingredients to match motor learning principles. The active video game ARRoW-CP for young people was developed, including a 4-week rehabilitation protocol consisting of a series of high-intensity walking sprints (Article 7). Today, we evaluate the ARRoW-CP active video game with young people with Cerebral Palsy at the Ellen Poidatz Rehabilitation Center.

This thesis results from a cross between several scientific areas sharing the ambition to be part of "tomorrow's medicine", which is participatory, predictive, preventive, customizable, and evidence-based. Thus, we have contributed to rehabilitation sciences by developing, in collaboration with stakeholders, an active video game for gait rehabilitation. We have investigated more fundamental notions in motor learning, notably on the impact of feedback. Finally, we have explored the field of information and communication sciences and technologies by developing tools and content usable in augmented reality.

**Keywords:** Augmented Reality, Feedback, Gait Rehabilitation, Cerebral Palsy, Active Video Game, Motor Learning, Single-Event Multi-Level Surgery

# Résumé

Notre recherche a pour objectif général de contribuer à l'amélioration de la prise en charge rééducative des enfants atteints de paralysie cérébrale après intervention chirurgicale des membres inférieurs. Cette pathologie regroupe un ensemble de troubles du mouvement et de la posture responsable de limitations d'activité et de restriction de participation. Elle est imputable à une lésion cérébrale non évolutive ayant eu lieu au cours du développement fœtal ou durant la première année de vie de l'enfant. Durant l'enfance, les inégalités de croissance musculo-squelettiques engendrent des anomalies de la marche altérant fortement la qualité de vie de ces jeunes. Une intervention chirurgicale pratiquée à l'adolescence permet, dans le même temps opératoire, de rétablir ces dés-équilibres biomécaniques. Notre premier travail a été de réaliser une revue de la littérature sur la rééducation qui suit cette intervention chirurgicale, qui n'était jusqu'alors pas standardisée (Article 1). Nous avons mis en évidence que cette rééducation pouvait être décomposée en 5 étapes clés. Au cours de la quatrième étape, l'intensification de l'entraînement de la marche est recommandée comme une stratégie d'optimisation de la capacité de marche post-opératoire. La question du maintien de la motivation des jeunes au cours d'une si longue période de rééducation, ainsi que celle du moyen utilisé pour proposer une rééducation de la marche efficace, se sont posées.

A cette fin, nous avons choisi d'investiguer le potentiel des jeux vidéos actifs. En effet, au cours de la dernière décennie, l'intérêt pour les jeux vidéo actifs utilisés dans la rééducation motrice n'a cessé de croître. Cependant, la littérature rapporte que tous les jeux utilisés à des fins thérapeutiques testés cliniquement ne sont pas efficaces, loin de là. Il existe aujourd'hui un vaste mélange, source de confusions, avec notamment l'utilisation de jeux commerciaux détournés pour être utilisés en thérapie. Ces derniers ne sont pas toujours adaptés aux enfants atteints de handicap moteur, et surtout ils ne sont pas développés en tenant compte des ingrédients essentiels de l'apprentissage moteur et de la rééducation motrice. De plus, le jeu vidéo actif peut être délivré à l'aide de différents dispositifs technologiques, qui vont de systèmes de réalité virtuelle à la réalité augmentée. Ce choix est crucial.

Dans notre contexte, la réalité augmentée a été préférée comme support de notre jeu vidéo actif afin que les enfants puissent marcher dans leur environnement réel, sans restriction de mouvement, en utilisant leurs aides de marche habituelles. La réalité augmentée est une technologie associant des éléments virtuels, générés par ordinateur, pour modifier ou augmenter l'environnement réel. Ces objets virtuels sont enrichis par des informations perceptives multimodales, notamment visuelles et/ou auditives. Ainsi, l'individu perçoit une scène virtuelle qui se surimpose à son environnement réel et dans laquelle il peut interagir en temps réel. Les domaines d'application de cette technologie sont étendus. Nous avons souhaité ouvrir le champ de recherche et d'utilisation de la réalité augmentée au domaine de la rééducation motrice, et plus spécifiquement de la rééducation de la marche en neuropédiatrie. En effet, pour la rééducation de la marche, il n'existe à ce jour aucune recherche, fondamentale ou clinique, portant sur l'utilisation des jeux vidéo actifs en réalité augmentée. De nouvelles approches sont nécessaires afin de permettre aux patients de se rééduquer de manière ludique et efficace mais également de fournir aux thérapeutes une solution numérique autonome pour augmenter le temps de rééducation à proposer à leur patient.

L'outil utilisé pour délivrer notre jeu vidéo actif a été le casque de réalité augmentée Microsoft Hololens<sup>®</sup>. Ce choix a été validé par l'ensemble de l'équipe de recherche en fonction d'un cahier des charges précis. Nous avons mené deux études afin de confirmer la fiabilité du calcul de la position tridimensionnelle du casque dans l'espace (Article 2) et de valider la précision de l'algorithme de détection des paramètres spatio-temporels que nous avons développé et implémenté dans le casque Hololens (Article 3). En parallèle de ce versant technologique, nous avons orienté notre recherche fondamentale sur les retours sensoriels (ou feedback) en élaborant un modèle théorique des caractéristiques des feedbacks délivrés en réalité augmentée applicable dans le cadre de la rééducation. Nous avons ensuite testé auprès d'une population adulte (Article 4) et d'une population d'enfants atteints de PC des scenarii combinant différentes caractéristiques de feedbacks visuels (Article 5 & Article 6). Certaines observations issues de ces travaux ont conduit à de nouvelles questions de recherche, notamment celle de la caractérisation des profils de patients « non-répondants » aux feedbacks. Enfin, une réflexion pluridisciplinaire comprenant une analyse de la littérature a été menée afin d'élaborer le protocole de rééducation de la marche le plus adapté, notamment en termes d'intensité et de répétition, pour satisfaire les théories de l'apprentissage moteur. Sur la base de ces travaux, le jeu vidéo actif ARRoW-CP destiné aux enfants atteints de paralysie cérébrale a été développé, incluant un protocole de rééducation de 4 semaines composé de séries de sprints de marche à haute intensité (Article 7). Aujourd'hui, notre jeu vidéo actif ARRoW-CP est en cours d'évaluation clinique auprès des jeunes atteints de paralysie cérébrale au sein du Centre de Rééducation Ellen Poidatz.

Cette thèse est le fruit d'un croisement entre plusieurs disciplines scientifiques partageant l'ambition de s'inscrire dans la médecine de demain, qui se veut participative, prédictive, préventive, personnalisable, et basée sur les preuves. Ainsi, nous avons contribué aux sciences de la rééducation en développant en collaboration avec les parties prenantes un jeu vidéo actif de rééducation de la marche, nous avons investigué des notions plus fondamentales en apprentissage moteur, notamment sur l'importance des feedbacks et nous avons exploré le champ des sciences et technologies de l'information et de la communication par le développement d'outils et de contenus utilisables en réalité augmentée.

**Mots-clés :** Réalité Augmentée, Retours Sensoriels, Rééducation de la Marche, Paralysie Cérébrale, Jeux vidéo actifs, Apprentissage Moteur, Chirurgie Multisite

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"Il y a des êtres mystérieux, toujours les mêmes, qui se tiennent en sentinelles à chaque carrefour de notre vie."

Patrick Modiano

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# Acronyms

Acronym	Meaning
2MWT or 6MWT	2- or 6-Minutes Walking Test
AR	Augmented Reality
ARRoW-CP	Augmented Reality Rehabilitation Of Walking - Cerebral Palsy
AVG	Active Video Game
CGA	Clinical Gait Analysis
COM	Center of Mass
COP	Center of Pressure
CP	Cerebral Palsy
ES	Effect Size
HAS	Haute Autorité de Santé (in french)
GMFCS - E & R	Gross Motor Function Classification System - Expanded & Revised
ICF	International Classification of Functioning, Disability and Health
OGT	Overground Gait Training
MINORS	Methodological Index for Non-Randomized Studies
MPST	Muscle Power Sprint Test
MR	Mixed Reality
RCT	Randomized Control Trial
ROM	Range of Motion
SCPE	Surveillance of Cerebral Palsy in Europe network
SDK	Software Development Kit
SEMLS	Single-Event Multi-Level Surgery
SRT	Shuttle-Run Test
SG	Serious Game
TD	Typically Developing
VR	Virtual Reality
WHO	World Health Organization

# Chapter 1

# General introduction

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Chapter 1 is a general introduction presenting the rationale, objectives, and structure of the thesis. Then the list of published and submitted articles is attached.

## 1.1 Rationale

Cerebral palsy (CP) is childhood's most common motor disability, causing activity limitations and participation restrictions. It is a group of permanent disorders of the development of movement and posture attributed to non-progressive brain lesions that occurred in the developing fetal brain or during the first year of the child's life. While children grow and age, musculoskeletal growth-related deformities lead to gait abnormalities that severely impair their quality of life. An orthopedic strategy, named single-event multilevel surgery (SEMLS), can be used to realign the musculoskeletal system in both lower limbs during a single operative period. The few studies with a long-term follow-up showed a slight improvement in all gait and functional parameters, but the results differed. These results could be influenced by the surgical gesture itself, the child's preoperative status, and the postoperative rehabilitation. This rehabilitation is long, with an average of 4.5 months. One crucial stage is gait rehabilitation, during which children face several obstacles. On the one hand, it is challenging for children with CP to integrate their new gait pattern after their new musculoskeletal system's "instantaneous acquisition" after surgery. On the other hand, keeping a high level of motivation for a long time is a challenge. It is essential to find complementary (and enjoyable) solutions to improve and intensify gait rehabilitation after surgery.

Using Active Video Game (AVG) for motor rehabilitation has generated an extremely positive reaction from therapists and children. In recent years, multimedia technologies, such as Virtual or Augmented Reality headsets, have become considerably democratized and diversified. In particular, Augmented Reality (AR) is a technology adding virtual elements (sometimes called holograms) to modify or augment the real environment. These virtual objects are enriched by multimodal perceptual information, notably visual, auditory, or haptic. Thus, the users perceive a virtual scene superimposed on their real environment and can interact in real-time. The fields of application of this technology are extensive, ranging from the industrial world to optimize production processes to the surgical environment by facilitating access to various medical information for the surgeon.

In this thesis, we wanted to explore and use AVG in AR for motor rehabilitation, specifically for gait rehabilitation in pediatric neurology. First, the last decade has seen a growing interest in AVG used for motor rehabilitation purposes. Their forms are diverse, from commercial games to customized games developed in a research laboratory. However, there is currently no research, fundamental or clinical, on the use of AVG in AR for gait rehabilitation. Second, if providing feedback is an effective strategy to improve motor function in children with CP, feedback characteristics are multiple and have not been explored in the context of AR rehabilitation. we hypothesize that an AVG in AR incorporating both motor learning ingredients and motor rehabilitation principles would lead to relevant innovation for postoperative gait rehabilitation of children with CP.

## 1.2 Objectives

The global objective of this thesis is to evaluate an AVG in AR to improve the walking capacity of children with CP after SEMLS. Through this thesis, we are in response to more specific objectives, including research investigation about step detection with an AR headset and feedback characteristics. The scientific issues are multidisciplinary in motor rehabilitation and mixed reality, both fundamental and clinical. We answer the following research questions :

- Is post-operative rehabilitation standardized ?
- $\square$  Do the spatiotemporal parameters of walking measured with an AR headset are reliable ?
- So What are the model and characteristics of feedback in motor rehabilitation ? Are they compatible with AR ?
- S Can walking speed be controlled using virtual feedback delivered in AR ?
- Does an AVG in AR improve walking capacity in children with CP ? Does an AVG in AR improve children's motivation for gait rehabilitation ?

## **1.3** Thesis structure

Chapter 1 is a general introduction presenting the rationale, objectives, and structure of the thesis. Then, the list of published and submitted articles is attached.

Chapter 2 is state of art about cerebral palsy and its therapeutic strategies. In particular, we have conducted a literature review on rehabilitation after Single-Event Multi-Level Surgery, and we propose a 5-step framework to standardize this rehabilitation (Article 1). We present the ingredients for effective motor rehabilitation. To conclude, we propose an in-depth overview of games, serious games, active video games, virtual reality, and augmented reality, clarifying their definition and their use in the field of motor rehabilitation.

Chapter 3 investigates the feasibility of using an AR headset as a unique sensor and device for gait recognition in a real (overground) environment. First, we present the experiment assessing the reliability of the device that we have selected (Microsoft Hololens AR headset) (Article 2). In our context, the Hololens is a sufficiently reliable tool to measure in real-time the position of the participants' head during different walking situations, in comparison with a motion capture system. Second, we detail our step detection algorithm (HoloStep - open source code), accurately measuring the spatiotemporal gait parameters, without additional sensors, from the head's position. We present its experimental validation with a population of children with CP (Article 3).

Chapter 4 focuses on feedback for motor rehabilitation in AR. We expose our theoretical model of feedback in AR. Then we detail the results of two clinical studies testing the impact of scenarios combining different visual feedback characteristics on walking speed in adult population (Article 4) and children with CP (Articles 5 & 6). We have demonstrated that specific feedback characteristics helped reach and maintain target speed. Finally, we discuss our observations of different profiles of people based on their responses to feedback.

Chapter 5 describes our final active video game, called ARRoW-CP, which proposes a 4-weeks protocol with a series of walking sprints to be performed at high intensity. We precise the game development framework used to develop ARRoW-CP. Finally, we expose the clinical study protocol designed to test the efficacy of ARRoW-CP (*Article 7*). Today, we evaluate ARRoW-CP with children with CP at the Ellen Poidatz Rehabilitation Center. We present the preliminary results of this clinical study.

# **1.4** List of Publications

- Article 1 Guinet AL, Khouri N, Desailly E. Rehabilitation after Single event Multi Level Surgery in children with cerebral palsy. Qualitative literature review. Am journal of PM&R, 2021
- Article 2 Guinet AL, Bouyer G, Otmane S and Desailly E. Reliability of the head tracking measured by Microsoft Hololens during different walking conditions. Comput Methods Biomech Biomed Engin, 2019
- Article 3 Guinet AL, Bouyer G, Otmane S and Desailly E. Validity of Hololens Augmented Reality Head Mounted Display for measuring gait parameters in healthy adults and children with cerebral palsy. Sensors, 2021, 21(8), 2697
- Article 4 Guinet AL, Bouyer G, Otmane S, Biddiss E and Desailly E. Development of a serious game for gait intervention in Augmented reality: a prospective study on the impact of virtual feedback on walking speed. (Submitted to JMIR Serious Game)
- Article 5 Guinet AL, Bouyer G, Otmane S and Desailly E. Towards an AR game for walking rehabilitation: Preliminary study of the impact of augmented feedback modalities on walking speed. IEEE ISMAR, 2020
- Article 6 Guinet AL, Bouyer G, Otmane S and Desailly E. Visual feedback in Augmented Reality to maintain a target walking speed. Cross sectional study including children with cerebral palsy. (Under review in IEEE TNSRE)
- Article 7 Guinet AL, Bams M, Payan-Terral S, Otmane S, Bouyer G and Desailly E. ARRoW-CP: Effect of an Augmented Reality Active Video Game for Gait Training in Children with Cerebral Palsy following Single-Event Multilevel Surgery - Protocol for a Randomized Controlled Trial. (Submitted to BMJ Open)

# Chapter 2

# State of the art

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Chapter 2 is state of art about cerebral palsy and its therapeutic strategies. In particular, we have conducted a literature review on rehabilitation after Single-Event Multi-Level Surgery, and we propose a 5-step framework to standardize this rehabilitation *(Article 1)*. We present the ingredients for effective motor rehabilitation. To conclude, we propose an in-depth overview of games, serious games, active video games, virtual reality, and augmented reality, clarifying their definition and their use in the field of motor rehabilitation.

## 2.1 Introduction

"Cerebral Palsy is a lifelong condition" \*

We begin this chapter by defining cerebral palsy to understand better its clinical symptoms and its different classifications (Section 2.2). We will see that behind this single name is to find for many patients. From impairment in body function and structure to gait abnormalities, children with CP are affected in different forms and degrees. These troubles also impact their activity and participation in daily life.

After that, we provide an overview of the current therapeutic strategies and their efficacy (Section 2.3), including motor intervention. Among treatments to solve contracture and joint alignment, we devote a section on Single-Event Multi-Level Surgery (SEMLS), an efficient strategy to optimize biomechanics. We describe this surgery, its results on strength, range of motion, and gait parameters. Some of these parameters, such as walking speed, did not improve after SEMLS. We present our literature review on rehabilitation after SEMLS (*Article 1*) (Section 2.4.2). This review suggests a five-step framework allowing a more precise and comprehensive description of postoperative rehabilitation protocol.

To further the discussion, we will expose the main ingredients of motor learning that should be applied and beneficial to motor rehabilitation (Section 2.5). Our objectives have been to implement these ingredients in a postoperative protocol to enhance walking speed after SEMLS. At this stage, we justify our hypothesis that the video game could be a solution to efficiently deliver a structured rehabilitation while respecting some essential principles of motor learning. Finally, we present an in-depth overview of the game, serious game, active video game already tested in motor rehabilitation, and the systems such as virtual reality and augmented reality to display games used with therapeutic purposes (Section 2.6).

# 2.2 Cerebral Palsy

### 2.2.1 Definition, Clinical symptoms and Classifications

The international community  $\dagger$  defines Cerebral Palsy (CP) as follows: "Cerebral palsy describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain." [2]. This definition reflects the concept that CP is one group of neurodevelopmental disorders which involves numerous developing functions. As in other neurodevelopmental disorders, various manifestations of the disordered brain may appear more significant in different persons or at different life periods, e.g. some aspects of the motor impairment [3], sensory loss [4], intellectual disability [5], attention difficulty, epilepsy [6], respiratory problems [7], musculoskeletal dysfunction [8] and many others maybe more prominent at different stages of the life of a person with CP (Figure 2.1).

<sup>\*.</sup> Livre Blanc de la Paralysie Cérébrale [1].

<sup>&</sup>lt;sup>†</sup>. An International Workshop on Definition and Classification of Cerebral Palsy was held in Bethesda, Maryland (USA), on July 11-13 2004, co-sponsored by United Cerebral Palsy Research and Educational Foundation in the USA and the Castang Foundation in the United Kingdom, supported by the National Institutes of Health / National Institute of Neurological Disorders and Stroke and the Dana Foundation.

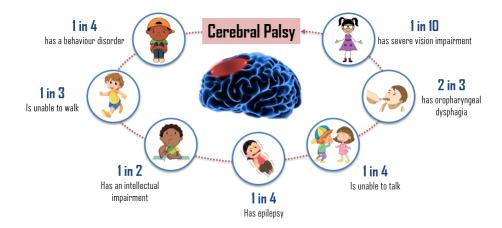


Figure 2.1 – **Infographic of Associated impairments of children with cerebral palsy**. This is an original figure. The estimated prevalence come from the Report of the Australian Cerebral Palsy Register published in 2018. [9]

In 2007, a collaborative group inside the Surveillance of Cerebral Palsy in Europe Network (SCPE) has proposed to classify CP into *three* main groups (or subtypes) based on neurological findings : *Spastic* (bilateral or unilateral), *dyskinetic* (dystonic or choreoathetotic) and *ataxic*. When it is a mixed CP form, i.e. spasticity with ataxia and/or dyskinesia, the child is classified according to the dominant clinical feature. The large majority of children are classified as spastic CP (54.8 % bilateral spastic, 33.5 % unilateral spastic), then dyskinetic for 8.0 % and ataxic for 3.8 % [10]. The topographic classification relies on the localization/limb distribution of neuromotor impairment in spastic CP : Monoplegia, Hemiplegia, Diplegia, Triplegia and Quadriplegia [11].

As noted previously, cerebral palsy includes a variety of clinical phenotype [12]. An easy way for clinicians to share a rapid profile about walking capacity of their patient is a classification system. In particular, to evaluate the functional capacities of children with CP, one of the most used classification system is the Gross Motor Function Classification System - Expanded & Revised (GMFCS - E & R) [13]. This 5-level classification system describes the gross motor function of children and youth with CP on the basis of their self-initiated movement with particular emphasis on sitting, walking, and wheeled mobility. Distinctions between levels are based on functional abilities, the need for assistive technology, including hand-held mobility devices (walkers, crutches, or canes) or wheeled mobility (Appendix 5.6). The reliability and stability of the GMFCS over time has been validated with inter-rater reliability of 0.93 and a test-retest reliability of 0.79 [14]. The GMFCS has been widely used around the world as a common language to describe gross motor function of children with CP (more than 1,500 citations) [15]. A recent cohort study in Canada have registered 36% GMFCS I, 12% GMFCS II, 12% GMFCS III, 17% GMFCS IV and 23% GMFCS V [16]. In our work, we are focusing on children with CP who are ambulatory [17], *i.e children with GMFCS I-III*.

Other tools are used to draw the patient profile. The International Classification of Functioning, Disability and Health (ICF) is a classification for health and health-related domains. ICF is the World Health Organization (WHO) framework including 5 categories that are linked together: Body functions and structure, Activity, Participation, Environmental Factors and Personal Factors. This interconnection means that changing in any area of the framework can potentially have influences elsewhere. This concept requires a more holistic approach to a person with disability.

In the field of childhood disability, Rosenbaum et al. have extended the ICF with the "F-Words": **Fitness, Function, Family, Friends, Fun, Future**. This makes it possible to define the child not only by functional or structural disorders but also to take into account their contextual factors (both environmental and personal), and their possibilities (both activity and participation). The "F-Words" approach has influenced our work, in the sense that we have followed this holistic approach to develop a therapeutic strategy that considers the person as a whole and in the different planes of her/his life.

#### 2.2.2 Prevalence and Risk Factors

CP is the most common motor disability of childhood with an estimated prevalence of 17 million people worldwide [18]. In Europe, the prevalence of CP decreased linearly from 1.90 for 1000 births in 1980 to 1.77 in 2003 with a range of 1.57–1.99 between countries, i.e. a mean decrease of 0.7 % per year. However, the absolute number of children "born with CP" rose from 2,701 in 1980 to 4,394 in 2003 (+ 62.3 %) [19]. The Australian Cerebral Palsy Register revealed a decreasing prevalence from 2.2 for 1000 births in 1997, 2.2 in 2003 to 1.4 in 2012, i.e respectively 1,259 births, 1,338 and 1,265 in 1997, 2003 and 2012 [9]. In the United States, the prevalence of CP for 2008 was 3.1 per 1000 8-year-old children, this trend has remained relatively constant since 1996. Using data from the North of England Collaborative Cerebral Palsy Survey for births during the period 1991 to 2000, Glinianaia et al. estimated there is 22,077 children living with CP in 2020, a 7.5 per cent increase compared with 2013 [20]. In Canada, the number of people living with cerebral palsy is expected to increase from more than 75,000 in 2011 to more than 94,000 in 2031 (prevalence rate from 0.222 to 0.236 per 100 population) [21]. Thus, although the relative prevalence is decreasing in some countries, the increase in births is leading to an increase in the absolute number of people living with cerebral palsy. This number is high in comparison to other severe childhood disabilities, such as Down syndrome.

Preterm birth is the most important risk factor for CP. This risk in infants born before 28 weeks of gestation is approximately 50-times that of full-term births [22]. Other factors are a difficult labour with neonatal asphyxia or oxygen deprivation, placental abnormalities and fetal growth retardation [23]. Correlation are observed between the socioeconomic status and CP, there is an increased risk of CP in low-income and middle-income countries versus high-income countries [24].

#### 2.2.3 Impairment in Body Function and Structure

Cerebral palsy is associated with various body function and structure deficits, which largely depend on the type (hypoxia and ischaemia are the most reported) and the location of the brain lesion. The disruption of cortico–striatal–thalamic–cortical and cortico–cerebellar–cortical networks impairs motor planning, coordination, muscle strength regulation, motor learning and fine motor skills [18]. But the clinical symptoms can also be a consequence of activity limitations during critical periods for activity-dependent and use-dependent plasticity. For example, impairments such as attention and perceptual deficits, communication, executive functions and arithmetic performance could be considered as a secondary consequence of activity limitation. Another significant finding is that restricted sensorimotor experience, caused by less neural stimulation owing to fewer or less-complex movements, impedes motor learning. As a consequences of poor range of movement, but also by lack of muscular activities and of many other factors, children with CP develops muscle contractures (defined as limited joint movement that results from high passive muscle force) and bone deformities amongst other problems [25]. The most common bone deformities in adolescents with CP are excessive femoral anteversion, hip subluxation, patella alta, excessive external tibial torsion, and pes valgus [26].

Muscles and muscle fibres are different for CP than typically developing (TD) children: shorter, smaller and reduced diameter. All CP children do not present the same alteration. The sarcomere are longer and fewer in number conducing to low active force, high passive muscle forces [27]. There is a decrease in the number of muscle stem cells, that is responsible for the growth in skeletal muscle and regeneration [28].

Weakness, Selective Motor disorders, Spasticity and Dystonia are the most common clinical phenomenons observed in children with CP. Weakness is a major cause of disability, positive correlations have been found between strength, gross motor function and functional outcomes [18]. Lance defined spasticity as "a motor disorder characterised by a velocity-dependent increase in tonic stretch reflexes (muscle tone) with exaggerated tendon jerks, resulting from hyperexcitability of the stretch reflex, as one component of the upper motor neuron syndrome" [29]. Recently, an expert consensus based on Delphi method, suggested that the term hyper-resistance should be used to describe the phenomenon of impaired neuromuscular response during passive stretch, instead of spasticity or hypertonia. They distinguished non-neural (muscle tissue properties involving elasticity, viscosity and muscle shortening) from neural (velocity dependent stretch hyperreflexia and non-velocity dependent involuntary background activation) contributions to hyper-resistance. The term spasticity should only be used next to stretch hyperreflexia, and the term stiffness next to passive (muscle) tissue-related contributions to hyper-resistance [30]. Dystonia is defined as a movement disorder characterized by sustained or intermittent muscle contractions or co-contractions causing abnormal and repetitive movements and/or postures. Dystonia is often initiated or worsened by voluntary action. All these symptoms lead to activity and participation restrictions, for example a limited walking capacity affecting the daily life of children with CP.

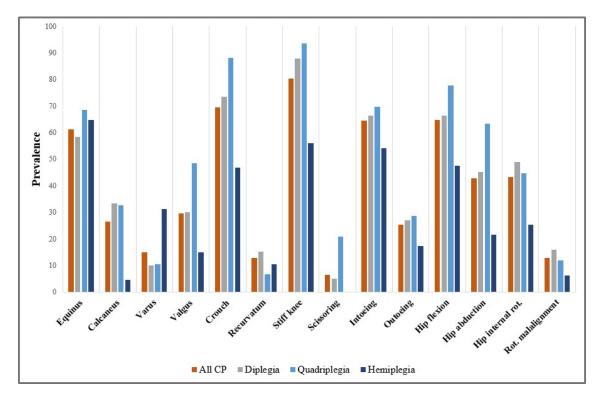
#### 2.2.4 Gait Abnormalities

Motor disability can range from minimal to profound, depending on the individual deficits in the ICF domains. As noted above (Figure 2.1), a third of people with CP cannot walk. In a recent cohort study including 336 patients, 45% achieved walking 10 steps without aids with a mean age 40 months (standard deviation of 22.1 months, upper age 120 months), 10% only ever achieved walking with the use of aids (mean age 45 months, standard deviation 15.2, upper age 94 months) and 45% never achieved walking [31]. Moreover, walking ability is an activity that may decline with age, more than 25% of adults with CP experience decline in mobility [32]. The pattern of walking in children with CP gradually deteriorates over time. Several factors, such as physiological muscle change, physical growth, surgical interventions, fatigue, and psychological aspect, are potentially implicated in this degradation [18,33]. According the gait definitions from Wren et al. [8], the most common gait problem in their group of patient was stiff knee in swing (80%), followed by crouch (69%), excessive hip flexion (65%), intoeing (64%), and equinus (61%) (Figure 2.2a) [8]. But these definition of gait abnormalities is now more than 15 years old and therefore does not take into account the latest advances in motion analysis. In 2019, based on a systematic review including articles published after 2005, six reliable and valid multiple joint patterns for children with CP reached a consensus in literature [17]. This classification provides an update and a common language among healthcare and researchers (Figure 2.2b). The prevalence of gait disorders will likely change with this new classification if it is adopted by clinicians. In any case, the gait disorders of children and adolescents with CP are extremely varied. More and more clinical assessment tools allow to measure precisely the differences within the same "class" of gait disorders. This allows a more personalized adaptation of medical, rehabilitative and surgical treatments. We will discuss some of these strategies in Section 2.3.

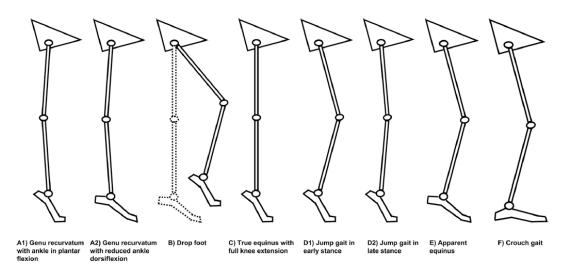
A recent meta-analysis including 35 articles identified and quantified gait variables and measures of dynamic stability in children with CP (n=822) and typically developed (TD) children (n=592) to find out the subset of most deteriorated gait parameters (See Figure 2.3) [34] :

For spatio-temporal parameters, a large effect size (ES  $\geq 0.8$ ) was observed for gait velocity, step length, stride length at the preferred-walking speed in favour of TD children whereas stride time, step width and double limb support time are significantly higher in children with CP. These parameters are the most deteriorated spatio-temporal gait parameters for children with CP. Figure 2.3 brings out these differences. A moderate effect size was observed on cadence (ES=0.29) and single limb support time (ES=0.39) in favour of TD children (Figure 2.3). Moreover, in other study, the comparison of spatiotemporal gait parameters revealed that TD children walked faster (1.79 vs. 1.07 m/s), with higher cadence (115.44 vs. 89.39 step/min) and longer stride length (1.11 vs. 0.81 m). Step width was larger in children with CP due to poor balance and gait instability (0.12 vs. 0.074 m) [35].

Kinematic parameters such as range of motion of the knee (ROM knee) in the sagittal plane and ROM pelvis (sagittal and transverse planes) exhibited a large effect in favour of TD. These parameters are the most deteriorated kinematics gait parameters for children with CP. The effect of CP was moderate (ES  $\geq 0.5$ ) on COM-COP inclination angle (frontal and sagittal planes) and ROM ankle (sagittal plane) parameters. Foot progression angle, ROM hip (transverse plane) and ROM pelvis (frontal plane) were least influenced by CP (Figure 2.3).



(a) **Prevalence of Gait abnormalities in children with CP** A stiff knee, hindering foot clearance, is more common than equinus for children with diplegia and quadriplegia. *From Wren et al.* [8]



(b) **Classification of Multiple joint pattern.** The bold black lines indicate the deviations comprising the pathological multiple joint patterns, the dashed lines in B represent the controlateral lower limb. *From Papageorgioua et al.* [17]

Figure 2.2 – Gait impairments prevalence and classification in children with CP. These gait abnormalities are the most frequent in this population. Each of them are well-described in [8,17].

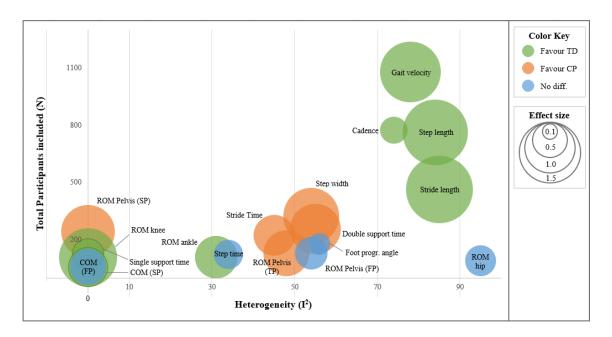


Figure 2.3 – Gait variables differences between TD and CP children. Favour TD indicates larger value of the corresponding gait variable in TD vs CP people. Favour CP indicates larger value in CP vs TD people. No diff. means that there are no differences between CP vs TD people.  $I^2$  is the heterogeneity among the studies. The threshold of  $I^2$  was determined from the Cochrane interpretation i.e. 0% to 40% might not be important, 30% to 60% may represent moderate heterogeneity, 50% to 90% may represent substantial heterogeneity, 75% to 100% considerable heterogeneity. COM: Center of Mass-Center of Pressure; ROM: range of motion; TP: tranversal plane; SP: sagittal plane; FP: frontal plane; progr.: progression. This is an original figure created with the supplementary data from Chakraborty et al. [34]

In a recent ecological study, Carcreff et al. have compared multiple gait parameters between two distinct environments both in children with CP and TD: the laboratory to measure the walking capacity and real life to evaluate walking performance using five synchronized Inertial measurement units-based devices (Physilog4®, GaitUp, Switzerland) [36]. In both groups of children, no difference was found for the amplitude (knee angle), smoothness (Higuchi's fractal dimension [37]) and coordination (walk ratio and cyclogram) domains. However, stride time rhythm, stride length and also stride time, stance time and stride length variability found increased in daily life. As a contrary, speed decreased in daily life for both groups, from 1.15 to 0.91 m/s for CP children, and from 1.28 to 1.15 m/s for TD (median value). The authors observed more difference between conditions in TD children than in CP children, that could be due to the heterogeneity among children with CP. Moreover, this can be the consequence of better capability of TD children to adapt their gait to the context. Children with lower capacity may have difficulties to efficiently respond to unpredictability [38].

To summarize, children with CP have different gait abnormalities causing a huge diminution of walking speed and step length, a larger step width, and more time in double support. These differences were observed both in laboratory and in real environment. These gait disorders contribute to reduced mobility and motor function, which affects their daily activity. *This initial observation has contributed to the choice of focus point on spatiotemporal gait parameters, and especially walking speed.* 

# 2.3 Therapeutic strategies

Although the brain injury that causes CP cannot be healed, the resulting physical impairment can be managed with a wide range of treatments and therapies, more or less invasive. Not every child with CP will follow the same "healthcare pathway" because of the diversity of impairments, that we have exposed previously. Physical therapy, occupational therapy, speech and language therapy, along with adaptive equipment, are the most common options for children with CP. Other therapies such as drug therapy, surgery, assistive technology, complementary medicine also exist.

In the following section, we will overview the therapeutic strategies adopted for children with CP, from clinical evaluation (2.3.1) to conventional care (2.3.2).

### 2.3.1 Clinical Evaluation

The first therapeutic strategy is a thorough clinical evaluation to individualize and adapt these interventions. The most common assessment tool to measure body functions and structure is **physical examination**, assessing joint mobility, tone, spasticity, muscle strength, and the degree of selective muscle control. In 2017, in the Fondation Ellen Poidatz gait laboratory, two phases had been completed to standardize this clinical examination: a review of the literature and a consultation with focus groups (multidisciplinary team). As a result, we have identified 34 measures plus their standardized instructions.

In complement to this clinical examination, and to quantify more precisely musculoskeletal impairments, and also to monitor progress and outcomes on the gait patterns which evolve, the **Clinical Gait Analysis** (CGA) is currently integrated into clinical practice [39]. Moreover, statistical models and machine learning techniques using patient data from the clinical exam and CGA can also provide essential and relevant information for treatment planning [40–42].

Supplementary questionnaires, tests, clinical observation, based on the ICF framework assess Participation, Quality of Life and Activity. Participation is defined as a person's involvement in a life situation, including domestic life, school, community, social, and civic life. Examples of outcome measures are the Paediatric Evaluation of Disability Inventory (PEDI) and the ABILOCO-Kids. Quality of Life is defined as the impact of disease and treatment on physical, psychological and social functioning, that could be assessed with the Cerebral Palsy Quality of Life questionnaire for Children (CP-QOL). Activity is divided into three sub-domains :

- a. activity capacity that represents a person's ability to execute a task in a standardised environment. This could be assessed with the Gross Motor Function Measure 66- or 88item (GMFM-66 or GMFM-88). Walking capacity could be evaluated more specifically with standardized tests such as 6 Minutes Walking Test (6MWT), Muscle Power Sprint Test (MPST) and Shuttle Run test (SRT). We will describe in details these three tests in Section 5.4.2 because we have used it in our randomized control trial (Chapter 5).
- b. activity capability that defines a person's ability to execute a task in his or her daily environment. This could be evaluated with the Activities Scale for Kids (ASK) and International Physical Activity Questionnaire (IPAQ).
- c. **activity performance** represents what a person actually does in his or her environment. This could be assessed with accelerometers and pedometers.

## 2.3.2 Conventional care

In 2020, Novak et al. have updated their fantastic work first published in 2013 about the interventions for preventing and treating children with CP. This state of the evidence "Traffic Lights" is probably the most broadly shared in the community [43] (Figure 2.4).

In this subsection, we will describe intervention strategies for Motor (2.3.2.1), Early Intervention (2.3.2.2), Tone (2.3.2.3) and Contracture & Alignment (2.3.2.4), linked to our work and we will make a brief overview of their efficacy.

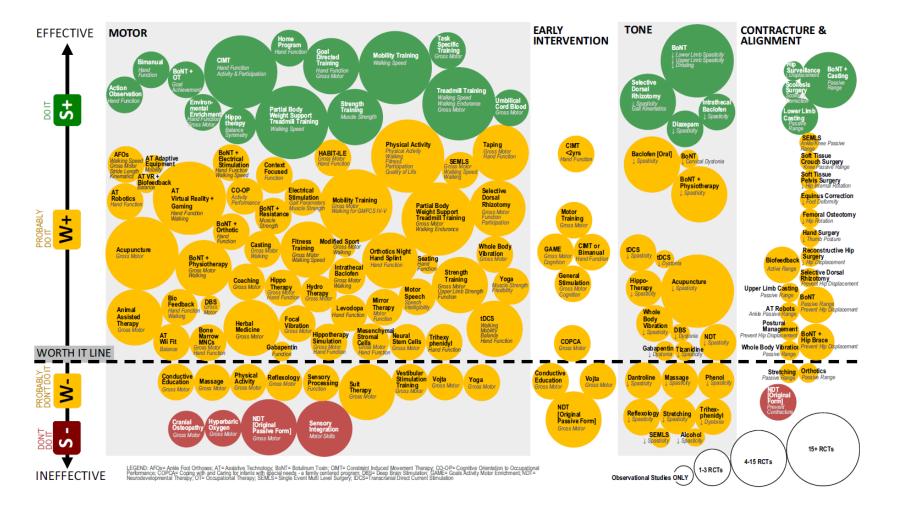


Figure 2.4 – State of the Evidence Traffic Lights 2019. Systematic Review of Interventions for Preventing and Treating Children with Cerebral Palsy. From Novak et al. [43]

#### 2.3.2.1 Motor interventions

Novak et al. presented an apparent dichotomy in the evidence base for what works and does not. Motor interventions that improve function and performance are action observation training, bimanual training, constraint-induced movement therapy, functional chewing training, goal-directed training, mobility training, treadmill training (green lights, "go" intervention). Additionally, virtual reality serious gaming is classified as yellow lights, weak positive. When combined with task-specific motor training, this may augment the positive effects of training [44].

Furthermore, the French Haute Autorité de Santé (HAS) prioritized strengthening exercises, aerobic exercise, cardiorespiratory training, and functional gait training [45]. The HAS also recommended intensive rehabilitation programs, such as Hand and Arm Bimanual Intensive Therapy Including Lower Extremity (HABIT-ILE). During ten days, therapists delivered 90 hours of functional and play activities. Both upper and lower extremity functions (AHA, ABILHAND-Kids, PEDI, 6MWT, ABILOCO-kids) progressed [46].

A recent randomized controlled trial tested a combined progressive resistance and functional anaerobic training program. They showed improvements in plantar flexor strength, peak anaerobic power (MPST), functional strength (total number of repetitions performed in 30 s for three functional exercises), agility  $(10 \times 5 \text{ m sprint test})$ , and walking distance (6MWT) [47]. An example for an effective gait training protocol was given by Zwinkels et al. [48]. They have evaluated the effects of 8-weeks of High-Intensity Interval Training, including a series of walking sprints on fitness in youth with physical disabilities. They highlighted positive effects on health, in particular on anaerobic performance (MPST), agility  $(10 \times 5 \text{ m sprint test})$ , aerobic performance (SRT), and systolic and diastolic blood pressure. Moreover, a recent systematic review reported the positive effect of functional gait training on walking speed, gross motor function, walking endurance, and functional mobility [49].

Physical activity has significant health benefits for the heart, body, and mind among motor intervention. The WHO recommended that adolescents living with disability should do at least an average of 60 minutes per day of moderate-to-vigorous intensity, including aerobic activities, strengthening exercises, at least three days a week [50]. Although the HAS recommended including physical activity and sport as part of the overall management of people with CP, the report pointed out that there were few clues as to the dose, intensity, and activity to be preferred [45]. Physical activity intervention is classified as yellow lights, weak positive on walking, fitness level, participation, and quality of life [43].

#### 2.3.2.2 Early Intervention

The improvement of early diagnosis in CP allows the development of early interventions. These child-active motor learning early interventions, such as baby-CIMT (which is an adaptation of the Constraint-Induced Movement Therapy), appeared to confer improvement in motor and cognition skills [51]. Other novel interventions (baby-bimanual, Goals Activity Motor Enrichment, small steps) have reported positive gains in movement skills (yellow lights, weak positive) [43].

#### 2.3.2.3 Tone

Spastic contractures could be treated with casting and night splinting. Intramuscular injection of BoNT-A is widely practised but the benefits and risks are not well known. Novak et al. highly recommended Botulinum Toxin (BoNT) to reduce spasticity, and BoNT+casting to improve passive range of motion (green light) [43]. But some studies point out the negative long-term effects such as weakness and atrophy of muscle [52].

Selective Dorsal Rhizotomy (SDR) is an invasive procedure that consists cutting the excitatory nerve fibres emerging from the proprioceptors in the muscle spindles. This procedure is questioned today, because it has been shown that 22% to 59% of patients required additional treatment with oral medication, BoNT, or intrathecal baclofen pumps and the long-term effect on spasticity remains uncertain [53]. Moreover, the interventions that reduce spasticity do not increase the activity or gross motor function in children with CP [18].

#### 2.3.2.4 Contracture & Alignment

Once a contracture has begun to develop, serial casting can be applied to effectively reduce or eliminate early/moderate contractures in the short term (green light) [43]. An efficient strategy to correct severe contracture to maintain alignment, muscle length and optimize biomechanics is Single-Event Multi-Level Surgery (SEMLS). This orthopedic surgery is defined as corrections of soft tissue and/or bony deformities at a minimum of two anatomical levels during a single operative event. The advantage is that only one hospital admission and recovery period are required. Surgical Interventions can be :

- Muscle lengthening (Psoas, Medial and/or lateral hamstrings, adductor, apneurotic gastrocneumius-soleus, apneurotic gastrocnemius)
- Transfer (Rectus femoris transfer, foot tendon, patella lowering)
- **Release** (Rectus femoris release)
- Tendon lengthening (Tendon Achilles, foot tendon)
- Osteotomy (Femoral derotation or deflexion osteotomy, tibia derotion osteotomy, foot osteotomies)

Therefore, SEMLS is a powerful intervention to simultaneously address the biomechanics of gait and minimize repeated surgeries (yellow light). We will develop the results of this surgery in the next section 2.4.

## 2.4 Focus on Single-Event Multi-Level Surgery

As we have seen in the previous paragraph, SEMLS has been recommended to treat contracture and joint alignment [43]. In the following section, we are providing a brief outline of the results of SEMLS (2.4.1) and we will present our literature review on rehabilitation after SEMLS (*Article 1*) (2.4.2).

### 2.4.1 Postoperative Results

#### 2.4.1.1 Muscle Strength

Thomason et al. [54] assessed static muscle strength using a manual dynamometer before and 1 year after SEMLS. In their study, the authors tested a lower extremity muscle strengthening program with progressive resistance between the  $3^{rd}$  and  $6^{th}$  postoperative months. The results POST-PRE were not statistically different. Moreover, as reported in Table 2.1, muscle strength parameters grew slightly, remained the same or decreased.

	Pre-operative	1 y Post-operative
Leg Press 1 1RM (kg)	116 (+/- 33)	110 (+/- 40)
Quadriceps (kg)	11.5 (+/-2.5)	11.2 (+/-4)
Hip Extensors (kg)	6.4 (+/-2.3)	7.2 (+/-3.3)
Hip Abductors (kg)	5.5 (+/-1.5)	6.7 (+/-2.4)
Ankle Extensors (kg)	7 (+/-2.8)	10.3 (+/- 4.5)

Table 2.1 – **Pre- and post-operative muscle strength outcomes in children with cerebral palsy.** The one repetition maximum (1RM) is a measurement of the greatest load (in kilograms) that can be fully moved (lifted, pushed or pulled) during a single exercise. *In red, negative results. In green, positive results.* 

#### 2.4.1.2 Range of Motion

One year postoperative, most of the passive joint range of motion (ROM) at the lower limb joints after SEMLS were improved. These results have persisted 2 years after surgery. From 8 studies, hip and knee flexion deformity (in french, "flessum") decreased and hip, knee and ankle ROM were also enhanced in most of studies [54–61]. To facilitate interpretation of the various results, an average of these parameters for the included studies has been calculated (Figure 2.5).

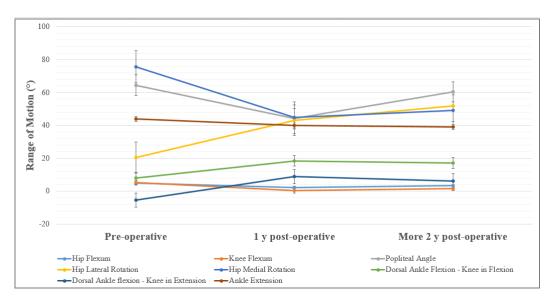


Figure 2.5 – Evolution of ROM after SEMLS on children with cerebral palsy. Means are extracted from 8 surgical case series [54–61].

#### 2.4.1.3 Kinetics & Kinematics Gait Parameters

SEMLS resulted in good improvement in most kinematics and kinetics gait parameters, in particular for knee ROM, knee flexion and ankle dorsiflexion at initial contact and minimal knee flexion in stance phase, but also for maximum dorsiflexion in stance and in swing phase, and foot progression angles [62, 63].

#### 2.4.1.4 Spatiotemporal Gait Parameters

In their systematic review in 2012, McGinley et al. showed that general spatiotemporal gait parameters were unchanged or declined after SEMLS in 7/15 studies [62]. In a more recent systematic review, Lamberts et al. [63] reported significant improvement for stride length in 46% of the studies. On the other hand, mixed results were found for walking speed (69% unchanged, 24% improvement and 7% deterioration) (Figure 2.6). The Minimal Clinically Important Difference (MCID) for increase in walking speed is 0.1 m/s [49]. This number has been reached in only 4/17 studies in [63], and only these with a long-term follow-up (more than 3 years). Similar findings concerning unchanged walking speed were reported in other studies [64,65]. The stagnation or even degradation of walking speed observed during the first year after surgery encouraged us to focus our work on this specific spatiotemporal gait parameter.

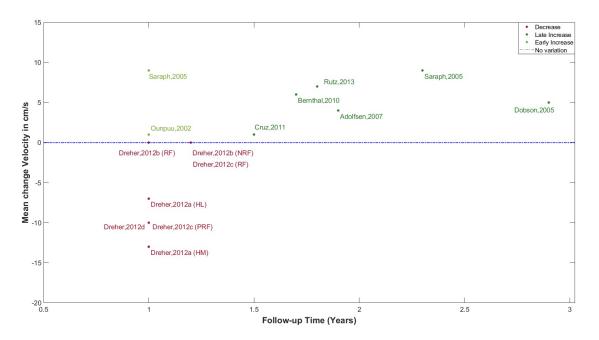


Figure 2.6 – Walking Speed Evolution before and after SEMLS. This figure presents a synthesis of the results published by Lamberts et al. [63] Color Code : in red, studies that show a decrease walking speed after SEMLS ; in green, studies that show an early improvement (during the first year after SEMLS) or a late improvement (more than 1 year after SEMLS)

#### 2.4.1.5 Activity & Participation

These ICF domains are still poorly evaluated after SEMLS. The Gillette Functional Assessment Questionnaire (FAQ) was improved in 3/3 studies whereas no improvement on the Gross Motor Function Measure (standing and walking dimensions) was observed [62].

### 2.4.2 Rehabilitation after SEMLS (Article 1)

The term "SEMLS" actually encompasses a wide range of surgical techniques, patients' characteristics are very diverse, and, as we will see, postoperative rehabilitation differs significantly among studies. As we have demonstrated above, SEMLS effectively improves several parameters, but results are mixed for some spatiotemporal parameters such as walking speed. Additionally, its effects on activity and participation remain discussed. Regarding those results, we would like to clarify that these studies do not solely assess the impact of SEMLS but the impact of SEMLS associated with postoperative rehabilitation. Indeed, the community unanimously agrees that postoperative rehabilitation is crucial to SEMLS success. But there is no actual consensus regarding intensity, frequency, type of exercise, period of immobilization, and prescription of orthoses. Both clinicians and researchers recommended that the postoperative rehabilitation content/duration should be better described in reports of SEMLS studies [62].

At the outset of this thesis, we have conducted a literature review on the rehabilitation after SEMLS for children and young adults with CP. This review sought to describe and analyze published protocols for rehabilitation after SEMLS for people with CP, to identify their differences and limits, and to introduce a common step-by-step framework for future descriptions and assessments of postoperative rehabilitation protocols.

The MEDLINE, Embase, CINAHL, and the Cochrane Library databases were searched. Inclusion criteria were as follows: (1) single-event multilevel surgery, (2) full-text reports published after 1985, and (3) articles with a method section describing the rehabilitation protocol. Interventions were coded using the Oxford Levels of Evidence and the Methodological Index for Non-Randomized Studies Index (MINORS).

Twenty-four articles were included in the review. Studies included patients aged 4–30 years with spastic cerebral palsy (hemiplegia, diplegia, and quadriplegia). Postoperative rehabilitation in most included reports took place in a rehabilitation center (69%) and continued for 4.5 months (4 sessions/week). However, the difference among studies was large, regardless of the surgery type and the patient's functional level. This review provides relevant information about the modalities, contents, limits, and difficulties associated with the post-SEMLS rehabilitation protocol reported in the literature. This review identified five steps in the post-SEMLS rehabilitation protocol that could/should be described more precisely in terms of objective, content, and intensity in all future studies assessing SEMLS. This *five-step framework* allows a more precise and comprehensive description of postoperative rehabilitation (Figure 2.7). Pain during the first and second steps was identified as a major problem. Moreover, the authors insisted on two main axes of the fourth step : *muscular strengthening and gait training that should be more intensive and more qualitative*. This review has considerably oriented our work on this specific step, and the idea of *gait training is certainly a path which we have attempted to explore*.

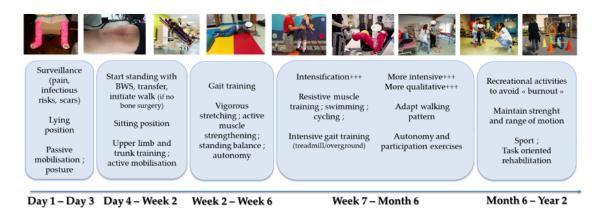
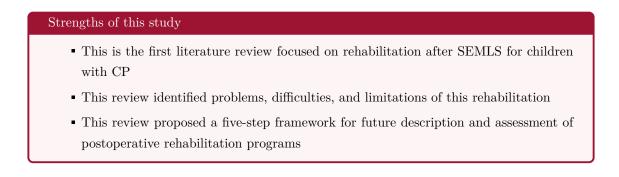


Figure 2.7 – The 5-step Framework for Rehabilitation after SEMLS



This work has been presented during the Société Française de Médecine Physique Et Réadaptation - World Federation for NeuroRehabilitation Congress in October 2020. This paper is attached to this thesis manuscript as published in the American Journal of Physical Medicine & Rehabilitation journal in 2021 [66].

## Rehabilitation After Single-Event Multilevel Surgery for Children and Young Adults With Cerebral Palsy

## A Systematic Review

Anne-Laure Guinet, MS, Néjib Khouri, MD, PhD, and Eric Desailly, PhD

Abstract: This review sought to describe and analyze published protocols for rehabilitation after single-event multilevel surgery for people with cerebral palsy, to identify their differences and limits, and to introduce a common step-by-step framework for future descriptions and assessments of postoperative rehabilitation protocols.

The MEDLINE, Embase, CINAHL, and the Cochrane Library databases were searched. Inclusion criteria were as follows: (1) singleevent multilevel surgery, (2) full-text reports published after 1985, and (3) articles with a method section describing the rehabilitation protocol. Interventions were coded using the Oxford Levels of Evidence and the Methodological Index for Non-Randomized Studies Index.

Twenty-four articles were included in the review. Studies included patients aged 4-30 yrs with spastic cerebral palsy (hemiplegia, diplegia, and quadriplegia). The mean postoperative rehabilitation duration was 4.5 mos, with 4 sessions per week, and rehabilitation took place in a rehabilitation center. This review provides relevant information about the modalities, contents, limits, and difficulties associated with the post-SEMS rehabilitation protocol reported in the literature. Pain was identified as a major problem.

A more precise and comprehensive description of post-SEMS rehabilitation protocols would be useful. The proposed five-step framework could be used by future studies to standardize their protocol description in terms of objective, content, and intensity.

Key Words: Cerebral Palsy, Child, Orthopedic Procedures, Rehabilitation, Systematic Review

(Am J Phys Med Rehabil 2021;00:00-00)

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- Foundation and Association Nationale Recherche et Technologie (ANRT), in collaboration with the University of Evry-Paris-Saclay.
- The Ellen Poidatz Foundation manages 29 healthcare institution around Paris. It provides support to more than 2200 children and 300 adults with disabilities. The Research and Innovation Department develops research and innovative projects that aim to propose new practices and care modalities for people with disabilities. The Fondation Ellen Poidatz funded a part of Anne-Laure Guinet's employment contract.
- The ANRT is the public-private intersectoral network for French research. Their work is intended to contribute to the organizational transformation of French and European research and innovation systems, as well as to the themes of energy, digital technology, education, sport, space, health, etc. The ANRT funded a part of Anne-Laure Guinet's employment contract.
- Financial disclosure statements have been obtained, and no conflicts of interest have been reported by the authors or by any individuals in control of the content of this article. Supplemental digital content is available for this article. Direct URL citations appear
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• erebral palsy (CP) is a group of permanent disorders of - movement development and posture, causing activity limitation, which are attributed to nonprogressive disturbances of development in the fetal or infant brain.<sup>1</sup> Cerebral palsy is the most common cause of childhood disability, affecting 1.77 per 1000 neonates (3.2 per 1000 in the United States),<sup>2</sup> with an estimated global prevalence of 17 million people.<sup>3,4</sup> During growth, children with CP develop secondary musculoskeletal and bone disorders, such as muscle-tendon contractures or torsions in long bones, caused by multiple factors.<sup>5,6</sup> Consequently, children with CP become less active, contributing to gait impairment, fatigue, muscle and weakness.<sup>7–9</sup> Since 1985, an orthopedic strategy, named single-event multilevel surgery (SEMLS) in both lower limbs during a single operative period, has been used to realign the musculoskeletal system and perform tendon transfer, muscle lengthening, derotation or deflexion osteotomy, and joint stabilization in individuals with CP. The results at 1 yr after SEMLS have been varied. The passive range of motion (ROM), kinetics and kinematics gait parameters, overall gait index (Gait Deviation Index, Gillette Gait Index), and energy efficiency were improved.<sup>10–15</sup> On the other hand, muscle strength and spatiotemporal parameters remained the same or worsened. In terms of the International Classification of Functioning, Disability and Health activity and participation domain, the Gillette Functional Assessment score was improved,<sup>12,15,16</sup> whereas improvement on the Gross Motor Function Measure (standing and walking dimensions) was inconsistent, particularly in high-functioning children.<sup>17,18</sup> In the quality-of-life domain, most outcome measures remained unchanged after SEMLS, but this outcome has only recently been introduced. The few studies with a long-term follow-up showed a slight improvement in all gait and functional parameters, but the results differed among studies.<sup>18-20</sup> A previous systematic review found low levels of evidence to support the use of SEMLS, because most of the included studies were level 4 on Oxford Center for Evidence-Based Medicine scale.<sup>2</sup> They reported that SEMLS must be accompanied by follow-up and a rehabilitation program. They recommended that the postoperative rehabilitation protocol/duration should be better described in reports of SEMLS studies.<sup>21</sup> The authors unanimously agreed that a rehabilitation program is crucial to SEMLS success.<sup>20,22-24</sup> No study has compared the effect of SEMLS only versus SEMLS associated with a well-described rehabilitation protocol. In terms of postoperative rehabilitation protocols, there is no actual consensus regarding intensity, frequency, and type of exercise, period of immobilization, as well as prescription of orthoses. It was hypothesized that the rehabilitation protocol could explain the low surgical effectiveness in terms of function (e.g., walking ability), quality of life, and long-term results. Before studying the effectiveness of a specific rehabilitation protocol, a systematic review

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of the rehabilitation protocols proposed in the literature is needed. Thus, this systematic review aimed (1) to describe postoperative rehabilitation protocols in studies reporting SEMLS, (2) to identify the problems and limits of these rehabilitation protocols, and (3) to propose a step-by-step framework for postoperative rehabilitation to standardize their future description and assessment.

### **METHODS**

### Search Strategy

A literature search of four electronic databases indexing health-related journals, that is, MEDLINE, Embase, CINAHL, and the Cochrane Library, was conducted between February 2019 and July 2021. The search equation was cerebral palsy [Title] AND ((orthopedic surgery) OR (multilevel surgery) OR (multilevel surgery)) AND (physiotherapy) OR (rehabilitation) OR (physical exercises). Filters were: humans, child: birth to 18 yrs, and young adult: 19–30 yrs, from 1985 to 2021. The search was limited to English articles, between 1985 and July 2021, and to children and young adults (younger than 30 yrs) with CP. Targeted manual searching of reference lists and key journals identified additional reference titles. No authors were contacted to provide additional data. The search strategy was developed and reviewed by multiple authors (ALG, ED, NK).

This systematic review was performed following the Preferred Reporting Items for Systematic Reviews and Meta-Analyses statement (see Supplementary Checklist, Supplemental Digital Content 1, http://links.lww.com/PHM/B384).<sup>25</sup> The original method of the systematic review, with a full list of search terms, was registered on PROSPERO and can be accessed online (CRD42019127502).

### **Eligibility Criteria**

Titles and abstracts were initially reviewed by two authors, and any potentially eligible titles were identified. Full-text articles were independently examined for eligibility by two of the authors. If consensus could not be reached, agreement was obtained through discussions with a third person (NK). Studies were included if they met the following criteria: (1) the surgical strategy was SEMLS, defined as a surgical intervention involving two or more joints and including two or more bony or soft tissue procedures; (2) patients with CP; (3) full-text reports published after 1985; and (4) detailed description of the post-SEMLS rehabilitation protocol (with objectives, content, steps or periods, intensity, exercises, indications/contraindications); brief descriptions and simple references to a physiotherapy program were excluded.

This review included only original, peer-reviewed published literature and "gray literature," such as the team approach published in scientific books.

### Data Extraction

The articles were independently reviewed by two authors (ALG, ED) and the following data were extracted:

- (1) Study design.
- (2) Study location.
- (3) Demographic data, including sample size, age, and sex distribution.
- 2 www.ajpmr.com

- (4) Diagnosis and description of CP. Information regarding movement disorder, topographical distribution, and gross motor function, including the Gross Motor Function Classification System (GMFCS),<sup>26</sup> was sought.
- (5) The description of the orthopedic surgery was scrutinized to determine the procedures and the number of surgical procedures performed.
- (6) Information pertaining to the postoperative rehabilitation protocol was classified as follows: place of rehabilitation (hospital, rehabilitation center...), time of rehabilitation, time and modalities of immobilization, time and modalities of wearing a gait orthosis, number of sessions per week, types of physical exercises and steps in the rehabilitation protocol, difficulties encountered, and the limits of the rehabilitation.

The method for defining a framework for post-SEMLS rehabilitation included extraction of both the general modalities and the special instructions described by the authors and dividing the overall description of the rehabilitation protocol into separate steps. To this end, the protocol description in the reports was screened, and a new step was considered when the overall objective, content, and/or intensity of the rehabilitation changed. When most authors described the same step, it was considered validated and integrated into the framework.

### **Quality Appraisal**

The Methodological Index for Non-Randomized Studies (MINORS) tool was chosen to classify the studies because of the significant number of nonrandomized controlled trials (RCTs).<sup>27</sup> When the study did not include participants, the MINORS tool was not applied, but descriptive and comparative elements of the different rehabilitation protocols were collected. This was typically the case for "team approach" publications.

#### RESULTS

#### Search Yield

The database search yielded 1136 published articles (Fig. 1). After removing duplicates, identification of additional records through manual searches and other sources, and reviewing the titles and abstracts, 71 full-text reports were retrieved. After the application of predefined inclusion and exclusion criteria, 24 fulltext reports were included in the systematic review (Table 1).

#### **Study Design**

Six of the 24 studies were retrospective in nature.<sup>28–32</sup> One of these was a comparative retrospective study.<sup>11</sup> Nine were prospective cohort studies,<sup>20,33–36,38</sup> with three comparative prospective studies and nonrandomized controls.<sup>13,18,39</sup> Two studies were RCTs.<sup>22,40</sup> One study was a feasibility study, with a control group versus an interventional group.<sup>41</sup> One study used a single-case experimental design,<sup>37</sup> with introduction and withdrawal of the independent variable.<sup>45</sup> Five were team approach articles, with complete recommendations for rehabilitation after SEMLS.<sup>23,24,42–44</sup> Sample sizes ranged from 6 participants.<sup>38</sup> to 47 participants.<sup>28</sup> Power calculation was not described in any publication. All studies, except the team approach articles, assessed the efficacy of SEMLS on gait parameters.

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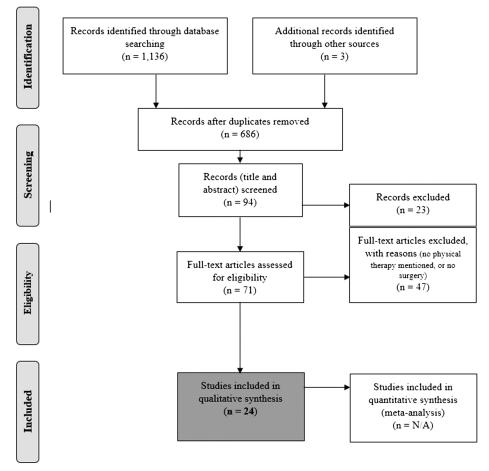


FIGURE 1. Flow chart detailing the literature search and selection process.

### Study Quality

The level of evidence varied from 1 (RCT) to 5 (expert opinion) on the Oxford Center for Evidence-Based Medicine Scale. The quality of the studies varied considerably, with MINORS scores ranging from 6 to 12 and from 15 to 19 using the additional MINORS criteria for comparative studies (Table 1).

## **Study Location**

Most of the studies originated from Europe (14 of 24) and North America (5 of 24), and a few from Asia (3 of 24) and Australia (2 of 24). Some articles had been published by the same multidisciplinary teams: in total, 20 different teams published at least 1 article describing rehabilitation after SEMLS (Fig. 2).

#### Participant Characteristics

Patient ages ranged from 4 to 30 yrs (mean = 10.7 yrs). Male patients predominated (219 male vs. 126 female), in accordance with the sex distribution in CP registers.<sup>46,47</sup> This information was missing from four experimental studies. Only persons with spastic CP were included in the studies, but all topographical patterns of the movement disorder were represented: monoplegia, hemiplegia, diplegia, triplegia, and quadriplegia. Spastic diplegia was the most commonly included form. Most patients were at GMFCS levels II (159) and III (112), whereas the remainder were

at GMFCS level I (59) or IV (15). This information was missing for team approach studies and for one prospective study published before introduction of the GMFCS level.<sup>33</sup> Overall, 345 patients were included (mean = 18 individuals per study; Fig. 2).

## **Surgical Procedures**

All studies included both soft tissue and bone procedures, except for three studies that reported only tendon lengthening and muscle release.<sup>34,36,38</sup> The range and details of the procedure were provided in 15 reports. A mean of 5.1 procedures were performed per patient (range = 2–13). The ratio of soft tissue to bone procedures was 75:25. Six studies detailed the surgical procedures.<sup>13,18,29,36,38,40</sup> In total, 27 different surgical procedures were identified. The most commonly used method was hamstring lengthening. Hereafter, the outcomes were evaluated considering that every patient, although treated with SEMLS, may have received different combinations of surgical procedures (Fig. 2, supplementary data additional, Supplemental Digital Content 2, http://links.lww.com/PHM/B385).

#### **Rehabilitation Protocol**

### **Place of Rehabilitation**

Rehabilitation took place in hospital (primary referral place), rehabilitation centers (secondary referral place), and at home, with

Age	Study			Population			Inter	Intervention				Rehabi	Rehabilitation <sup>b</sup>			õ	Quality
$ \begin{array}{c c c c c c c c c c c c c c c c c c c $			n i	Age (Mean/Range),	C.	GMFCS	Surgical Procedures,			How				•	Phases Described		
$ \begin{array}{cccccccccccccccccccccccccccccccccccc$	First Author	Design	(F, M)	yr	Type	Level	Mean	Outcomes"	Where	Long				Exercises	Implicit	Oxford'	MINORS
	Lofterød <sup>28</sup>	Retro	47 (18, 29)	10.1 (NR)	H, D	III-I	3.9	B, A	7	2	7	2	0	1	2/3	4	8
$ \begin{array}{c c c c c c c c c c c c c c c c c c c $	Saraph <sup>29</sup>	Retro	22 (NR)	12.6 (7.4–16.6)	D	II-II	8	В	7	7	0	7	2	1	2/3	4	6
Gaminalistication         Retron         86, (12)         146 (12–30)         D         111         3.8         B, A         2         2         2         2         2         34         4           Debler <sup>21</sup> Retron         416 (12, 3)         38 (NR)         D         1-111         4.1         B, A         0         0         1         2         0         1         24         4           Gaugitii         Proop         155 (5, 20)         86 (4-12)         D, Q         1-1V         NR         B, A         0         0         0         2         1         24         4           Name <sup>31</sup> Proop         18 (1, 7)         85 (6-10)         D, H-1U         NR         B, A         2         2         2         1         24         4           Amichai <sup>30</sup> Proop         18 (1, 7)         85 (6-10)         D         H-11         NR         A, A         2         2         2         0         1         23         4         4         3         4         4         5         5         3         4         4         5         5         3         4         4         5         5         5         5	Hoiness <sup>30</sup>	Retro	7 (3, 4)	13 (9–16)	D, Bi	Ш	4.7	B, A, Q	7	7	7	2	1	1	2/3	4	6
$ \begin{array}{c ccccccccccccccccccccccccccccccccccc$	Ganjwala <sup>31</sup>	Retro	18 (6, 12)	14.6 (12-30)	D	Π	3.8	B, A	7	2	7	2	2	2	3/4	4	10
$ \begin{array}{cccccccccccccccccccccccccccccccccccc$	Dreher <sup>32</sup>	Retro	44 (16, 28)	9.8 (NR)	D	III-I	4.1	B, A	0	0	1	2	0	1	2/4	4	12
$ \begin{array}{c c c c c c c c c c c c c c c c c c c $	Gough <sup>11</sup>	Retro	24 (NR)	6.1 (5.1–6.9)	Bi	I–IV	2.5	В	2	0	0	2	0	1	2/5	4	18
$ \begin{array}{c c c c c c c c c c c c c c c c c c c $	Nene <sup>33</sup>	Prosp	18 (6, 12)	12.4 (8-16)	D	NR	4.3	B, A	0	0	0	2	1	1	2/5	4	9
$ \begin{array}{cccccccccccccccccccccccccccccccccccc$	Chang <sup>34</sup>	Prosp	25 (5, 20)	8.6 (4-12)	D, Q	VI–II	NR	B, A	2	7	0	2	0	2	3/4	б	8
$ \begin{array}{cccccccccccccccccccccccccccccccccccc$	Amichai <sup>35</sup>	Prosp	18 (11, 7)	8.5 (6.8–10.5)	D, H	III–III	1.8	B, A	0	7	7	1	0	2	5/2	4	8
$ \begin{array}{cccccccccccccccccccccccccccccccccccc$	Dequeker <sup>20</sup>	Prosp	34 (12, 22)	12.4 (6–25)	D	III-I	NR	Α, Ρ	7	2	7	0	7	1	3/4	С	11
$ \begin{array}{rrrrrrrrrrrrrrrrrrrrrrrrrrrrrrrrrrrr$	Kondo <sup>36</sup>	Prosp	25 (15, 10)	8.2 (4–16)	D, Q	I–IV	13.4	A	7	2	2	0	2	1	2/3	4	11
Haberfehlner <sup>18</sup> Prosp $6(3,3)$ 13.8 (10.6-17.3)         NR         II-III         1.8         B         2         2         2         1 $5/5$ 4           Grecco <sup>30</sup> Prosp         15 (NR)         11.05 (8-15)         NR         II-III         3.5         B, A         2         2         2         2         2         3         3           Thompson <sup>13</sup> Prosp         15 (NR)         11.05 (8-15)         NR         II-III         3.5         B, A         2         2         2         0         2         2         2         3         3           Thompson <sup>13</sup> Prosp         20 (8,12)         11.05 (6-12)         D         II-III         8         B, A, Q, P         2         2         0         0         2         3         3           Meyns <sup>41</sup> Prosp         26 (6,19)         6.2 (+10.3)         D         I-III         6         B, A, Q, P         2         2         3         3         3         3         3         3         4         5         5         5         5         5         5         5         5         5         5         5         5         5	Akerstedt <sup>37</sup>	Prosp	11 (1, 10)	13.8 (9–18)	H, Bi	III-I	NR	B, A, Q	7	7	0	0	2	2	4/5	4	12
$ \begin{array}{cccccccccccccccccccccccccccccccccccc$	Haberfehlner <sup>31</sup>		6(3,3)	13.8 (10.6–17.3)	NR	III–III	1.8	В	7	2	2	0	2	1	5/5	4	12
$ \begin{array}{cccccccccccccccccccccccccccccccccccc$	Grecco <sup>39</sup>		15 (NR)	11.05 (8-15)	NR	III–III	3.5	B, A	0	7	0	0	2	2	2/3	С	15
$ \begin{array}{c ccccccccccccccccccccccccccccccccccc$	Thompson <sup>13</sup>	Prosp	20 (8, 12)	11 (9–14)	D	III-I	5.1	В	0	7	0	2	0	1	4/5	б	16
Patikas <sup>40</sup> Prosp         39 (12, 27)         9.8 (6-16)         D         I-IV         10.1         B, A         2         2         0         0         2         1         3/5         2         2           Meyns <sup>41</sup> Prosp         11 (7, 4)         11.6 (6-18)         D, H, Q         I-IV         3         A         2         2         0         0         2         1         2/3         3           Buckon <sup>18</sup> Prosp         11 (7, 4)         11.6 (6-18)         D, H, Q         I-IV         3         A         2         2         0         0         2         1         2/3         3           Buckon <sup>18</sup> Prosp         25 (6, 19)         6.2 (4-10.3)         D         I-III         6         B, A, Q, P         2         2         2         3/4         3           Harryman <sup>42</sup> Team appr         NA	Thomason <sup>22</sup>	Prosp	19 (NR)	NR (6–12)	D	III–III	8	B, A, Q, P	7	7	0	2	0	2	3/4	7	17
Meyns <sup>41</sup> Prosp         11 (7, 4)         11.6 (6-18)         D, H, Q         I-IV         3         A         2         2         0         0         2         1         2/3         3           Buckon <sup>18</sup> Prosp         25 (6, 19)         6.2 (4-10.3)         D         I-III         6         B, A, Q, P         2         2         0         0         2         2         3/4         3           Harryman <sup>42</sup> Team appr         NA	Patikas <sup>40</sup>	Prosp	39 (12, 27)	9.8 (6–16)	D	I–IV	10.1	B, A	7	7	0	0	2	1	3/5	7	18
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<ul> <li><sup>a</sup> The ICF domains: B, body functions and structure; A, activity; P, participation; Q, quality of life.</li> <li><sup>b</sup> Quality grades: 0, not reported; 1, reported but partially; 2, fully reported.</li> <li><sup>c</sup> Oxford Centre for Evidence-Based Medicine scale.</li> <li>D: Lincomb, D. Andonio, D. Andonio, D. Andonio, David Andre Study, Botto, percented and the structure for Evidence Participation for two himmobility for the structure for two himmobility for two himmobility for the structure for two himmobility for two himmobility for the structure for two himmobility for two himmobility for the structure for two himmobility for two himmobility for the structure for two himmobility for two himmobility for the structure for two himmobility for two himmobility for the structure for two himmobility for two himmobility for the structure for two himmobility for the structure for two himmobility for the structure for two himmobility for two himmobil</li></ul>	Gage <sup>24</sup>	Team app.		NA	NA	NA	NA	NA	1	7	7	2	0	2	5	5	NA
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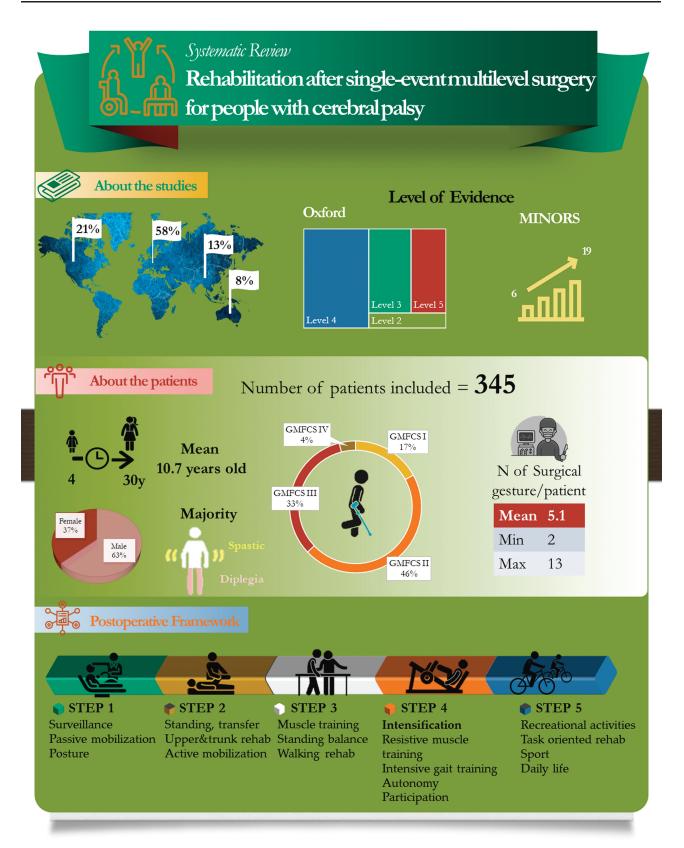


FIGURE 2. Infographic for the systematic review: rehabilitation after SEMLS for people with CP. Study characteristics (study location, level of evidence), patient characteristics, and five-step postoperative framework were presented.

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or without the presence of a physiotherapist (tertiary referral place). In most studies, patient care was initiated at the hospital. The patients stayed for an average of 7 days. After the first week, rehabilitation locations were redistributed heterogeneously: rehabilitation centers in 11 protocols; home in 5 centers; and home and a short period in a rehabilitation center in 1 study.<sup>42</sup>

### **Duration of Rehabilitation**

Eighteen teams started the rehabilitation protocol within 1-3 days after surgical intervention. The rehabilitation protocol duration ranged from 4 wks to 2 yrs postoperatively and was influenced by the rehabilitation location. A shorter period was associated with a hospital stay (4-6 wks), with no description in the rehabilitation program after this period.<sup>29,30</sup> When the rehabilitation protocol was implemented in a rehabilitation center, the protocol lasted between 9 wks and 1 yr, with an average of 4.5 mos. In three studies, when the children were able to walk independently with a K-Walker or crutches, they left the rehabilitation center and continued their physical therapy at home.18,20,41 Few studies proposed home programs after a rehabilitation center stay. In this case, the duration of stay at a rehabilitation center was shorter (mean = 8 wks), but the total duration of rehabilitation was 2 yrs. Most studies recommended returning to the usual program and frequency of rehabilitation sessions as soon as children reached or exceeded the preoperative functional state.<sup>13,23,24,35,43</sup> One protocol, a community-based rehabilitation program lasted 2 yrs.<sup>31</sup> Buckon et al.<sup>18</sup> specified the following criteria before discharge from inpatient therapy: ability to ambulate 100 feet (30 meters) on a level surface with and without an assistive device; ability to transfer in and out of bed, wheelchair, and/or appropriately sized chair, with and without assistive devices, with standby assist; ability to transfer from the floor to standing with the support of a stationary object; ability to transfer on and off the toilet and in and out of the tub, with or without assistive device, with minimal assistance; ability to don and doff clothing in a sitting position (excluding shoes, braces, fasteners); and ability to ascend and descend five stairs using a hand-rail and appropriate assistive device, with standby assist.

#### Time and Modality of Immobilization

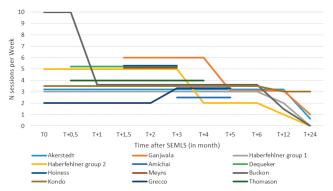
For immobilization, casts were recommended by most articles (16 of 22), although removable splints or braces were also mentioned (6 of 22). The length of casts depended on the joint affected by the surgical procedure, for example, above-knee cast, below-knee cast, short or long, cast with distal extension up to toes. The mean time for cast or splint immobilization was 5.1 wks (range = 2-12 wks). Soft tissue surgery required shorter cast immobilization than bony procedures. Some studies suggested the use of radiographic evaluation of all osteotomy sites when the casts were removed 6 wks after surgery, before encouraging full weight-bearing.<sup>31,35</sup>

## **Time and Modality of Gait Orthoses**

Every study recommended prescription of orthoses post-SEMLS. The type and time of implementation of orthoses varied according to the surgical procedure and other factors. Walking with orthoses started between weeks 4 and 6 postop-eratively, which corresponded to the time of weight-bearing.<sup>18,31,33</sup> The children had to retain their orthoses for several months (minimum = 3 mos, maximum = 6 mos). Orthoses were used until muscle balance at the knee was established or when the calf muscle power was sufficient to stabilize the knee during stance.<sup>29</sup> Various types of orthoses were used, depending on the age, the cooperation of the child, the aim of the orthoses, and the surgical stability. Three types of orthoses were identified: ground reaction force ankle foot orthoses, principally used for children who walked with apparent equinus or crouch gait; hinged or solid ankle foot orthosis, and knee-ankle-foot orthosis, with different properties: nonarticulated or articulated; carbon-fiber or polypropylene, custom-made or commercial. Some studies recommended night orthosis or knee immobilizers to reduce the risk of recurrence of knee flexion deformity and to maintain the other corrections, from 6 mos to 1 yr after SEMLS.

#### Number of Rehabilitation Sessions per Week

Eleven studies defined the number of sessions per week (1-10 sessions/wk) and the time per session (0.5-2 hrs/session; Fig. 3). At the beginning of the rehabilitation, the difference between the studies was notable (2-10 sessions/wk), and different studies did not start the program at the same time. By 3 mos, this difference was reduced (3-6 sessions/wk), and at 6 mos, there was only a difference of 2 sessions among studies. The mean number of sessions per week decreased over time after 6 mos.



**FIGURE 3.** Number of physical therapy sessions after SEMLS. The maximum of physical therapy sessions was 10 per week (2 daily PT sessions); the minimum was 1 per week. At 24-mo postsurgery, many authors reported that PT was stopped. The start and end of the rehabilitation protocols were not homogenous. The number of sessions per week decreased over the time, after 6 mos.

# Type of Physical Exercises and Step(s) of Rehabilitation

The most practiced types of exercises were muscle strengthening (cited 29 times), walking/gait rehabilitation (24), passive (14) and active (10) ROM, transfer (10), and positioning (10). Muscle strengthening and gait rehabilitation were not detailed in the reports, except for two studies that assessed the effect of additional muscle strengthening<sup>48</sup> and treadmill training.<sup>39</sup> Passive and active lower limb joint mobilization was practiced by the therapist. Transfer was described as the ability to change body position, such as sit-to-stand transfer. Positioning was defined as immobilizing a joint or the body for a period, extending the knees on a positioning table, or standing on a raising table.<sup>49</sup>

# Define a Five-Step Framework for Rehabilitation After SEMLS

This study proposes a five-step framework for rehabilitation after SEMLS. Among the 24 studies included, step 1 was described or mentioned by 18 reports; step 2, by 23 reports; step 3, by all reports; step 4, by 22 reports; and step 5, by 14 reports. The five-step framework of rehabilitation is described according to the details presented in the reports (time after SEMLS; Table 2), for SEMLS involving bone and soft tissue procedures, and for patients with GMFCS level I–III (most patients in this review). It may require adaptation for soft tissue procedures only and for patients with GMFCS IV–V:

- Step 1 (days 1–3): The first step started early after SEMLS. One day after surgery, according to most reports, and at most 3 days after surgery, in two reports. It consisted of pain and infectious risk monitoring and scar care. Some reports emphasized ensuring the patient's comfort and reassuring the family. The postoperative instructions were as follows: patients were to remain in a lying position and were not allowed to sit for meals. During this time, physiotherapists prevented contractures and attempted to inhibit abnormal movement patterns with passive mobilization of the lower limbs.<sup>33,35,37</sup> Positioning and splinting were also practiced to stretch the muscle affected by SEMLS (e.g., hip flexors after psoas lengthening).<sup>23,43</sup>
- Step 2 (day 4 to week 2): During the second step, the time spent seated in an armchair in a corrected position (lower limbs raised and stretched) increased. Standing practice

Author	Step 1: D1–D3 Surveillance, Passive Mobilization	Step 2: D4–W2 Transfer, Sitting, Active Mobilization	Step 3: W2–W6 Soft Gait Training and Muscle Strengthening	Step 4: W7–M6 Intensification, Autonomy	Step 5: M6–Y2 Task-Oriented, Maintain Muscle Strength and Gait Pattern
Akerstedt <sup>37</sup>	а	а	а	Ь	а
Chang <sup>34</sup>	с	а	а	b	а
Ganjwala <sup>31</sup>	с	а	b	а	а
Haberfehlner <sup>38</sup>	а	а	а	а	а
Harryman <sup>42</sup>	а	а	а	а	b
Amichai <sup>35</sup>	а	а	а	а	а
Saraph <sup>29</sup>	с	а	а	Ь	d
Georgiadis <sup>43</sup>	а	а	а	а	d
Dequeker <sup>20</sup>	а	а	b	а	d
Høiness <sup>30</sup>	а	а	b	d	d
Meyns <sup>41</sup>	с	а	b	а	d
Berker <sup>23</sup>	а	а	а	Ь	а
Gage <sup>24</sup>	а	а	а	а	d
Dreher <sup>19</sup>	а	а	b	Ь	d
Buckon <sup>18</sup>	а	а	b	Ь	а
Kondo <sup>36</sup>	с	а	b	Ь	а
Nene <sup>33</sup>	а	а	Ь	Ь	b
Thompson <sup>13</sup>	а	а	а	Ь	а
Gough <sup>11</sup>	а	а	b	Ь	Ь
Patikas <sup>40</sup>	а	b	а	а	Ь
Grecco <sup>39</sup>	с	а	Ь	а	d
Lofterød <sup>28</sup>	а	а	Ь	d	d
Thomason <sup>22</sup>	а	а	b	а	d
Thomason <sup>44</sup>	а	а	а	а	а

TABLE 2 The 5 step model of rehabilitation and the authors' description of objective content, and/or intensity of rehabilitation after SEMI S

Each protocol described by authors was screened, and if the description indicated a change in objective, content, and/or intensity, a new step was considered. "Step detailed as a change in objective, content, intensity.

<sup>b</sup>Step mentioned but not detailed.

<sup>c</sup>Step not described.

<sup>d</sup>Physiotherapy stopped.

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began with body weight support and walking aids. Patients should be able to sit in a neutral position 2 wks after SEMLS, while sometimes remaining supine.<sup>42</sup> If SEMLS did not include bone procedures, a standing position and walking between parallel bars could be initiated.<sup>24,29,32</sup> Two reports considered that weight-bearing was allowed on the second to fourth day after soft tissue surgery, whereas it depended on the quality of internal bone fixation in combined soft tissue and bony operations.<sup>11,23</sup> Sit-to-stand transfers were trained under a physiotherapist's care to facilitate movement and patient autonomy.<sup>18</sup> Trunk and upper limb muscle strength training was also encouraged.<sup>23</sup>

- Step 3 (weeks 2–6): During the third step, physical therapy shifted from walking and passive ROM exercises to vigorous stretching, active muscle strengthening exercises, and gait training. This important rehabilitation period aims to maintain muscle length, regain muscle strength, maintain lower limb alignment, and encourage weight-bearing, standing, and walking. The protocol included continuation of activities in the supine and prolonged sitting position and introduction of exercises to strengthen the hip and knee muscles without weight-bearing.<sup>44</sup> The protocol focused on muscle strength training (in particular, that of the gluteus, quadriceps femoris, rectus femoris, gastrocnemius, soleus, hamstring, and rectus abdominis) and gait training, which was emphasized.<sup>20,24,29,31,33,34,42–44</sup> Gait training started by using proper walking aids (K-walker and crutches) to improve endurance and walking patterns. The distance and velocity were to increase over time. Gait training should be guided and tailored to the specific needs and capacities of each patient.<sup>35</sup> Standing balance and body weight transfer were two other important objectives. The physiotherapist needed to verify that active knee control was acquired while standing and walking and that there was no lumbar compensation during standing. Proprioception exercises were performed.<sup>20</sup> Indoor cycling training and swimming were considered ideal during this period.<sup>24</sup> All the activities needed to be progressive, with increasing intensity.
- Some teams began this third step of rehabilitation after muscle healing at week 4, rather than at week 2. Hence, postoperative weeks 2 and 3 aimed to maintain optimum positioning and facilitate transfer, to reach passive 90-degree knee flexion, and to educate the family and child regarding compensation.
- Step 4 (week 7 to month 6): The fourth step was an intensification period in the rehabilitation process. Several exercises were proposed, such as resistive muscle training, swimming activities, cycling, and horseback riding. Intensive (overground or treadmill) gait training has been suggested by some authors.<sup>39,44</sup> Progressive withdrawal of walking aids was advised. At this step, one team evaluated a virtual reality (VR) rehabilitation protocol with the Nintendo Wii Balance Board.<sup>41</sup> A test group performed three sessions per week of VR (30 hrs of additional therapy compared with the control group). Another team evaluated a program of muscle strengthening that began at week 4. It consisted of seven exercises targeting the flexors/extensors of the hip and knee, and rectus abdominis.<sup>40</sup> Another team proposed intensive gait training on a treadmill during weeks 8–20 (month 5). This step aimed to improve the walking pattern

using the ground reaction force. Children had to appropriate their own (and new) musculoskeletal system and to adapt their walking patterns to improve their function, autonomy, and participation.<sup>30,42,44</sup>

• Step 5 (month 6 to year 2): The fifth step was fully described by one report<sup>44</sup> and was mentioned in 12 other studies. These reports explained that to avoid "burnout" and to shift the emphasis toward strengthening and independence, physio-therapy was replaced with recreational activities, including family walking, bicycle riding, and swimming to maintain strength, ROM, and abilities. Children can benefit from sports or recreational activities adapted to their interests and goals. Therapists analyzed the specific tasks and activities that the patient would like to perform (task-oriented therapy).

## Difficulties Encountered and Limitations of Post-SEMLS Rehabilitation

The main reported difficulty was pain.<sup>23,30,31,35,38,39,43,44,48</sup> Pain, frequently reported as significant, was mainly experienced during the first postoperative month and was due to cast immobilization, passive mobilization when the cast was removed, and standing position. Medical treatment, including analgesic and antispasmodic drugs, was proposed.<sup>13,23,43</sup> A need for surveillance and objective evaluation of the pain was mentioned.

On the other hand, appropriate alimentation and skin care were necessary to prevent complications, such as "pressure sores."<sup>23</sup> No other adverse events resulting from post-SEMLS rehabilitation were reported.

#### DISCUSSION

This systematic review provides relevant information regarding post-SEMLS rehabilitation protocols. It identified the problems, difficulties, and limitations of this specific rehabilitation. It describes protocols in terms of objectives, content, and intensity. Finally, this review proposed a five-step framework for future description and assessment of postoperative rehabilitation protocols.

## A Five-Step Framework for a Postoperative Rehabilitation Protocol

The organization of rehabilitation differs among countries, depending on the social, political, and health insurance systems of each country. Postoperative rehabilitation in most included reports took place in a rehabilitation center (69%) and continued for 4.5 mos (4 sessions/wk). However, the difference among studies was large, regardless of the surgery type and the patient's functional level. Surprisingly, the number of sessions varied from a daily session at weeks  $4-6^{30}$  to three sessions per week during year 1,<sup>37</sup> in two studies including patients with the same GMFCS level (II) and involving SEMLS with both bone and soft tissue procedures.

This review identified five steps in the post-SEMLS rehabilitation protocol. The timing of each step is relevant to SEMLS that included bone and soft tissue procedures, because this represented most included studies. Some adaptation is necessary for SEMLS involving soft tissue procedures only. For example, it is interesting to note that recent studies had prioritized removable splints or braces over casts, except for surgery involving bony procedures.<sup>20,34,38</sup> Moreover, the immobilization phase was shorter when only muscle release surgery was performed, but this represented only two included studies.<sup>36,38</sup> The first two steps were quasi-unanimously described, whereas the third step was more disputed. It consisted of starting more vigorous stretches, active muscular work, and walking training. Some teams began this phase during the second week, whereas others waited until the fourth week, when muscles were theoretically healed. The difference between SEMLS involving bone and that involving soft tissue procedures is important in this third step, because soft tissue surgery allows an earlier standing position. The fourth step, from the seventh week to the sixth postoperative month, is a phase of intensification: exercises become more intense and qualitative. Children must develop their new musculoskeletal system, adapt their walking patterns, and improve their function, autonomy, and participation. Thus, during the fifth step, which continues until 2 yrs postoperatively, rehabilitation should continue to maintain muscle strength and ROM. Sport and motor activities with oriented tasks are encouraged to maintain a high level of motivation.

Although only one report described the rehabilitation process after 6 mos in detail, most authors mentioned the importance of continuing rehabilitation after 6 mos. This could involve home-based sessions or an intensive phase in a rehabilitation center with a physiotherapist or a specialist sports coach. In some studies with a long-term follow-up after surgery, there was a decline in participants' gait quality from the short to the long term.<sup>50,51</sup>

Therefore, it could be suitable to follow this five-step framework for rehabilitation to standardize the practice description across studies and to allow SEMLS plus rehabilitation across countries. The specific content and duration of each step should be adapted to the patient (by GMFCS level) and the surgical procedure (bone/soft tissue surgery), but this framework presents a rehabilitation protocol framework that is valid in most cases (Fig. 2). The five-step rehabilitation protocol allows testing of specific therapy during a specific step, while keeping the other steps the same. For instance, it is necessary to test a new intensification strategy in phase 4, as the first three steps should be comparable between the groups.

## Focus and Key Questions About Intensification Strategy

Reports agreed that rehabilitation is a key factor in SEMLS success. They insisted on two main axes of rehabilitation: muscular strengthening and gait training. From the seventh week postoperatively, these two axes are described as essential, and the term "intensification" appeared in the reports. However, the content of this phase was imprecise and heterogeneous. Nevertheless, this may help improve postoperative results in terms of body functions and structures, activities, and participation, as defined by the International Classification of Functioning, Disability and Health,<sup>52</sup> but no systematic review or RCT has assessed this strategy.

but no systematic review or RCT has assessed this strategy. Patikas et al.<sup>40,48</sup> proposed evaluation of the effects of a muscle strengthening program starting from 4-wk postsurgery. It consisted of seven exercises targeting the hip extensors and flexors, knee extensors and flexors, and abdominal muscles.<sup>40,48</sup> Exercises were practiced at home, at least 3 times a week. The modalities consisted of two sets of five repetitions for each exercise. Resistance was progressively increased by gradually

eliminating external support during the exercise. The test group did not show any significant advantages over the control group. This contrasted with previous studies that documented the advantages of strength training in people with CP.53 A highintensity functional anaerobic and progressive resistance training of the lower limbs, in three sessions per week for 12 wks, increased muscle size, strength, and functional capacity in young adults (15–30 yrs) with CP.<sup>54</sup> A strength training program, based on maximum isometric contraction of the quadriceps (4 sets of 5 repetitions; 3 times/wk for 6 wks) showed an effect size of 5.27 for increasing muscle strength.<sup>55</sup> Another program that proposed upper and lower limb strength training with important resistance (6 repetition maximum: the heaviest weight the child could lift with maximum effort for 6 consecutive exercise repetitions; 1 set of 6 repetitions; 3 times/wk for 10 wks) showed good results (effect size = 1.99).<sup>56</sup> It was hypothesized that the modalities chosen by Patikas et al.<sup>48</sup> (home-based program without supervision, started at an early stage, with low-level intensity) were not adapted and sufficient to improve strength in children with CP.

Similarly, Grecco et al.<sup>39</sup> proposed intensive gait training, starting at the eighth week, for 12 wks. It involved 1 session of 30-min gait training on a treadmill, without bodyweight support, at 80% of maximal VO<sub>2</sub>, which was added to the conventional rehabilitation protocol.<sup>39</sup> Both groups underwent surgery (group 1: soft tissue surgery; group 2: soft tissue and bone surgery), conventional rehabilitation, and intensive gait training. Improvements in the Gross Motor Function Measure 88 and 6-min walking test were significant. This result is even more significant when considering other studies without intensive gait training, in which these parameters were not improved.<sup>13,17,18</sup>

Many studies have concluded that intensification of rehabilitation improves motor learning and skill acquisition in children with CP.<sup>57,58</sup> Although these studies were not conducted after surgery, adaptation of such principles to post-SEMLS rehabilitation could improve postoperative outcomes. These studies were all based on the consistent characteristics of motor learning theories that affirm that practice variability facilitates skill acquisition.<sup>59,60</sup> The optimal conditions of practice, particularly in a neurorehabilitation context, are not well-defined. Schmidt schema theory states that practicing variations of a movement in different environmental conditions, with a sufficient intensity and rest period, improves skill acquisition, retention, and transfer.<sup>61</sup>

Some authors have suggested that physiotherapy in rehabilitation centers should stop when people are able to walk independently using posterior walkers or crutches. However, the concept of "to walk independently" is not well-defined. Patients could walk alone in the rehabilitation center corridors, but not outside in an "insecure" environment (with obstacles, encountering other people). Buckon et al.<sup>18</sup> specified that children were required to meet short-term goals before being discharged from inpatient therapy. These criteria should be defined before SEMLS, to the family and medical staff, and should be adapted according to the patient's abilities.

# Difficulties and Limitations of Rehabilitation After SEMLS

Another possible way to improve existing programs is to analyze the difficulties and limitations of the actual rehabilitation.

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In the 24 included articles, this type of information was not commonly reported. The difficulties and limitations were either not evaluated, or they were not reported. However, pain is a major obstacle in rehabilitation progress. It is first necessary to evaluate this pain on a daily basis to prevent it. A systematic evaluation should be performed, rather than waiting for the child to complain. After identifying the cause of the pain, a drug treatment or a distraction (games, VR) could be implemented rapidly.<sup>62</sup> Immersive VR is an effective adjunctive pain reduction technique in the pediatric population who are undergoing painful rehabilitation therapy.<sup>63</sup>

The frequency of rehabilitation sessions increased considerably after SEMLS (4 times a week) and remained higher after completion of programs delivered in rehabilitation centers. One year after surgery, the patients continued physical therapy once a week. It may be problematic to maintain a high level of motivation and to avoid "burnout." The development of gamification for rehabilitation using VR could be a suitable method, providing the user with goals, challenges, problem solving, and rules, as well as a clear internal value and an interactive experience.<sup>64</sup>

From an economic and societal perspective, this long and specific rehabilitation represents a significant cost for the healthcare system or for the family when it is not covered by any insurance.

#### Study Limitations

This study highlighted the difficulty in creating universal protocols for rehabilitation after SEMLS, considering the diversity of patients, surgical techniques, social characteristics, and available resources. However, this review identified five steps shared by specialized medical teams. Eligible articles were included regardless of methodological quality. The first part of this study aimed to gather as much information as possible about published rehabilitation protocols. Therefore, the methodological quality of the included clinical studies was not considered a limitation in the analysis of the contents of reported rehabilitation protocols. A potential bias in the included studies was the lack of information concerning the rehabilitation protocol and surgery (missing information about specific details of surgery, lack of detail on all steps of rehabilitation, and lack of justification for the various rehabilitation steps).

#### CONCLUSIONS

This review highlights multiple rehabilitation protocols after SEMLS. However, based on this review, a five-step protocol for rehabilitation after SEMLS was proposed to provide a basis and serve as a guideline for future studies. The standardization of rehabilitation practices is necessary to allow comparison of the results after surgery and to be able to test a specific rehabilitation protocol in a particular step, with all other steps being equal. Pain was identified as a major problem in rehabilitation after SEMLS and needs to be kept in mind.

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To summarize, this review proposes a five-step framework for rehabilitation after SEMLS. From the seventh week to the sixth postoperative month, the fourth step is a phase of intensification. Exercises become more intense while retaining movement quality. Two main axes are essential to maximizing rehabilitation program efficiency: muscular strengthening and gait training. *In this work, we are focused on gait training.* This axe has been chosen at the beginning of the thesis with medical, paramedical, and researcher staff from the Fondation Ellen Poidatz. Therapists reported an ambition to bring significant evolution of practices for gait training after SEMLS to enhance walking speed. The main reasons were the low speed in children with CP compared to typically developed children in preoperative context and the stagnation or even degradation of this walking speed after SEMLS. The next step was to explore the means to improve it. Even if the systematic review of Novak et al. recommended SEMLS, the authors provided no details on the rehabilitation protocol following surgery [43]. We could not duplicate any existing - and validated - postoperative protocol at this stage. Before modifying the fourth step, and gait training protocol, the first step was to investigate the fundamental ingredients of motor learning theory that could be integrated into motor rehabilitation to develop an efficient therapy.

## 2.5 Ingredients for an effective Motor Rehabilitation

Among motor (Section 2.3.2.1) and early interventions (Section 2.3.2.2) included in their *State of the Evidence Traffic Lights*, Novak et al. provided recommendations on effective therapies (DO IT or PROBABLY DO IT) [43]. In this current section, we address the question of the key components of the success of these therapies to see if we could implement these ingredients into a postoperative protocol.

A recent International Clinical Practice Guideline provides 13 recommendations for interventions to straighten up physical function for children and young people with CP. These recommendations focus more on the best ingredients that should be included in the clinical strategy than a "to-do list". The authors highlighted the importance of client-chosen goals, whole-task practice within real-life settings, support to empower families, enjoyable intervention, and a team approach [67]. The motor learning principles inspire these recommendations. At this stage of our work and to develop an effective therapeutic strategy, it was important to define and establish a clear picture of motor learning and motor rehabilitation.

Motor learning defines the changes associated with practice and/or experience in internal process (or state) allowing a person to perform a motor skill or to produce a certain movement [68]. Motor relearning is a term used for the patient after brain injury, bone fracture, or accident with multiple damages. It consists for the patient to re-learn a completely lost motor skill or physical function or movement [69]. The term motor relearning needs to be distinguished from the term rehabilitation. Rehabilitation helps the individual regain the ability to perform a particular movement or action. The same movement pattern may be recovered or may require a compensatory adaptive movement pattern. It is essential to define the terms motor recovery and motor compensation to avoid confusion and misunderstanding when communicating between disciplines (especially between neuroscience and clinical rehabilitation) and to adapt our therapeutic strategies better.

Levin et al. defined recovery of motor performance as "the reappearance of elemental motor patterns present prior to central nervous system injury" [70]. For people after stroke, for body function and structure domain of ICF, recovery means restoring the ability to perform a movement in the same manner as it was performed before the injury. On the other hand, motor compensation was defined as "the appearance of new motor patterns resulting from the adaptation of remaining motor elements or substitution" [70]. Compensation was to perform an old movement in a new manner. In other words, functions were taken over, replaced, or substituted by different body segments.

In the context of rehabilitation, clinicians promoted motor learning, reinforcing and perhaps bolstering the brain neuronal plasticity (also called neuroplasticity) [71], and therefore optimize acquisition, retention, generalization, and transfer of motor skills. The common consensus was that the principles of motor learning stayed the same as those for initial learning, relearning, and the recovery of movement function in rehabilitation [72]. But there are essential differences in how best to facilitate the stages of motor skill acquisition in these different situations. The information and condition of practice to support change in movement coordination, control, and performance depend on the individual learner, the task to be learned, and the stage of motor skill acquisition, but also the environment (ecological approach) [72]. The determination of what information was used to regulate action was particularly important. In other words, what augmented information does an individual need to learn or relearn a motor skill? To answer this question, we explored the recent literature on key factors that would influence motor learning in the rehabilitation setting for people with motor disorders [46,73–78]. We particularly included articles and references from the last **international clinical practice guideline** focusing on the interventions to improve physical function for children and young people with CP published in 2021 [79].

Based on these articles, we have extracted a list of 10 ingredients that are effective for motor recovery (Figure 2.8):

- High intensity practice, defined as a minimum training dose of 5 hours per week, can accelerate functional recovery [46,67,80]
- Difficulty progression, which consists in progressively increasing the difficulty of the task while adapting it to the learner, avoids boredom [81]. Moreover, it exists a strong association between challenge (appropriate difficulty level) and motivation [73]
- Motivation is a key factors for learning. A high level of motivation both increases activity capacities and children's participation, thus keeping adherence to treatment [82]. Main elements of motivation include appropriately challenging tasks, variable practice, setting realistic goals transferable to daily life, and aspects of competition such as a reward system [83]
- Task-specific, context focus and goal-directed therapy, in which tasks incorporated functionally meaningful movements that promoted activity and participation in their daily lives, maximizes the learning process and facilitated their generalization and transfer [84]
- Family support has been recently introduced in the conception of the rehabilitation program, strongly encouraged by the "F-Words": Fitness, Function, Family, Friends, Fun, Future. Family support has not been evaluated but the international clinical practice guideline recommended as an important supplement to clinicians-delivered intervention, especially when interventions were home-based [79]

Finally, Feedback plays a crucial role to enhance motor learning and motivation level [75].
 We will develop this topic in the next subsection 4.2



Figure 2.8 – Ingredients for Motor Rehabilitation. Each ingredient helps to optimize acquisition, retention, generalization and transfer of motor skills. This is an original figure.

At this stage, and after brainstorming sessions, we have decided to explore the possibility of developing a video game to improve gait rehabilitation. First, we have hypothesized that video games can be a solution to match the motor learning ingredients. It allows maintaining a high level of motivation during a long rehabilitation period, provides high intensity for exercises, be goaldirected, and adapts to the difficulty progression. Second, we have been impressed by the Pokemon Go effect on walking performance. In December 2018, players collectively walked 4.6 billion miles for catching Pokemons. And third, we have been inspired by great results from Cho et al. assessing the impact of treadmill training with virtual reality on gait, balance, and gross motor function in children with CP [44] and recent work on biofeedback [85]. We hypothesize that gamification for rehabilitation using technologies, such as virtual or augmented reality systems, could be a suitable method, providing the user with goals, challenges, interactive and funny experiences.

## 2.6 Playing Video Game to Learn, are you Serious ?

A brief overview of the current video-game use has taught us that 84% of teens say they have or have access to a game console at home (girls: 75% and boys: 92%), and 90% say they play video games of any kind whether, on a computer, game console or cellphone (girls: 83% and boys: 92%) [86]. In France, video gamers play 56 minutes per day on average [87]. More interestingly, 25% of those who have played video games believe that video games have helped them develop good problem-solving and strategic thinking skills [88].

So, is a serious game an oxymoron? Can we learn with video games? Can we use this growing interest in games to promote motor rehabilitation? In this section, we will explore the definition and concept of **play**, **game** and **gamification**. We will investigate the terms **serious** 

game, active video game and exergame. Finally, we will overview the active video games used in pediatric motor rehabilitation.

## 2.6.1 Game Theory

In English, the two terms **play** and **game** coexist. In French, there is only one word: "jeu". The Latin etymology is  $l\bar{u}dus$  that means "jeu, amusement" (game, amusement), "bagatelle, enfantillage" (trifle, childishness) but also "école" (school). The second Latin etymology is  $j\bar{o}cus$ : "plaisanterie, badinage" (jesting or playfulness, joke or crack or jest). It is very interesting to observe that the words "game" and "school" have the same Latin root,  $l\bar{u}dus$ .

What does **play** and **game** means ? Basically, we could answer that a game is something you play. Susanna Millar defines **play** as: "any purposeful mental or physical activity performed either individually or group-wise in leisure time or at work for enjoyment, relaxation, and satisfaction of real-time or long term needs" [89]. The international Play Association defines that **play** is communication and expression, combining thought and action; it gives satisfaction and a feeling of achievement. It is instinctive, voluntary, and spontaneous. It is a means of learning to live, not a mere passing of time [90].

The French sociologist Roger Caillois argued that we can summarize the complexity of games by referring to four *play forms* (Agon (or competition) and Alea (or chance), Mimicry (or role playing) and Ilinx (or vertigo) and two opposing types of play (ludus from Latin term  $l\bar{u}dus$  and paidia from Greek Ancient paîs, the child) [91]. He defined ludus as a taste for difficulty, a tendency to set rules and impose conventions in order to reduce the uncertainty of the effect of an activity and to push it toward a specific goal. Ex. sports competitions, theater or alpinism. He defined paedia as a primary power of improvisation and joy, fantasy without rules. Ex. improvised races, childish imitations, merry-go-rounds. For Caillois, play existed on a spectrum from ludus or **game** to paidia or **free play** [91]. To summarize, **game** can be defined as follows:

- (a) "A game is an interactive structure of endogenous meaning that requires players to struggle toward a goal" [92]
- (b) "A game is a closed, formal system, that engages players in structured conflict, and resolves in an unequal outcome" [93]

From these definitions, Jesse Schell has extracted some essential characteristics of **games** [94] displayed in Figure 2.9.

To define the concept of **play**, Jesse Schell has proposed to think more about the player's true motivations not just the goals of the game because play involves willful action. He called this characteristic the lens of curiosity. For him *"play is manipulation that indulges curiosity"*. He finally proposed an elegant and interesting definition combining the concept of **game** and **play** [94]:

"A game is a problem-solving activity, approached with a playful attitude".



Figure 2.9 – Characteristics of games. To better define 'games', Jesse Schell have listed the main common features of games. This figure is an original production based on the Jesse Schell's book 'The Art of Game design' [94]

## 2.6.2 The Role of Play

Plato philosophised that reinforcing certain behaviours exhibited in play would reinforce those behaviours as an adult. The psychologist Jean Piaget said that "play is the work of children". He argued that play affords the consolidation of existing skills through repetition, as well as developing a sense of mastery. In the 19<sup>th</sup> century, the theorist David Cohen argues that "[if children] used some of that freedom to play, then play had to have some purpose". Since 1989, The Office of the High Commissioner for Human Rights recognizes the right of the child "to rest and leisure, to engage in play and recreational activities appropriate to the age of the child and to participate freely in cultural life and the arts" (Article 31 - the United Nations Convention on the Rights of the Child) [95]. One of the arguments advanced is that play helps children develop physically, mentally, emotionally and socially.

## 2.6.3 Games used with therapeutic purposes

Serious Game (SG) appears to be a contemporary manifestation of centuries-old theories and practices, and this is the best illustration of moving from free-play to purposeful rule-based games argued by Caillois [91,96]. The term Serious Games can be traced to the seminal work of Clark Abt [97]. In the 1970s, he defined **serious games** as "games (that) have an explicit and carefully thought-out educational purpose and are not intended to be played primarily for amusement. It does not mean that serious games are not, or should not be, entertaining". The consensus has come with David Michael's definition: "[Serious games are] games that do not have entertainment, enjoyment, or fun as their primary purpose" [98]. Nowadays, with the rising popularity of video games, we have observed that the current uses of the term SG often imply a digital form. The fields of SG application may belong to economics, commercials (advertising), education, training, health, industry, military, and politics [99]. In this context, **Gamification** is the process that aims to influence the user's behavior and motivation by adding game mechanics, game elements, and game experience design in contexts that are initially utilitarian or serious. Recently, Sardi et al. have identified the game elements employed in the digital healthcare domain (i.e., doing a therapy exercise a serious game): feedback/rewards (94% of studies investigated), progression (43%), social connection (37%), challenges/quests (26%), others (game currency, prizes) [100]. He also found that the most recurrently studied health topic for SG in healthcare is chronic disease management and rehabilitation [100]. To conclude, we propose a synthesis of definitions for the term Serious Game [94,99,101] :

## Serious Game

A game, often played using a digital device including specific rules, that uses game elements and game mechanics to further serious objectives, such as industry training, education, health, public policy and, strategic communication.

During the last International Conference of Virtual Rehabilitation in July 2021, a symposium has been organized to discuss about the Unifying Terminology for "Virtual Rehabilitation, Virtual Reality and Video Games". The International Society for Virtual Rehabilitation has proposed to distinguish two categories of SG used in Rehabilitation: Active Video Games (AVG) and Exergames.

## Active Video Game

Video games that are used for sensorimotor (upper and lower limb use, posture, gait) and cognitive rehabilitation across the body function structure, activity and participation domains of the ICF.

## Exergames

Games that use exercise for promoting health and wellness as well as fitness.

Serious Game, Active Video Game and Exergame can be displayed using different devices, that could include virtual/augmented/mixed reality systems. Recently, Huygellier et al. [102] have developed a taxonomy of mixed reality rehabilitation systems using Milgram's virtual continuum from real environment to virtual environment [103] (Figure 2.10).

On the left side of this taxonomy, there is **Augmented reality** (AR) that is an interactive experience where the objects in the real world are enhanced by computer-generated perceptual information, sometimes across multiple sensory modalities, including visual, auditory, (and more rarely haptic, somatosensory and olfactory) [104]. AR systems incorporate three basic features: a combination of real and virtual worlds, real-time interaction, and accurate 3D registration of virtual and real objects [105]. AR is used to enhance natural environments or situations and offer perceptually enriched experiences. In this way, AR alters one's ongoing perception of a real-world environment, whereas VR completely replaces the user's real-world environment with a simulated one. On the opposite side, there is Virtual reality (VR) is a simulated experience including images, sounds and other sensations that immerses the user in a virtual environment (real or imagined) in a way that allows the user to have a sense of being present in that environment, and allows to interact with it [104, 106]. VR is deployed on devices such as virtual reality headsets (e.g. HTC Vive Pro, Oculus Quest, Sony PlayStation VR, Lenovo Mirage Solo) or multi-projected systems (e.g. Cave Automatic Virtual Environment). And because the taxonomy is a continuum, there is a middle part Augmented VR (also called Mixed reality), that is the merging of real and virtual worlds to produce new environments and visualizations, where physical and digital objects co-exist and interact in real time. Mixed reality does not exclusively take place in either the physical world or virtual world, but is a hybrid of reality and VR [103].

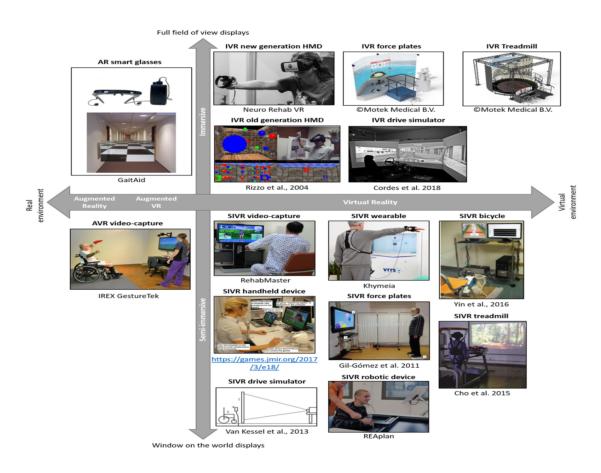


Figure 2.10 – **Taxonomy of mixed reality rehabilitation systems.** It is based on the extent to which real and virtual information is mixed, the level of immersion and the main input device. AVR: augmented virtual reality; IVR: immersive virtual reality; SIVR: semi-immersive virtual reality; HMD: head-mounted display. *From Huyguellier et al.* [102]

## 2.6.4 Active video game in Paediatrics Motor Rehabilitation

In the last decades, significant technological advances such as the miniaturization of external sensors, wearable systems, VR/AR headsets, or smartphones have contributed to the rise of gamification in many industries, especially in healthcare. For example, to promote fitness and physical activity by playing exergame, the Virtual Reality Institute of Health and Exercise<sup>‡</sup> evaluates energy expenditure when playing different commercial video games. Thus, they observed an energy expenditure of 6.55 to 7.45 kcals per minute during tests with the VR Game "Beat Saber" <sup>§</sup>, equivalent of playing tennis.

SG and AVG are used to motivate, engage patients in the therapy, increase treatment adherence, improve well-being, and promote activity. There is a lot of SG and AVG used and validated in pediatrics [107], for educational therapy [108], diversion during painful care [109], physical activity [110] and awareness of risky behaviours. Moreover, it can decrease the inequity of access to healthcare resources and reduce the healthcare costs [111].

We use Huyguellier's Taxonomy of mixed reality rehabilitation systems (Figure 2.10) to describe the AVG used in Paediatrics Motor Rehabilitation.

In a recent systematic review, Lopes et al. have investigated the role of both commercialoff-the-shelf games and customized AVG for motor rehabilitation. Of the 13 interventions involving children with CP, seven were classified as home-based interventions and six as clinical-setting interventions. These studies aimed to improve upper-limb function (46%), to promote motor performance (23%), functional mobility (15%) or balance (15%). The authors concluded that most of the studies reported high levels of compliance, motivation, and overall engagement with the game-based therapy, even at home with family support. However, they observed mixed results related to the efficacy of the intervention in improving the targeted skill. Still, the majority reported some therapeutic gains, but the results were not always significant against the control group. They recommended that these game-based interventions should be used as a complement to the conventional therapies, not as a substitute, and should be based on clear predefined therapeutic objectives [112].

In the same way, a systematic review found that most studies exhibited significant improvement after the game-based intervention, but not more effective than standard care [113]. Lower-limb gross motor function and walking capacity were significantly improved after AVG training, but there were no improvements in stair climbing ability or agility performance [114]. The authors highlighted the critical variation in game-based intervention dosages, ranging from 10 minutes to 1 hour, less than once per week to 5 days per week, and for a duration of 3 to 24 weeks [113].

In another systematic review of the use of AVG in rehabilitation for children with CP, 5/7 RCT found a difference between an AVG intervention compared to a control group for muscle strength, balance, motivation and participation level, motor performance (e.g., moving an object) and bone density [115].

<sup>‡.</sup> More information available on the website https://vrhealth.institute/

<sup>§.</sup> Demo https://store.steampowered.com/app/620980/Beat\_Saber/

In a home-based intervention, Levac et al. [116] have included 11 children with CP divided into 2 groups: 5 children received 1 hour of IREX (AVR video capture) system training  $\P$  for five days followed by a 6-week home AVG program, supervised online by a physical therapist, and six children completed only the 6-week home AVG program. The AVG program consisted of 30 min/5 days a week expected session with commercial-off-the-shelf AVG (Kinect Xbox 360). The games selected were sport-based, incorporating full-body movement. There were no significant differences between groups for the 6MWT and the Gross Motor Function Measure Challenge Module (GMFM-CM). The home AVG-only group demonstrated a statistically and clinically meaningful improvement in GMFM-CM scores following the 6-week AVG intervention (median difference 4.5 points, IQR 4.75). The IREX + AVG group demonstrated a statistically and clinically significant decrease in 6MWT distance following the intervention (median decrease 68.2 m, IQR 39.7 m). The authors concluded that neither intervention improved outcomes. Regarding the ingredients for effective motor learning exposed in 2.5, we think that other issues also need to be considered. In this study, the AVG-only group played an average of 42.1 min (SD 4.9 min) per day, an average of 14.71 minutes more per day than the IREX + AVG group (mean 27.4 min, SD 1.4 min). This dosage, even if this is more in the first group, didn't match the minimal recommended intensity of practice of 5 hours/week [73]. Moreover, the author admitted that the online video supervision by therapists did not provide good results. However, it has been demonstrated that home-based intervention required a strong implication of families and the necessary follow-up by health care professionals [75]. Second, AVG displayed during this program was not explicitly designed for children with CP. It could have induced some trouble in their use. The authors also suggested a better-individualized exercise challenge to match the difficulty progression principle. Third, playing games as Just Dance was not goal-oriented or relevant for activities of daily living. Lastly, the games did not provide real-time feedback about their walking performance but only about their success in the game. Improving level in the game doesn't necessarily mean an improvement in the 6MWT, which assesses functional walking capacity.

In another study, Sandlund et al. have explored the feasibility of using low-cost motion AVG as a home-based intervention for children with CP (EyeToy for PlayStation 2). The program was a four-week intervention with at least 20 min/day of practice. Participants did not significantly improve their 1-minute walk test [117]. Again, that can be explained because the intervention did not incorporate the motor learning ingredients: 20 min per day was not enough, and playing with EyeToy did not include the specific task of walking (therapy was not goal-directed and task-specific).

In a large RCT, Salem et al. included 40 children. The intervention group practiced two weekly sessions for ten weeks using Nintendo Wii Sports and Nintendo Wii Fit (only balance, strength training, and aerobics games). Even if they noticed trends toward more significant gains in the experimental group compared to the control group, they did not find a significant difference between groups [118]. Once again, the main ingredients for effective motor rehabilitation were not fulfilled.

Clinical-setting interventions seemed to provide better but mitigated results. For example, Cho et al. have compared two groups of children with CP: the AVG-group performed treadmill gait

<sup>¶.</sup> The IREX, short for Immersive Rehabilitation Exercise, uses a camera connected to a computer, to place the live real-time, full-body image of the patient onto the screen, where they see themselves immersed in the AVG.

training connected to an AVG (commercial-off-the-sheld Nintendo Wii Fit Plus) vs. the control group performed gait training on a treadmill without the application of AVG (Figure 2.11). Each participant of the AVG-group was treated for 30 minutes per day (5 min warm-up, 10 min of exercise, 5 min rest, 10 min of exercise) 3 times per week for a total of 8 weeks. In the field of walking, running, and jumping, gross motor function increased significantly from 52.7 to 57.9 in the AVG-group and from 47.1 to 51.2 in the control group after training (no difference between groups). The distance traveled in the 2MWT increased significantly, from 54.83 to 116.07 m in the AVG-group and from 72.87 to 88.87 m in the control group. The change in the 2MWT was considerably more remarkable in the AVG-group than in the control group [44]. Unlike the previous study, the skills acquired during walking training on the treadmill were transferred to natural walking environments. Moreover, walking speed and walking endurance improved to greater extents in the AVG-group than the control group. We hypothesize that task-oriented gait training improves patients' motor function with neurological impairments. Moreover, AVG provides optimal flow that encourages/motivates patients to walk with more intensity.



Figure 2.11 – Example of treadmill training in virtual environment using commercialoff-the-sheld Nintendo Wii Fit Plus. According to the Huyguellier's taxonomy of MR rehabilitation systems, this is classified as Semi-Immersive Virtual Reality treadmill. *From Cho et al.* [44]

In another study, Wu et al. have evaluated the use of a robot-assisted ankle movement in ambulatory children with CP. After six weeks with three sessions/week, a significant improvement was found in the 6MWT [119]. High intensity through repetition of the same movement probably promoted transfer between active ankle movement and walking ability.

To summarize, as we report in the red box below, the potential benefits of AVG are essential, BUT motor learning ingredients should not be ignored in Motor Rehabilitation using AVG in AR/VR. The game interest, which raises the child's level of motivation for rehabilitation, is not sufficient to improve motor skills. To be efficient, game-based interventions should integrate motor learning ingredients such as appropriate dosage (high intensity), variable practice, difficulty progression, task-specific exercises, feedback in real-time and with delay, motivation of the patient, context-focused and goal-directed therapy (Section 2.5).

The potential benefits of Active Video Game

- Increase intensity by repetition and consistent practice of the same task
- Individualize difficulty and difficulty progression
- Record and analysis performance outcomes
- Provide a safe environment to undertake tasks which may be difficult/unsafe in real life
- Offer appealing games that may make therapy tasks more fun and engaging, which may increase compliance with therapy
- Enhance motivation which may lead to longer practice duration and more practice repetitions
- Provide augmented feedback about task performance or task results
- Expand therapy at home including family

In addition, we have identified, from three systematic reviews focusing on AVG used in children with neuro-motor impairments, 10 additional and complementary recommendations to promote motor rehabilitation in AR/VR presented in the blue box below [112, 113, 115] (See Subsection 2.6.4 for more details).

## Additional Guideline for Motor Rehabilitation in AR/VR

- $\checkmark$  Goals should be pre-defined AND well defined
- ✓ Goals and dosage should be discussed with all the professional team AND the child AND the family (in particular for home-based therapy)
- $\checkmark$  Short intensive interventions or massed practice should be preferred
- $\checkmark$  A high enough dose of practice must be undertaken to achieve functional goals
- $\checkmark$  The difficulty must be progressive and adapted to the child's abilities
- $\checkmark$  The transfer of skills to the real life should be anticipated
- $\checkmark$  Prefer games created specifically for the population of interest
- $\checkmark$  Children with severe motor impairment should also benefit from motor rehabilitation in AR/VR
- $\checkmark$  Virtual reality rehabilitation should be considered as a complement, not a substitute
- $\checkmark$  Use adaptive and mixed feedback modalities depending on the task complexity

These recommendations were not always integrated into virtual reality interventions, as we have seen previously and as confirmed by Demers et al. in 2020 [73]. They analyzed the key components of motor learning in commercial video game platforms and custom virtual reality systems. All studies provided continuous feedback but essentially visual feedback. Macintosh et al. highlighted that most studies provided feedback consistently and concurrently throughout the intervention regardless of the individual's desire or progress contrary to recommendations [85]. Most of the systems delivered variable practice and used functionally relevant and motivating virtual tasks. The main negative point was the dosage, which was variable and often not intensive enough, with low repetition of the gesture or task. In the same way, the difficulty progression and the assessment of skills retention and transfer were poorly incorporated, particularly for commercial video games. Finally, they concluded that motor learning ingredients should be better integrated into the development of future virtual reality systems for optimal motor recovery in children with CP [73]. We must keep in mind the recommendations that we outlined above to better design our future active video game.

Moreover, the environment's choice was crucial: AR, VR, MR? Our decision was made early and was based on two main arguments. First, children must be free to move in the global environment without the restriction of movement. In the systematic review, Booth et al. concluded that functional overground gait training is a safe, feasible, and effective intervention to improve walking capacity for children and young adults with CP (especially walking speed) [49]. Second, children must use their usual walking aids (crutches or posterior walkers). Under these considerations and taking into consideration each system's comparative advantage, AR has been preferred to VR. (Figure 2.12) The choice of the AR device will be more detailed in Chapter 3.



Figure 2.12 – **Overground Gait Training in AR vs. Treadmill Gait Training in VR.** On the left, overground gait training with an AR headset (extract of the ARRoW-CP AVG), child is free to walk in the real environment using her/his walking aids. On the right, treadmill gait training with the Motek GRAIL system with body-weight support and walking constraints due to the treadmill

## 2.7 Conclusion

Cerebral palsy is the most common motor disability of childhood. The clinical picture is at the same time very diverse between each individual, but for the same individual, it evolves during his life. Various manifestations of the brain lesion may appear more significant in different persons or at different life periods, causing activity limitation and participation restriction. This is due, in particular, to the individual's natural growth, treatments, care, and the family and educational environment.

Gait disorders are an essential part of this clinical picture. 1 out of 3 patients will never walk. Among walking children, a quarter of them will experience a decrease in their walking capacity in adulthood. During growth, children with CP develop secondary musculoskeletal and bone disorders, such as muscle-tendon contractures or torsion in long bones, caused by multiple factors. In recent years, considerable progress has been made in diagnosis, clinical and instrumental assessment (use of standardized and validated tools such as CGA), and care (development of the early intervention, intensive therapeutic strategies, surgery...).

Among the validated treatments, SEMLS is indicated to improve passive range of motion, kinetics and kinematics gait parameters, and overall gait index. However, spatiotemporal gait parameters, motor function, and quality of life remain mixed. Several authors suggest that the benefits of this surgery can be better if rehabilitation care (both pre and post-operative) is optimized. Our state of the art defines five critical steps in this rehabilitation after SEMLS. One step, in particular, seems optimal for introducing new therapeutic strategies: intensification (step 4).

According to the literature, this intensification strategy must be based on motor learning principles, among them appropriate dosage (high intensity), difficulty progression, variable practice, task-specific exercises, context-focused and goal-directed therapy, motivation, and feedback. A way to intensify gait reeducation without causing boredom, without putting a lot of time pressure on the therapists and while making the children happy, and that can easily integrate motor learning principles could be: Active Video Game.

Active video games and exergames are emerging today in motor rehabilitation thanks to the miniaturization of devices and the rise of virtual reality and augmented reality. But there are many ways to deliver these game therapies. The literature reports that not all games and protocols tested are effective, far from it. Today, there is an extended mix, source of misunderstanding, between commercial games diverted for use in therapy but not always adapted to children with disabilities, and especially not developed taking into account the essential ingredients of motor learning and motor rehabilitation. In our context, augmented reality has been preferred to virtual reality so that children feel free to walk in their actual environment, without restriction of movement, using their usual walking aids.

Therefore, this thesis aims to develop an active video game in AR including most of the ingredients of motor learning, adapted and individualized for each child, and co-developed with therapists and children with a unique objective: to improve overground gait rehabilitation. We hypothesize that a play-based AR approach would enhance the intensification step of post-SEMLS rehabilitation, leading to an overall improvement of the child's walking capacities.

## Chapter 3

# Using an Augmented Reality Headset for Gait Rehabilitation

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Chapter 3 investigates the feasibility of using an AR headset as a unique sensor and device for gait recognition in a real (overground) environment. First, we present the experiment assessing the reliability of the device that we have selected (Microsoft Hololens AR headset) (Article 2). In our context, the Hololens is a sufficiently reliable tool to measure in real-time the position of the participants' head during different walking situations, in comparison with a motion capture system. Second, we detail our step detection algorithm (HoloStep - open source code), accurately measuring the spatiotemporal gait parameters, without additional sensors, from the head's position. We present its experimental validation with a population of children with CP (Article 3).

## 3.1 Introduction

In the previous chapter, we identified that the package SEMLS and Rehabilitation on functional capacities' clinical results are mixed. The literature review suggested improving rehabilitation after SEMLS, especially Step 4 (i.e., Intensification). There was also the question of keeping children motivated and focused on achieving their goals over such a long period. What do children love more? PLAY! So, the preferred choice was developing an active video game focusing on this fourth phase of intensive gait rehabilitation. The emergence of active video games used for motor rehabilitation purposes, as well as the growing development of new technologies, allow us today to consider their use in a post-operative context for children with motor disabilities.

The device's choice was crucial and implied a prerequisite that we will detail in Section 3.2. An essential point of this thesis was to evaluate the reliability of the calculation of the tridimensional position of the AR Headset. We had to be sure that the AR headset was sufficiently accurate to estimate the user's position and displacement in the global environment. After that, we developed an algorithm to measure the spatiotemporal gait parameters only with the AR Headset. This constraint corresponds to the recommendations of health professionals who require a standalone tool (or an all-in-one tool).

In this third chapter, we will overview the main characteristics of the AR Microsoft Hololens Headset (Section 3.3). We will present the experimental study assessing the accuracy of the AR Headset (Section 3.4), and we will expose the HoloStep algorithm that we have developed to measure spatiotemporal gait parameters with the AR headset (Section 3.5).

## 3.2 Necessary Prerequisite

Before rushing towards the game development, we selected the best device. The involvement of the multidisciplinary team has led to a discussion on the technological device to be used. The team identified four crucial technical aspects. First, children must be free to move in the global environment without the restriction of movement. In the systematic review, Booth et al. concluded that functional overground gait training is a safe, feasible, and effective intervention to improve walking ability for children and young adults with CP (especially walking speed) [49]. Second, children must use their usual walking aids (crutches or posterior walkers). Thirdly, the device must be accurate to calculate spatiotemporal gait parameters (speed, cadence, step length) [44]. Fourth point, more technical constraint, this device should offer ample possibilities in software customization, providing technical documentation, SDKs, and dedicated development support. These four considerations impacted the choice of technological device.

To summarize, the device should be adaptable for overground gait training rehabilitation, hands-free, be compatible with exercises including variable intensity, and be well-documented. The Microsoft Hololens AR Headset has been selected because its technical characteristics matched the specification.

As soon as the project team validated the choice of the tool, we conducted experiments with two children from the Fondation Poidatz. Neither of them had experienced Microsoft Hololens headset before. The goal was to verify the children's behavior with the Hololens, as a device, and with the holographic content. After explaining augmented reality, we put the headset on their heads without turning it on. We asked the children about the headset's comfort, and we studied their motor behavior while they were wearing the headset without turning it on. Then we proposed to children to play the game RoboRaid developed by Microsoft Corporation, available for free on Windows Store. It is a mixed reality first-person shooter, where the player defends his home from a robot invasion. These robot invaders break through the walls; the player must move to avoid enemy fire and blast their foes using his gaze, gesture, and voice. After a time of adaptation, we studied their motor behavior. In particular, we evaluated the risks of falling, the imbalance, the safety, the interaction with the holograms, the comprehension, and the enjoyment. We noted all their reactions during the session. Neither child did anything dangerous with the headset off or during play. No falls, no imbalances. On the contrary, the necessary "evasion" during the robot attacks allowed them to explore new motor behaviors (e.g., squat down quickly, lateral body translation). They quickly understood the game's purpose and were able to interact appropriately with the holograms. They were both satisfied with the experience. The only negative point was the headset's weight on the nose, which made them feel uncomfortable after 15 minutes of play. This problem has been solved by adding the overhead straps provided by Microsoft. This experiment was crucial to adopt the Microsoft Hololens headset for the next steps of the ARRoW-CP project.

## 3.3 Augmented Reality Headset characteristics

## 3.3.1 Basic Specifications

Microsoft HoloLens (1<sup>st</sup> generation) is the world's first commercial fully untethered holographic computer, using Gaze tracking, Gesture input and Voice support to understand user actions (Figure 3.1). And Spatial sound to understand the environment. It's an Augmented Reality Head-Mounted Display of 579 grams. The Microsoft website provided complete specifications of the Hololens (https://docs.microsoft.com/en-us/hololens/).

Bluetooth can be used to talk peer to peer between multiple HoloLens if the customers' application supports it or to other Bluetooth devices. The autonomy of the HoloLens is estimated to 2-3 hours of active use and up to two weeks of standby time. The battery lifetime is unavailable. The FCC ID for HoloLens is C3K1688. An FCC ID is the product ID assigned by the FCC to identify wireless products in the market. HoloLens has been tested and found to conform to the basic impact protection requirements of ANSI Z87.1, CSA Z94.3, and EN 166. HoloLens (1st gen) supports device encryption using BitLocker.

HoloLens (1st gen) has entered Long Term Servicing (LTS) state. For developers, this means that HoloLens (1st gen) apps will not support the OpenXR API. These headsets remain supported in Unity 2019 LTS. All the applications that we have developed are compatible with the Hololens 2.

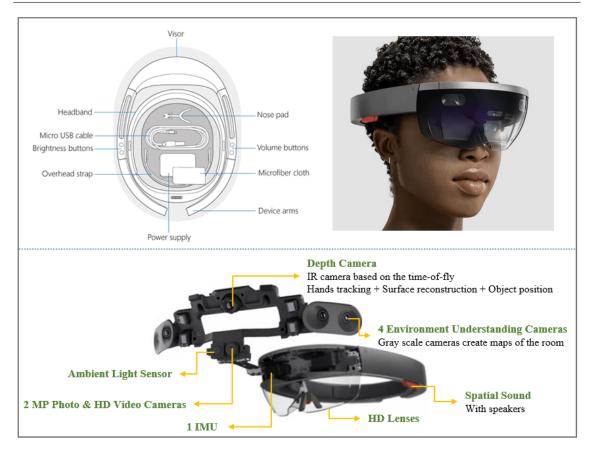


Figure 3.1 – Microsoft Hololens AR Headset. Top left: Basic features and accessories. Top right: Woman wearing the Microsoft Hololens  $(1^{st}$  generation) used in our work. Bottom: Specifications of the Hololens

## Hololens Specifications Displays

**Processor** Intel 32-bit architecture with TPM 2.0 with Custom-built Microsoft Holographic Processing Unit (HPU 1.0).

Memory 64 GB Flash with 2 GB RAM.

**Optics** See-through holographic lenses (waveguides).

Holographic resolution 2 HD 16:9 light engines producing 2.3M total light points.

Holographic density >2.5k radiants (light points per radian).

Eye-based rendering Automatic pupillary distance calibration.

## Hololens Specifications Sensors & Connectivity

1 inertial measurement unit (IMU)

4 environment understanding cameras; 1 depth camera

2MP photo / HD video camera; 4 microphones; 1 ambient light sensor

Built-in speakers; Audio 3.5mm jack; Volume up/down; Brightness up/down; Power button; Battery status LEDs; Wi-Fi 802.11ac; Micro USB 2.0; Bluetooth 4.1 LE

## 3.3.2 Examples of applications

HoloLens gets online software applications through the Windows store. The easiest way to develop an app for Hololens is using Unity and the Mixed Reality Toolkit (MRTK). MRTK-Unity is a Microsoft-driven project that provides a set of components and features, used to accelerate cross-platform MR app development in Unity. Some applications are presented in Figure 3.2.



Figure 3.2 – **Examples of Hololens Applications.** Hololens Applications have been developed in the field of industry (bottom left), for surgical assistance (top left), for teaching (top right), for entertainment (gaming or watching sports)

## 3.4 Reliability of the head tracking with an Augmented Reality Headset during different walking conditions (Article 2)

The first step to developing a relevant application for gait rehabilitation, using position, velocity, and acceleration of the headset, was to assess the reliability of the headset tracking, in comparison with a reference motion analysis tracking system, during different conditions. We hypothesize that the accuracy of the Hololens is sufficient to measure the position and the velocity of the headset, and by extension, the user, without spatiotemporal drift.

During nine walking trials in 3 different conditions, the 3D raw coordinates of the Hololens were measured simultaneously by the HoloLens and a VICON motion capture system. An Iterative Closest Point algorithm was used to align the 3D positions given by the two systems minimizing the distance between them, using geometric transformations (rotations R and translations T). Pearson's correlation coefficient and root mean squared error (RMSE) between the two systems were calculated.

Minimal errors between signals were about 6 to 27 mm. Maximal errors were 55 to 250 mm. and concerned the antero-posterior axis. RMSE was from 9 to 53 mm. Speed variation between the different sets didn't influence the accuracy of the Hololens tracking. There was no signal drift over time.

In conclusion, the accuracy of the Hololens is sufficiently high to evaluate the position and the velocity of the AR headset, and the user by extension, without time drift.

We have shown that the Hololens allows strong and comprehensive tracking capabilities. It could be used as a tracking tool for the child's position while she/he is walking. The possible use of the AR headset in the "unlimited" real environment is of significant interest. The child can walk in the rehabilitation center without constraint while she/he is playing the game. The next step of our work is to develop an algorithm to extract the spatiotemporal parameters from the AR headset (Section 3.5).

This paper is attached to this thesis manuscript as published in *Computer Methods in Biomechanics and Biomedical Engineering* in 2019 [120]. This work was presented during the *Société de Biomécanique Congress* in October 2019.

## Reliability of the head tracking measured by Microsoft Hololens during different walking conditions

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Keywords: motion capture; biomechanics; headset tracking; augmented reality

## 1. Introduction

Augmented reality (AR) is a technology that expand the real environment by adding digital holograms into it. AR appears to be a promising field of development for serious games, especially for walking rehabilitation applications. The HoloLens device is one of the most popular and advanced AR Head Mounted Display (HMD). It includes optical and inertial sensors for position and orientation tracking. Although the algorithm combining the sensors information in the HoloLens can estimate the headset's pose in an absolute reference frame, its reliability in walking condition is undocumented. Yet, the first step to high realistic application for gait develop rehabilitation, using position, velocity and acceleration of the headset, is to assess the reliability of the headset tracking, in comparison with a reference motion analysis tracking system, during different conditions. We hypothesis that the accuracy of the Hololens is sufficient to measure the position and the velocity of the headset, and the user by extension, without spatiotemporal drift.

## 2. Methods

#### 2.1 Data collection

During 9 walking trials in 3 different conditions the 3D raw coordinates of the HMD were measured simultaneously by the HoloLens and a VICON motion capture system (MOCAP) both at 100Hz :

Condition 1 - "L-shaped" walking path (5 x 2,5m) at three different speeds (self-selected, low and fast). Three repetitions.

Condition 2 – random walking path during 2 minutes. Three repetitions.

Condition 3 – same as condition 1 with intercalated 2 minutes walking exits outside off the laboratory between each repetition.

The Hololens tracking was performed with an application created with Unity. This app contained a script which logged the position of the HMD and Spatial Mapping prefab which provided a detailed representation of real-world surfaces in the environment around the HoloLens. A MOCAP system was used to track 5 retro-reflective markers (19 mm) attached to the headset, with 15 infrared cameras.

#### 2.2 Data analysis

The Hololens signals were synchronized to the MOCAP signals by an automatic time shifting using a local minimum detection. An ICP algorithm (Besl & McKay 1992; Chen & Medioni 1992) was used to align the 3D positions given by the two systems minimizing the distance between them, using geometric transformations (rotations R and translations T). The ICP algorithm has two steps: The first step consists of determining the correspondence pairs (p, m) from two data sets H and G. The aim is to find for each point p in H its closest point in G. The second step is to apply a transformation (R and T) in order to minimize the distance between the correspondence pairs:

$$E(R,t) = \frac{1}{N_H} \sum_{i=1}^{N_H} ||G_i - RH_i - t||^2$$

With  $H{xH yH zH}$  and  $G{xG yG zG}$  the corresponding points, from Hololens and from MOCAP respectively. These two steps are repeated until the error is below a given threshold or until the maximum number of iterations is reached.

Before components and norm velocities computations, trajectories of both devices were low pass filtered with a fourth-order Butterworth filter with a cut-off of 6 Hz. The correlation between the position and the velocity of the headset given by the Hololens and the MOCAP system was calculated with Pearson's correlation coefficient. The distances between the Hololens and the MOCAP measured parameters were computed by root mean squared error (RMSE).

## 3. Results and discussion

The ICP algorithm permitted to compare a headset position data from Hololens with a model shape gived by MOCAP system, after the shape was moved (registered, positioned) to be in best alignment (Figure

1). For trials 1 to 3, realized in condition 1, correlation between  $H_i \{ x H_i \ y H_i \ z H_i \}$ and coefficients  $G_i \{ xG_i \ yG_i \ zG_i \}$  were quite close to 1. Minimal errors between signals were about 10<sup>-2</sup> to 10<sup>-4</sup>. Maximal errors were of 55 to 250 mm. Maximal errors concerned antero-posterior axis. RMSE was from 9 to 53 mm. Speed variation between the different sets didn't influence the accuracy of the Hololens tracking. There was no signal drift over the time. The error increased when there was a sudden shift of trajectory. Results obtained in condition 2 were similar. For trials 7 to 9, realized in condition 3, signal processing was more complex. Indeed, for each trial, the first laboratory exit systematically generated a "jump" on  $xH_{exit}$  and  $yH_{exit}$ , for no apparent reason. For example, in trial 9, there was a drift of 0,45 m (mediolateral axis) and 40 m (antero-post axis). No drift for vertical axis. However, these calibration movements were realized at the same place inside the laboratory. An hypothsis shoud be the Spatial Mapping function, which could be influence the position and orientation in space. The corridor route was not pre-mapping unlike the laboratory. Nevertheless, after this offset was eliminated, the headset tracking with Hololens presented the same good results than in condition 1 and 2. No signal drift over the time again (Table 1).

		Х	Y	Z
Cond. 1	r	1,00	1,00	0,99
	max Err	98	251	81
	min Err	21	25	6
	mean Err	24	44	6
	RMSE	32	50	9
Cond. 2	r	1,00	1,00	0,99
	max Err	75	117	105
	min Err	13	14	6
	mean Err	14	24	8
	RMSE	19	28	10
Cond. 3	r	1,00	1,00	0,95
	max Err	113	146	55
	min Err	24	27	6
	mean Err	24	42	6
	RMSE	35	53	10
TT 11	1 4	1 1 6	11 . 1 1	• •

Table 1 Averaged results for all trials in each experimental conditions. Errors are given in mm.

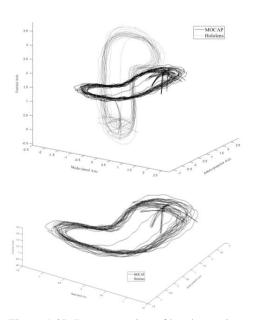


Figure 1 3D Representation of headset trajectory with Hololens and VICON tracking for Trial 1 before ICP (top), and after ICP (bottom)

For each trials, correlation coefficients for the norm of the HMD vector velocity were high (between 0,96 and 0,99) (detailed not presented in this paper).

## 4. Conclusions

The accuracy of the Hololens is sufficiently high to evaluate the position and the velocity of the headset, and the user by extension, without time drift. There was however uncertainty as to the exact drift of the Hololens signal, when the environment changed. This problem, with no obvious explanation, should not stop to develop gait rehabilitation application using position and velocity of the headset given by the Hololens.

## Acknowledgements

The authors thank the Fondation Ellen Poidatz and the SESEP for the support to this research.

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## 3.5 Validity of gait parameters calculation in real time with an Augmented Reality Headset (Article 3)

As we have previously demonstrated, the accuracy of the Hololens is sufficiently high to evaluate the position and the velocity of the headset, and by extension, the user, without time drift (Section 3.4). The next step of our work is to develop an algorithm to extract the spatiotemporal parameters from the AR headset.

The objective of this work was to develop and validate a new algorithm, **HoloStep**, calculating in real-time gait spatiotemporal parameters with the head's position of the user. The HoloStep Computational Method is described in detail in the published article. Briefly, the principle of detection used by HoloStep was based on the fact that during walking, the body displacement is pseudo-periodic. The body slightly leaned to the left/right side. Moreover, at each initial contact (when the foot was touching the ground), the body was lower on the vertical axis. Therefore, when the user was leaning to the left (x min) and the body was at a lower position (y min), the user's left foot started to touch the ground. A similar situation happened on the other side (x max and y min). HoloStep was developed using a combination of locking distance, locking time, and peak amplitude detection with custom thresholds for children with CP. At each frame, HoloStep calculates: Time, Position, Filtered position, step detected (Boolean), Step Length, and Walking distance from the beginning of the trial. We hypothesize that HoloStep is accurate enough to be used in clinical practice to calculate walking speed, step length, and cadence of people with cerebral palsy.

We included thirteen healthy adults and sixty-two children with CP. The inclusion criteria for people with CP were between 10–18 years, GMFCS I to III. Children were divided into two groups: Group 1: Children with CP walking without walking aids (N = 32) (GMFCS I), Group 2: Children with CP walking with crutches or posterior walker (N = 30) (GMFCS II/III).

Participants were instructed to walk at a comfortable speed straight along an 8-m path in the gait lab. Data were collected for three successful trials. The performance of the HoloStep algorithm was compared to the Gold Standard (Zeni algorithm using a set of pelvic and feet markers from a 3D motion capture analysis system). Bland and Altman analysis was performed to compare walking speed, step length, and cadence between the two methods. The Intraclass Correlation Coefficient for absolute agreement among measurements, also known as criterion-referenced reliability, was calculated. The Pearson correlation coefficient r and determination coefficient  $r^2$ were calculated. The confusion matrix allowed visualization of the performance of the HoloStep algorithm.

For the 32 participants from group 1 (GMFCS I), sensitivity, specificity, accuracy, and precision of the HoloStep algorithm were excellent (> 0.964). The Bland and Altman analysis showed a bias d = 0.017 m for step length and 0.018 m/s for walking speed. The ICC coefficients were excellent for step length and walking speed (ICC = 0.922 and 0.996 respectively) and good for cadence (ICC = 0.642). The mean difference for step length between Zeni and HoloStep was 2.6 cm. For the 30 participants from group 2 (GMFCS II-III), sensitivity, specificity, accuracy, and precision of the HoloStep algorithm were excellent (> 0.984). The Bland and Altman analysis showed a bias d = 0.005 m for step length and 0.018 m/s for walking speed. The ICC coefficients were excellent for step length and walking speed (ICC = 0.863 and 0.990 respectively) and good for cadence (ICC = 0.625). The mean difference for step length between Zeni and HoloStep was 4 cm.

In conclusion, we have developed and evaluated a new algorithm called HoloStep to calculate spatiotemporal gait parameters using only the head pose provided by an augmented reality headset (Microsoft Hololens). We have shown that HoloStep is accurate enough to be used in clinical practice compared to the gold standard. We have deployed the HoloStep algorithm in our active video game ARRoW-CP for gait rehabilitation in children with CP (Chapter 5). We can use HoloStep to provide patient feedback on their spatiotemporal parameters through the AR headset. HoloStep algorithm is open-source, and it can be implemented in other active video games for motor rehabilitation.

This paper is attached to this thesis manuscript as published in Sensors in 2021 [121]. This work has been presented during the  $30^{th}$  European Society for Movement Analysis for Adults and Children Congress in October 2021.





## Article Validity of Hololens Augmented Reality Head Mounted Display for Measuring Gait Parameters in Healthy Adults and Children with Cerebral Palsy

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Abstract: Serious games are a promising approach to improve gait rehabilitation for people with gait disorders. Combined with wearable augmented reality headset, serious games for gait rehabilitation in a clinical setting can be envisaged, allowing to evolve in a real environment and provide fun and feedback to enhance patient's motivation. This requires a method to obtain accurate information on the spatiotemporal gait parameters of the playing patient. To this end, we propose a new algorithm called HoloStep that computes spatiotemporal gait parameters using only the head pose provided by an augmented reality headset (Hololens). It is based on the detection of peaks associated to initial contact event, and uses a combination of locking distance, locking time, peak amplitude detection with custom thresholds for children with CP. The performance of HoloStep was compared during a walking session at comfortable speed to Zeni's reference algorithm, which is based on kinematics and a full 3D motion capture system. Our study included 62 children with cerebral palsy (CP), classified according to Gross Motor Function Classification System (GMFCS) between levels I and III, and 13 healthy participants (HP). Metrics such as sensitivity, specificity, accuracy and precision for step detection with HoloStep were above 96%. The Intra-Class Coefficient between steps length calculated with HoloStep and the reference was 0.92 (GMFCS I), 0.86 (GMFCS II/III) and 0.78 (HP). HoloStep demonstrated good performance when applied to a wide range of gait patterns, including children with CP using walking aids. Findings provide important insights for future gait intervention using augmented reality games for children with CP.

**Keywords:** spatiotemporal gait parameters; augmented reality; wearable device; cerebral palsy; concurrent validity

## 1. Introduction

Cerebral Palsy (CP) is the most common cause of childhood disability, affecting 17 million people worldwide [1,2]. CP describes a group of permanent disorders of the development of movement and posture, causing activity limitation, which are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain [3]. Children with CP have different functional abilities, classified from Gross Motor Function Classification System (GMFCS) Level I (can walk and climb stairs without using hands for support) to Level V (impaired in all areas of motor function). In addition, gait in children with CP is characterized by a slower speed, a shorter-step length, a lower cadence, and more time spent in double support [4–6]. The natural history in children with CP is a gradual decline in ambulatory function as children grow and age, in particular during adolescence [7,8].

To reverse this trend, physiotherapists propose overground or treadmill-based gait training, with varying body weight support [9]. Functional gait training includes many



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**Copyright:** © 2021 by the authors. Licensee MDPI, Basel, Switzerland. This article is an open access article distributed under the terms and conditions of the Creative Commons Attribution (CC BY) license (https:// creativecommons.org/licenses/by/ 4.0/). different interventions and level of intensity to improve walking capacity. Grecco et al. highlighted the fact that rigorous gait training protocol, performed at the aerobic threshold with increasing intensity, was the key point to improve endurance, walking speed and daily functional performance [10]. But both for adults and children, with multiple repetition of the same task, the loss of motivation can decrease treatment adherence.

The application of technology in the field of rehabilitation is becoming increasingly popular. Within the range of choices available to therapists and patients, virtual/augmented reality (VR/AR) systems are feasible, effective and have positive effect on compliance with therapy and motivation of the patient [11,12]. These systems promote multi-sensory stimulation and user interaction, and can deliver feedback in real-time that could enhance motor learning and skill acquisition [13,14]. A meta-analysis by Chen et al. based on the ICF (International classification of functioning, disability and health [15]) concluded that VR/AR had a large effect size for the activity component (including ambulation function) for children with CP [16]. For example, after VR treadmill training, the distance travelled in the 2-Minutes Walk Test increased significantly, from 54.83 to 116.07 m in the VR group in children with CP [17].

Thanks to the miniaturization of the devices and the commercial development of affordable products, wearable technology-assisted devices appear to be an interesting tool for gait rehabilitation in overground condition (i.e., gait training without treadmill) [18–21]. Among existing tools, AR technology does not fully immerse the user in a simulated environment but superimpose virtual elements over the real-world. For example, Microsoft Hololens is an AR Head Mounted Display (HMD) which includes optical and inertial sensors for position and orientation tracking. It allows patients to walk with their walking aids and augments real environment with visual and auditory feedback. These features offer the possibility to develop a serious game based on motor learning theories to improve walking rehabilitation with gait pattern recognition.

Today, there is no AR system or serious game designed for overground gait training, including a rigorous protocol based on the patient's walking performance and abilities. One of the essential components of such a system is the real-time tracking of the spatiotemporal gait parameters in people with gait disorders. Several methods exist that use full kinematic captures [22–24] or lighter inertial sensors [25–30]. Two recent works exploit an AR HMD but do not involve children with CP [31,32]. In this paper, we present a new algorithm, called HoloStep, to compute spatiotemporal gait parameters of children with CP using only the head pose recorded with a Hololens AR HMD. It is based on the detection of peaks associated to initial contact event, and uses a combination of locking distance, locking time, peak amplitude detection with custom thresholds for children with CP. We have evaluated its concurrent validity (i.e., between-systems agreement) in healthy adults and in children with CP using different walking aids (GMFCS I-II-III). Gait parameters (speed, step length, cadence, step detection) were compared to a reference algorithm from Zeni [22] which uses pelvic and feet kinematics data extracted from a motion capture system. The objective of this study was therefore to develop and validate an AR HMD based algorithm providing gait spatiotemporal parameters, in a real environment, both in healthy and children with gait disorders. We assume that HoloStep is accurate enough to be used in clinical practice to provide feedback on gait performance to the patient through the AR HMD.

# 2. Related Work

Multiple methods exist for calculating spatiotemporal gait parameters, whether based on force plates or reflective marker systems [22–24]. However, these techniques require expensive equipment and are only applicable in a gait laboratory.

Wearable sensors based on inertial measurement units (IMUs) or accelerometers have been validated in both normal and pathologic gait to detect gait events both in controlled laboratory conditions [25–28] and in real-life behaviour [29,30]. Using tri-axial accelerometer, Zijlstra et al. developed an algorithm predicting spatiotemporal gait parameters with trunk acceleration data for healthy participants, but step length and speed were underestimated both in overground and treadmill condition [25]. Trojaniello et al. have tested five methods for the estimation of gait events and temporal parameters from the acceleration signals of a single IMU. Data were acquired from healthy participants. Some methods estimate step time (i.e., the time between two consecutive initial contacts) and stride time (i.e., one complete gait cycle) to determine spatial parameters such as the step length. Some methods require the determination of both initial contacts (IC) and foot contacts (FC), and two methods associate acceleration signals and physical characteristics of gait to identify gait events [27]. They concluded that all methods are acceptable for clinical use (mean error in estimating stride time and step time are non-significant, maximal percentage of error is 2 to 4% for stride time and 2 to 8% for step time). McCamley at al. used vertical acceleration from IMU and means of continuous wavelet transform to detect foot contacts (initial and final). They reported an average error of 0.02 s and 0.03 s representing 2% and 3% of mean stride duration [28]. Storm et al. have compared two algorithms to determine temporal gait parameters based on two shank-worn IMUs or a single waist-worn IMU in free-living condition. IC and FC were detected inside predefined search windows. Then, the IC is identified as the instant of minimum angular velocity in the sagittal plane between the beginning of the IC search window and the instant of maximum anterior-posterior acceleration. The FC is identified as the instant of minimum anterior-posterior acceleration in the FC search window. For the second method using single waist-worn IMU, a first Gaussian continuous wavelet transformation is applied to the vertical acceleration signal, and the minima are identified as the IC timings. Results showed that the stride and step time absolute errors recorded using these methods were higher during outdoor free walking but generated only a small increase in percentage error (6 to 9 ms for stride time, and 9 to 14 ms for step time). Step length calculation was not assessed [29]. These previous studies concerned only healthy participants.

In a study including people with gait disorders, two magneto-inertial units including a tri-axial accelerometer, a tri-axial gyroscope and a tri-axial magnetometer (MIMUs) were fixed to the malleoli for the determination of both temporal and spatial parameters [26]. The gait events were detected using a specific period within which no gait events were expected and additional conditions also had to be satisfied. This complex algorithm was adapted to pathologic gait patterns to limit the risk of extra and missed gait events detection. Neither missed nor extra gait events were observed. Percentage of absolute error in estimating stride length was excellent both for hemiparetic people (3%), Parkinson's disease people (2%) and choreic people (2%), both at comfortable speed and higher speed.

In children with CP-GMFCS I-II (i.e., less affected), some previous studies have demonstrated acceptable validity of accelerometry to detect mobility-related metrics, such as the total number of steps per day, walking distance [33–35], and cadence [36]. Sala et al. evaluated the accuracy of the wrist-based Fitbit Flex and the hip-based Fitbit One in quantitatively measuring the ambulation of children with CP, classified in GMFCS levels I to III, in a clinical setting. Participants were children with CP using different walking aids: any assistive device (n = 28), a posterior rollator (n = 7), one forearm crutch (n = 3), and two forearm crutches (n = 3). They demonstrated that wrist-based was not accurate for counting steps (range of errors between -484 to 35 steps). They reported better results for hip-based device (range of errors between -52 to 6). They concluded that for people having reduced mobility (walked slowly, took small steps, and used a rollator), the step counts for a hip-based and a wrist-based Fitbit must be considered with prudence [33]. These devices did not provide step length and gait speed. Other study assessed the accuracy in distance walked and step count of two commercial devices: the Minimod combining three accelerometers and the AMP (inertial sensors). Participants were diplegic CP and typically developing children. When the walked distance increased, both devices became less accurate and showed greater underestimation of actual distance walked and step count, steps differences were as high as 40 [34]. Both studies showed that commercial devices using a standard algorithm for detecting temporal gait parameters

were not suitable for patients with gait disorders. Recently, Paraschiv-Ionescu et al. have developed a custom-made algorithm based on detection of peaks associated to heel-strike events, and included several processing stages such as peak enhancement and selection of the steps-related peaks using heuristic decision rules. They used the norm of trunk acceleration signals from Physilog4<sup>®</sup> (device including 3D accelerometer, 3D gyroscope, 3D magnetometer and barometer) worn on the trunk. They showed a very good sensitivity, specificity and precision for detection of locomotion period: between 86% to 97%. But, they highlighted that for short period of locomotion and/or if the gait pattern is unsteady with high variability, the error can be important [36]. Moreover, IMU has been successfully used as a tool for diagnosing pathological gait providing estimation about joint kinematics parameters. Glowinski et al. developed a new algorithm combining discrete Fourier transform (DFT) and continuous wavelet transform (CWT). Based on IMU, they identified significant differences in knee flexion during gait in patient with lumbar discopathy [37].

Globally, results indicate that adaptive and custom algorithm is suitable for calculating spatio-temporal gait parameters in people with gait disorders. But these techniques, despite their great robustness, have potential drawbacks for coupling them with AR/VR systems in clinical context. The need for multiple devices to maximize accuracy, the difficulty in synchronizing with AR/VR systems and the complexity of the user's equipment mean that the system is not "plug and play". A simpler configuration, with a single device, could allow for wider clinical application.

To our knowledge, there are very few studies using an AR HMD (Hololens) for calculating spatio-temporal gait parameters. As a preliminary study, Guinet et al. showed that the accuracy of the Hololens was sufficiently high to evaluate the position of the user's head, without spatial drift, in comparison to MOCAP system. They found an absolute errors between 55 to 250 mm in all 3 planes [38]. Using the same device, Geerse et al. tested a method using head vertical maximal position to estimate foot step location [32]. This algorithm has shown a good test-retest reliability and a good concurrent validity at different walking speeds for healthy participants and for people with Parkinson's disease (PD). Still, they observed significant differences between their method and the reference for walking speed, step length and cadence. They also had measurement biases increasing with faster instructed walking speeds. Finally, Ju et al. have proposed a machine learning approach for detecting whether a healthy subject was touching the ground with the left or right foot while walking [31]. This method required the sound recording of footsteps in real-time, that is impossible in clinical use because of the ambient noise. Moreover, this algorithm was not validated for people with gait disorders.

# 3. Materials and Methods

## 3.1. Participants

Thirteen healthy adults and sixty-two children with CP were included. Healthy adults had to be 18 years or older and normal or corrected vision. Subjects were recruited at the Poidatz Rehabilitation Center adjacent to the gait lab. The inclusion criteria for people with CP were an age between 10–18 years, a Gross Motor Function Classification System (GMFCS) [39] I to III. Children for whom a gait analysis test was planned were invited to participate in this study when they met the inclusion criteria. Written consent was previously obtained from each child's parent or guardian and assent from each child to collect and use their clinical data. Children were divided in two groups: Group 1: Children with CP walking without walking aids (N = 32) (i.e., GMFCS I), Group 2: Children with CP walking with crutches or posterior walker (N = 30) (i.e., GMFCS II/III). Characteristics of children included were summarized in Table 1. The study was conducted according to the guidelines of the Declaration of Helsinki, and approved by the National Ethics Committee (see Institutional Review Board and Informed consent statement below). Test session was held in July 2020.

Characteristics	Healthy Participant (n = 13)	Children Group 1 (n = 32)	Children Group 2 (n = 30)
Age (mean)	35.9	12.6	12.3
Sex $(F/M)$	5/8	16/16	13/17
GMFCS	NA	Ι	II–III
Walking aids	No	No	Crutches (18) Posterior walker (12

Table 1. Characteristics of population included.

# 3.2. Gait Analysis Systems

# 3.2.1. MOCAP System

The gait laboratory used a fifteen-camera VICON system (8 MX 20, 5 T 40, 2 T 160) (PluginGait marker set, VICON, Oxford Metrics, UK). Data were collected from markers placed on the headband (4), pelvic (4) and feet (6). Data were recorded at 100 Hz and filtered using a real-time 2nd order low-pass Butterworth filter (with a cutoff frequency of 6). VICON system was considered as a reference for gait analysis and spatiotemporal gait parameters calculation. Hereafter in this article, VICON system has been called MOCAP.

# 3.2.2. Hololens AR HMD

The HoloStep algorithm has been written in C# language. It was a part of the AR application developed with Unity 2019.2.8f1 (64-bit) using Mixed Reality Toolkit version 2 for Microsoft Hololens HMD. This version of AR application did not contain any hologram in order to be comparable to MOCAP. Data were recorded at 100 Hz and filtered using a real-time 2nd order low-pass Butterworth filter (with a cutoff frequency of 6). This application has been deployed in Microsoft Hololens version 1 [40]. In the following, Microsoft Hololens is called AR HMD.

# 3.3. The HoloStep Computational Method

# 3.3.1. Step Detection

When the application starts, the spatial coordinate systems of the HMD were righthanded, which means that the positive *X*-axis points right, the positive *Y*-axis points up (aligned to gravity) and the positive *Z*-axis points towards you (Figure 1) [40].



**Figure 1.** Spatial coordinate system of the AR HMD is  $(0; \vec{x}; \vec{y}; \vec{z})$ .

The data acquisition rate was 100 Hz. At each frame, HoloStep calculates: Time (s), Position  $(x_H, y_H, z_H)$  (m), Filtered position  $(x_F, y_F, z_F)$  (m), Step detected (*Boolean*), Step length (m) and Walking distance from the beginning of the trial (m). These data are stored in memory for direct use in the AR application, and logged in a *.csv* file for later analysis.

The initial position  $P_{t_0}$  of the AR HMD was:

$$P_{t_0} = (0, AR headset y level, 0)_{t_0}$$

Then at any time *t* the AR HMD position  $P_t$  was given in this reference frame:

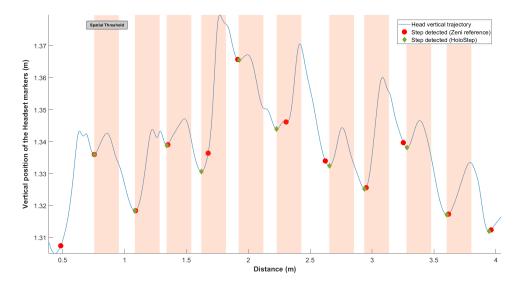
$$P_t = (x_H, y_H, z_H)_t$$

The position signal *P* was then filtered using a second-order zero-lag Butterworth low pass filter with a 6 Hz cut-off frequency to get  $P_F$ . Minimum peaks were detected using  $y_F$  the filtered AR HMD vertical position signal.

Each gait cycle was divided into two phases: stance and swing. Stance consisted of the entire time that a foot was on the ground, starting with an initial contact (IC) when the foot touched the ground, and ending with a toe off (TO) when the foot left the ground. Swing corresponded of the entire time that a foot was in the air, starting with TO and ending at the next IC. The principle of detection used by HoloStep was based on the fact that during walking, the body displacement is pseudo-periodic. The body slightly leaned to the left/right side. It created a characteristic variation on *x*-axis. Moreover, at each IC (i.e., when the foot was touching the ground), the body was at a lower position on *y*-axis. Therefore, when the user was leaning to the left (*x* min) and the body was at a lower position (*y* min), the left foot of the user started to touch the ground. The similar situation happened on the other side (*x* max and *y* min).

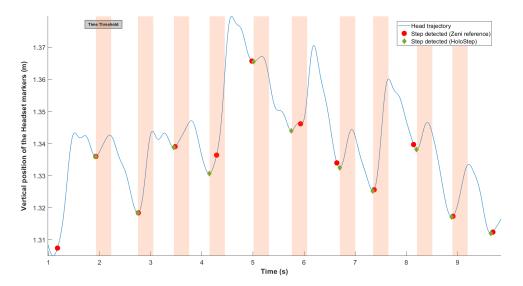
HoloStep was developed using a combination of locking distance [41], locking time [42,43] and peak amplitude detection with custom thresholds for children with CP. In each window, the minimum peak position on  $y_F$  signals was used to detect initial contact IC. In order to define if the distance between two peaks should be considered as a real step, three conditions were checked:

• First, the *locking distance* was defined: new IC was considered only if the distance between two IC was greater than this threshold (Figure 2). In order to make Holostep the most suitable for children with CP, we reviewed a separate data set from the gait analysis of 188 children with CP in a specialised laboratory. The mean distance between two IC was 44.31 cm, with SD = 11.87. We have set the locking distance at 20 cm (rounded down of *mean*  $- 2 \times SD$ ).



**Figure 2.** Representation of the locking distance threshold. In blue, vertical head position  $y_F$  of the user wearing the AR HMD over the walkway. The green diamonds were IC detected with HoloStep. The red circle were IC detected with Zeni algorithm reported to  $y_F$ . The orange bands were the locking distance threshold (minimum peaks ignored by HoloStep).

• Second, the *locking time* was defined : new IC was considered only if the time between two IC detected was greater than this threshold (Figure 3). As before, after analysis of



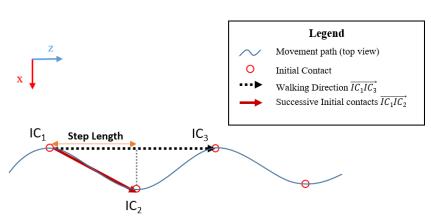
the specific data set for children with CP, mean time between two IC was 56.01 ms, with SD = 12.54. The locking time was set to 30 ms (rounded down of *mean*  $- 2 \times SD$ ).

**Figure 3.** Representation of the locking time threshold. In blue, vertical head position  $y_F$  of the user wearing the AR HMD over the walkway. The green diamonds were the IC detected with HoloStep. The red circle were IC detected with Zeni algorithm reported to  $y_F$ . The orange bands were the locking time threshold (minimum peaks ignored by HoloStep).

• Third, the *peak amplitude* threshold was defined: the minimum difference required between previous maximal  $(P_y)_f$  and current minimal  $(P_y)_f$ . As before, after analysis of the specific data set for children with CP, mean peak amplitude detection was small at 0.3 cm. This is the value we have retained for the peak amplitude threshold.

3.3.2. Step Length and Walking Distance

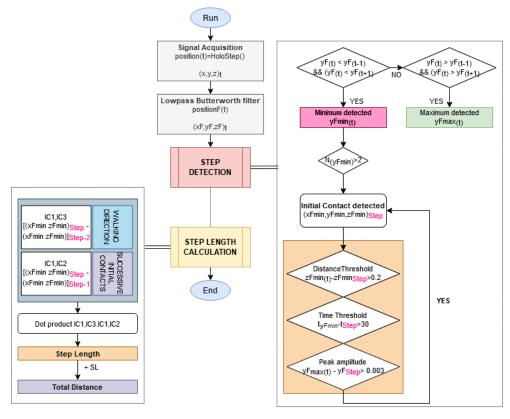
After detecting 2 consecutive steps, step length (SL) was the dot product between walking direction  $\overrightarrow{IC_1IC_3}$  and two successive initial contacts  $\overrightarrow{IC_1IC_2}$  (Figure 4):



$$\overrightarrow{IC_1IC_2} \cdot \overrightarrow{IC_1IC_3} = IC_1IC_2 \times IC_1IC_3 \times \cos(I\widehat{C_2IC_1IC_3})$$

**Figure 4.** Representation of Step Length in the coordinate system of the AR headset  $(0; \vec{x}; \vec{y}; \vec{z})$ .

Finally, the total walking distance was the sum of the successive step lengths. Figure 5 summarises the flow chart of the HoloStep algorithm.



**Figure 5.** Flow chart of the proposed method HoloStep for step detection and step length calculation of children with CP.

## 3.4. Experimental Procedure

Participants were instructed to walk at a comfortable speed in a straight line along an 8-m path in the gait lab. Data were collected for three successful trials.

## 3.5. Data Processing

The user's head position was measured with the AR HMD and with MOCAP using 4 reflective markers placed on the AR HMD. Pelvic and feet position were measured with MOCAP using reflective markers. Dataset was processed using two different algorithms:

- Reference: Zeni algorithm using a set of pelvic and feet markers calculating spatiotemporal gait parameters with high accuracy [22];
- Challenger: HoloStep algorithm using head pose.

From HoloStep algorithm (C#) deployed in the AR HMD, user's position  $(x_H, y_H, z_H)$ , user's position filtered  $(x_F, y_F, z_F)$ , walking speed, step length, number and timing of step detected were extracted in .csv format. From MOCAP, user's position filtered was extracted from the reflective markers placed on the AR HMD  $(x_G, y_G, z_G)$ , walking speed, step length and number of step were calculated using Zeni algorithm. The AR HMD signals were synchronized to the MOCAP signals by an automatic time shifting procedure using a local minimum detection. An Iterative Closest Point (ICP) algorithm was used to align the 3D positions given by the two systems minimizing the distance between them, using geometric transformations (rotations *R* and translations *T*) [44,45]. The ICP algorithm had two steps: The first step consisted of determining the correspondence pairs ( $\vec{p}, \vec{m}$ ) from two data sets *H* and *G*. The aim was to find for each point *p* in *H* its closest point in *G*. The second step was to apply a transformation (*R* and *T*) in order to minimize the distance between the correspondence pairs:

$$E(R,T) = \frac{1}{N_H} \sum_{i=1}^{N_H} ||G_i - RH_i - T||^2$$

with  $H = (x_H, y_H, z_H)$  and  $G = (x_G, y_G, z_G)$  the corresponding points, from AR HMD and from MOCAP respectively. These two steps were repeated until the error was below a given threshold or until the maximum number of iterations was reached. Data processing was performed with MATLAB 2019a.

## 3.6. Statistical Analysis

Bland and Altman analysis was performed to compare walking speed, step length and cadence between the two methods given the bias  $\bar{d}$  (the mean of the differences between the two methods) and the limits of agreement (LoA). The Intraclass Correlation Coefficient for absolute agreement among measurements, also known as criterion-referenced reliability, was calculated (ICC(A, 1)) [46]. The Pearson correlation coefficient r and determination coefficient  $r^2$  were calculated to compare the two methods.

The two-sample t-test was done to compare means of the two methods for walking speed, step length and cadence. The 5% significance level was used to reject the null hypothesis. The confusion matrix allowed visualization of the performance of HoloStep algorithm: sensitivity (true positive rate), specificity (true negative rate), accuracy (true negative and positive rate) and precision (positive predictive value) of step detection for HoloStep algorithm, for each group, were calculated. Each row of the matrix represented the instances in the challenger class (HoloStep), while each column represented the instances in the reference class (Zeni).

# 4. Results

## 4.1. Healthy Participants

For the 13 healthy participants, the total number of IC detected was 65 with Zeni and 63 with HoloStep. The HoloStep algorithm ignored 2 IC (false negative) for 2 participants. Sensitivity, specificity, accuracy and precision of HoloStep algorithm were excellent (Table 2). The Bland and Altman analysis shown a bias  $\bar{d} = 0.054$  m for step length and 0.035 m/s for walking speed. The mean difference between the two algorithms for all variable were not significant. The ICC coefficients were excellent for walking speed (ICC = 0.973) and good for step length and cadence (ICC = 0.778 and 0.534, respectively). The mean difference for step length between Zeni and HoloStep was 5.6 cm (Table 3).

**Table 2.** Sensitivity, specificity, accuracy and precision of step detection with HoloStep algorithm for healthy participants and children with CP.

	Sensitivity	Specificity	Accuracy	Precision
Healthy participant (n = 13)	0.969	1.000	0.999	1.000
Children Group 1 (n = 32) Children Group 2 (n = 30)	0.969 0.989	0.999 1.000	0.999 1.000	0.964 0.984
All CP children (n = 62)	0.979	1.000	0.999	0.974

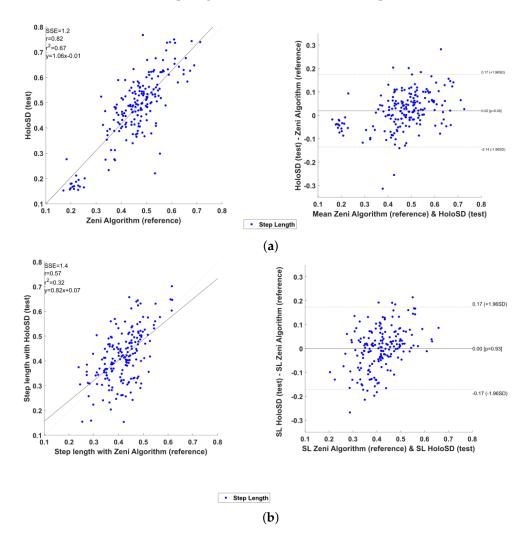
**Table 3.** Concurrent validity for spatiotemporal gait parameters in healthy participants (HP) and in children with cerebral palsy (Group 1 and Group 2).

		MOCAP Zeni	Hololens HMD HoloStep				
		Mean $+/-$ SD	Mean $+/-$ SD	Bias (95% LoA)	t-statistics	ICC(A,1)	r corr
Walking Speed (m/s)	1	1.044 +/- 0.254 0.667 +/- 0.180 1.277 +/- 0.199	0.648 + / - 0.174	0.018 (-0.012 0.048) 0.018 (-0.017 0.053) 0.035 (-0.026 0.096)	$\begin{array}{l} t(30) = -0.28,  p = 0.78 \\ t(28) = -0.40,  p = 0.69 \\ t(11) = -0.45,  p = 0.66 \end{array}$	0.996 0.990 0.973	0.998 0.995 0.988
Step Length (m)	1	$\begin{array}{c} 0.488 + / - \ 0.090 \\ 0.430 + / - \ 0.064 \\ 0.623 + / - \ 0.079 \end{array}$	0.434 + / - 0.079	0.017 (-0.106 0.140) 0.005 (-0.152 0.162) 0.054 (-0.048 0.156)	$\begin{array}{l} t(30) = 1.07,  p = 0.29 \\ t(28) = 0.18,  p = 0.86 \\ t(11) = 1.74,  p = 0.095 \end{array}$	0.922 0.863 0.778	0.885 0.649 0.802
Cadence (steps/s)	1	$\begin{array}{c} 1.980 + / - \ 0.272 \\ 1.178 + / - \ 0.308 \\ 1.908 + / - \ 0.196 \end{array}$	1.425 + / - 0.294	0.142 (-0.249 0.534) -0.247 (-0.589 0.095) 0.0099 (-0.288 0.486)	$\begin{array}{l} t(30) = -2.11, p = 0.29 \\ t(28) = 3.18, p = 0.86 \\ t(11) = -1.18, p = 0.095 \end{array}$	0.642 0.625 0.534	0.726 0.833 0.582

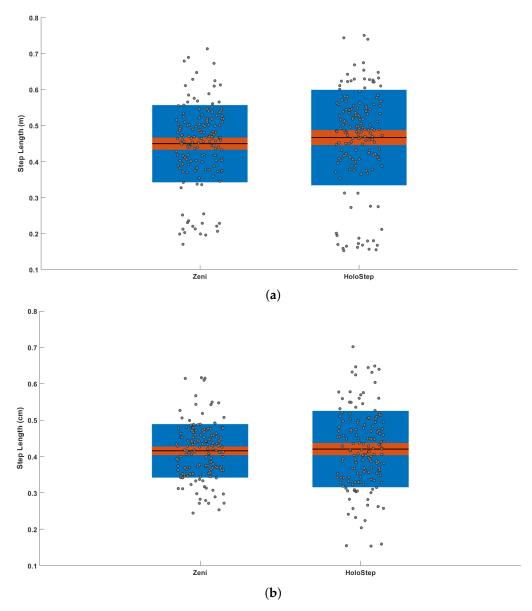
#### 4.2. Children with CP

For the 32 participants from group 1 (children with CP walking without aids/GMFCSI), the total number of IC detected was 194 with Zeni and 195 with HoloStep. The HoloStep algorithm ignored 6 IC (false negative) and added 7 false IC (false positive) for 7 different patients. Sensitivity, specificity, accuracy and precision of HoloStep algorithm were excellent (Table 2). The Bland and Altman analysis shown a bias  $\bar{d} = 0.017$  m for step length and 0.018 m/s for walking speed (Figure 6a). The mean difference between the two algorithms for all variable were not significant (Figure 7a). The ICC coefficients were excellent for step length and walking speed (ICC = 0.922 and 0.996 respectively) and good for cadence (ICC = 0.642). The mean difference for step length between Zeni and HoloStep was 2.6 cm (Table 3).

For the 30 participants from group 2 (children with CP walking with aids/GMFCSII-III), the total number of IC detected was 184 with Zeni and 185 with HoloStep. The HoloStep algorithm ignored 2 IC (false negative) and added 3 false IC (false positive) for 3 different patients. Sensitivity, specificity, accuracy and precision of HoloStep algorithm were excellent (Table 2). The Bland and Altman analysis shown a bias d = 0.005 m for step length and 0.018 m/s for walking speed (Figure 6b). The mean difference between the two algorithms for all variable were not significant (Figure 7b). The ICC coefficients were excellent for step length and walking speed (ICC = 0.863 and 0.990 respectively) and good for cadence (ICC = 0.625). The mean difference for step length between Zeni and HoloStep was 4 cm (Table 3).



**Figure 6.** Statistical analysis graphics for children with CP. Left: Linear Distribution of step length between the two algorithms. Right: Bland and Altman plot representing step length. A bolt horizontal line representing the bias. Additional dotted horizontal lines, limits of agreement, are added to the plot at  $d \pm 1.96$  SD. (a) Children with CP—Group 1 (b) Children with CP—Group 2.



**Figure 7.** Difference between HoloStep and Zeni algorithm. Box plot visualizing raw data along the mean, 95% confidence interval, and 1 SD. (**a**) Children with CP—Group 1 (**b**) Children with CP—Group 2.

# 5. Discussion

This study is the first to describe and assess the validity of a custom algorithm deployed in the Hololens AR HMD to calculate spatiotemporal gait parameters both in healthy participants and children with CP. The results of our study suggest that HoloStep algorithm using Hololens AR HMD calculate spatiotemporal gait parameters with sufficient accuracy even in people with gait disorders using walking aids.

# 5.1. Comparison with Other Methods

HoloStep performance for speed calculation was comparable to those obtained with an IMU. For ex., Zijlstra et al. obtained a mean difference between predicted and real speeds below 0.05 m/s [25]. HoloStep had a mean difference from 0.018 m/s to 0.035 m/s between groups. For foot contacts detection, the sensitivity across various methods using IMU varied between 81% and 100% [27], whereas HoloStep was between 97% to 99%. For step length, the mean absolute error in estimating stride length for adult with gait disorders varied from 1.8 cm for hemiparetic to 2.6 cm for choreic people. HoloStep error varied from 2.6 cm to 4 cm for children with CP. This gap could be explained by the use of a posterior rollator which led to the detection of false steps. HoloStep using AR HMD obtained better results than other commercial devices like the wrist-based FitBit Flex and the hip-based FitBit One in quantitatively measuring the ambulation of children with CP [33]. In this study, mean absolute error in number of steps detected for children varied from 6 to 17 depending on the used walking aids. For HoloStep, those parameters varied from 2 to 7. Paraschiv-Ionescu et al. have developed a robust algorithm that used data from IMU device worn on lower back (L5 vertebrae) to calculate gait parameters. Performance metrics (sensitivity, specificity and precision) were excellent, from 0.90 to 0.98 for children with CP. But, this algorithm could not be used in the AR HMD because it used the norm of trunk acceleration signals.

Recently, Geerse et al. proposed a method to calculate spatiotemporal gait parameters using a Hololens AR HMD and compared this method to the reference Interactive Walkway System (IWS). They found that ICC were excellent for between-systems agreement for walking speed, step length and cadence for healthy adults and people with Parkinson's Disease (PD) (ICC > 0.92). But limits of agreement obtained with Bland-Altman analysis were quite narrow. Still, walking speed and step length were underestimated with biases increasing with faster walking speeds (min bias of 0.6 for people with PD to 3.1 for healthy people walking faster) [32]. They also found statistical difference between step length measured with Hololens and IWS (p < 0.05). These between-systems biases were justified by the authors because of the drift in tracking and deviations in the map (caused by the Hololens AR HMD spatial mapping component). Using data available in supplementary material, we have calculated step length with HoloStep. Using population-best matched thresholds, we have found no statistical difference between step length calculated with HoloSD and IWS, and HoloStep seemed to have better results on the biases (min bias of 0.07 for people with PD to 1.9 for healthy walking faster). These results suggest that the use of custom thresholds enhances the calculation of spatiotemporal gait parameters, but a stronger method to compare these two algorithms is necessary to conclude.

Sun et al. have developed two AR-based automated functional mobility test using Hololens AR HMD: Sit To Stand (STS) and Time Up and Go (TUG). In comparison with reference inertial sensor (Opal, APDM), vertical kinematic data (displacement, velocity and acceleration) shown a bias less than 0.02 s for STS and 0.13 s for TUG, with range of error within  $\pm 0.8$  s. Correlation coefficient for kinematic measurement agreement between Hololens and reference sensors were from 0.74 to 0.99 [47]. We obtained similar results with HoloStep for kinematic measurement.

## 5.2. Gait Detection for Children with CP

Children with CP present alteration of dynamic stability during gait because of deficits in balance and postural control. Hsue et al. demonstrated that children with CP showed significantly larger vertical and medio-lateral displacements of the Center of Mass than TD group. But, the trajectories have the same shape (sinusoidal pattern) both for TD group than children with CP. In vertical directions, the Center of Mass reached a maximum peak at mid-stance, and a minimum at the end of terminal stance (when the two feet are in contact with the ground). It was interesting to observe that the minimum peaks were shifted for children with CP (4% of gait cycle after) [48]. When children have an asymmetrical gait (this is often the case for children using crutches), this pattern was not that regular: amplitude of the first peak (more affected side) is higher than the second [48]. Moreover, children with CP walking with crutches presented some small and inconsistent peaks on vertical axis. This variation of walking pattern conducted to a lot of false IC detection by existing algorithms. HoloStep used custom thresholds, based on the analysis of CP gait pattern, in order to minimize these bias. Eyes, head and chest orientation are strongly correlated when people walk with a fixed gaze direction [49]. The head and trunk trajectories are also linked during locomotion (signals have the same sinusoidal shape) [50]. These considerations encouraged to develop wearable technology fixed to the participant's head [51] which is the case of a HMD.

# 5.3. HoloStep Limitations

HoloStep is based on the kinematics of the head, which requires special attention as it has 6 degrees of freedom and can have variable patterns. In sagittal plane, the variability of head displacement for children with CP is high during gait [52]. But, at initial contact, Heyrman et al. reported that ICC within and between session were above 0.7 (comparable to the thorax kinematics) [52]. In our study, children were asked to look straight ahead during walking tests to minimize head movement, which could be considered as non spontaneous gait. However, this constraint is relevant to the future use of the Hololens AR HMD, which will display holograms in front of the subject, encouraging them to keep their gaze on the horizon. Each child produced distinctive walking pattern, peak amplitudes could be different (regular, asymmetric, short, round...). Thus, a method based on individual signals could be better than predefined thresholds [41]. The IC detection could be more specific to the individual's gait pattern. In the same way, the locking period threshold could be customized for one gait pattern. The use of machine learning method could contribute to improve step detection, and by extension step length [31]. Another limitation is that HoloStep has been tested only with the Hololens AR HM version 1. Using another device would probably require some adjustments to the thresholds used. Furthermore, it can be assumed that future versions of the various manufacturers' HMD will further improve the robustness and accuracy of head pose capture.

# 6. Conclusions and Future Work

We have developed and evaluated a new algorithm called HoloStep to calculate spatiotemporal gait parameters using only the head pose provided by an augmented reality headset (Hololens). It is based on the detection of peaks associated to initial contact event, and used a combination of locking distance, locking time, peak amplitude detection with custom thresholds for children with CP. An experimental comparison with a reference algorithm based on full motion kinematics shown that HoloStep accurately detected foot contact, and calculated step length, total distance walked and gait speed both for children with CP and healthy participants.

Once the gait parameters have been obtained, another step before designing relevant rehabilitation exercises is to investigate feedback on gait performance in AR. We therefore started to model and evaluate different visual feedback on speed, and their effects on patients' walking performance. We have also started the development of a serious game using a process framework that involves a multidisciplinary team and is inspired by existing methodologies [53,54]. This serious game includes sprint training sessions and adaptive feedback. We will start a larger clinical study to assess acceptance, usability [55] and therapeutic effects on children with CP.

Beyond this specific application, we believe that the HoloStep algorithm can be implemented in other serious games aimed at a wider range of different patients for motor rehabilitation purposes.

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# Abbreviations

The following abbreviations are used in this manuscript:

AR	Augmented Reality
СР	Cerebral Palsy
FC	Foot Contact
GMFCS	Gross Motor Function Classification System
HMD	Head Mounted Display
HP	Healthy Participants
IC	Initial Contact
ICC	Intra-Class Coefficient
SD	Standard Deviation
SL	Step Length
TD	Typically Developing
TO	Toe Off
VR	Virtual Reality

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# 3.6 Conclusion

The Microsoft Hololens AR Headset was selected because its technical characteristics matched the project team's crucial technical aspects and specifications. In our first experiment, we have shown that the accuracy of the Hololens is sufficiently high to measure the position and the velocity of the headset, and the user, by extension, without spatiotemporal drift. Second, we have developed and validated an algorithm, HoloStep, calculating in real-time gait spatiotemporal parameters with the only position of the Hololens. It is based on detecting peaks associated with initial contact events and uses a combination of locking distance, locking time, peak amplitude detection with custom thresholds for children with CP. These two essential results encourage us to go further with the Hololens and use this device for our gait rehabilitation active video game.

# Chapter 4

# Multimodal Feedback in Augmented Reality for Motor Rehabilitation

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Chapter 4 focuses on feedback for motor rehabilitation in AR. We expose our theoretical model of feedback in AR. Then we detail the results of two clinical studies testing the impact of scenarios combining different visual feedback characteristics on walking speed in adult population (Article 4) and children with CP (Articles 5 & 6). We have demonstrated that specific feedback characteristics helped reach and maintain target speed. Finally, we discuss our observations of different profiles of people based on their responses to feedback.

# 4.1 Introduction

As soon as the multidisciplinary team and children validated the choice of the tool, the next steps of the ARRoW-CP project focusing on motor learning, motor rehabilitation, and feedback have been initiated. As we have seen in Section 2.6.4, new technologies, such as augmented reality (AR), appear to be an excellent way to combine repetition, practice specificity, motivation, feedback, and reward, both in a clinical context and at home. While the potential of AR feedback, particularly for enhancing motivation, has been demonstrated, few studies have delved into how different forms or presentations of AR feedback may impact gait parameters and performance. Then, after defining the term feedback, we propose a model of feedback in Augmented Reality (AR) for Motor Rehabilitation (Section 4.2). Indeed, AR feedback can be displayed to the user through different characteristics. This theoretical work was necessary before designing feedback and testing specific characteristics of the model. The final aim was to optimize the augmented information provided to the patient. To carry out this work, we extended and adapted the biofeedback model described by Macintosh et al. [85], and the qualitative model from Martinez et al. [122], to the AR context. Our descriptive model of feedback in AR application helped us design two applications, called Best-Of ARRoW v1 and v2. In the next section, we present the two clinical studies involving healthy adults (Section 4.3) and children with CP (Section 4.4). We have tested with ARRoW v1 and v2 the impact of different virtual feedback characteristics on walking speed. Finally, the last section of this chapter presents an exploratory analysis on the profile of responders to the visual feedback (Section 4.5). We have observed that not all children with CP performed equally with AR feedback. Some children never responded or were disturbed by feedback regardless of the feedback characteristics, while other children responded to some but not all the tested feedback.

# 4.2 Model of Feedback in Augmented Reality

As previously presented, augmented feedback is crucial for enhancing motor (re)learning and motor rehabilitation. Before detailing the feedback model that we have developed, we have to define feedback in a general context.

In Engineering Sciences, to "feed back" is to return to an earlier position in a mechanical process [123]. Feedback was defined as the information about the gap between the actual level and the reference level of a system parameter that is used to alter the gap in some way. The information itself was not feedback unless translated into action [124]. Common application was the proportional-integral-derivative controller (PID controller), a control loop mechanism employing feedback to calculate an error value continuously to reduce system error. Human intervention was rarely present (the goal was sometimes to avoid it). The system was autonomous and automated according to pre-established human settings and configuration parameters. Feedback was included in the system or was the closed-loop system. In industry, it was used to minimize error for sampling analysis (autosampler) to increase gains in productivity by accelerating cadence production. It was not the same approach when the feedback was used for motor learning. In that case, feedback was designed for human beings, for human development. The human being was the center of the system, the beneficiary of the system. Humans should be able to perceive, understand the feedback, and adapt their behavior. It was more complicated than in automation because one feedback did not conduce to the same output for each person from a specific input. We have to keep in mind the superior thinking ability of humans compared to machines... In motor learning theory, feedback

was no longer the total system but an element of the system that helped the user/learner. The system was more defined as how two components (the learner and the rehabilitation device or the learner and the therapist) interacted. However, we should not neglect the definitions we have inherited from Engineering science. We should use these historical feedback definitions and adapt them to the rehabilitation context.

We conducted a literature review to propose a standard definition of feedback applied in (and applicable to) rehabilitation. We included publications related to the effectiveness of interventions in individuals with motor disabilities, including feedback. The PubMed, Google Scholar, IEEE Xplore, and Cochrane databases were searched in March 2020, using a combination of keywords related to motor rehabilitation and feedback. We included all studies with a definition of "feedback" in the Introduction, Method, or Discussion section. Note that most of these studies were clinical trials assessing a specific device, tool, platform, video game, and system displayed feedback. For all included articles, we extracted the following information into an Excel spreadsheet: author/date, the word used to qualify feedback (feedback, augmented feedback, biofeedback), nature of feedback (information, action, property, process), what were the input/output of the system, the action verb associated, what were the characteristics and the aim of the feedback. We included 22 definitions from 22 articles. All these definitions are available in Appendix 5.6.

Our qualitative analysis of occurrence keywords provided the following definitions for feedback in the Motor Rehabilitation context (See Annex 5.6 for all references) :

The feedback retraining paradigm is based on the conversion, supplementation, and augmentation of sensory information that are usually accessible only by an internal focus of attention to accessible information. In this paradigm, *augmented feedback is defined as augmented sensory information provided by an external resource (therapist or display) to the patient.* Sensory channels used to deliver information are visual, auditory, or haptic, linked to the proprioception properties of humans.

The timing of feedback delivery is critical. Concurrent feedback is delivered while the skill is being performed, terminal feedback is provided after the skill is performed, with or without delay.

Augmented feedback is sensory information added to interventions to improve motor learning and motor recovery in the rehabilitation of persons with neurological disorders. The information provided to the user could refer to the movement's pattern (knowledge of performance) or the result on the environment or the outcome of action concerning the goal (knowledge of results). In the rehabilitation context, the feedback content could vary according to the aim of the therapy, for example, movement kinematics or kinetics motor performance gait pattern.

Augmented feedback is efficient to modify specific motor behaviors to make the learning process more explicit, to enhance the practice environment, to facilitate learning of complex tasks, to speed up the learning process. The effectiveness of this augmented feedback depends on therapist competency, display parameters, augmented feedback frequency, and timing.

Note that specific augmented feedback, named biofeedback, defines a method of treatment that uses an electronic or electromechanical instrument to accurately measure, process, and deliver physiological processes to an individual. It is a non-invasive technique that requires sensing, processing, and output (e.g., visual, haptic, or auditory) technologies. Biofeedback retraining paradigms are based on the patient's active interpretation and response to feedback cues of individual acquired signals.

To summarize :

# Feedback for Motor Rehabilitation (short version)

Feedback is based on the conversion, the supplementation and augmentation of sensory informations that are usually accessible only by an internal focus of attention, to accessible information. In this paradigm, augmented feedback is defined as augmented sensory information (visual, auditory, haptic, proprioceptive) provided by an external resource (therapist or device) to the patient.

Thus, as pointed out in the definition, augmented feedback characteristics are multiple. The next step was developing a feedback model suitable for both motor rehabilitation and the augmented reality context. Our model is a practical extension of the theory of feedback to formalize their characteristics better. We remind that a model is a simplified, relative, incomplete and temporary representation of a phenomenon. Through its qualities of description, representation, and analysis, a model allowed us to discover new links, to state new hypotheses, to formalize common characteristics or divergences, and to define innovative methods of intervention [125].

Moreover, our model allows a projection of the feedback theory to the AR environment. Thus, it would be possible to determine the impact of each feedback characteristic on motor rehabilitation by testing different combinations of our model. In a more practical sense, when creating a future AR application including feedback, a researcher, developer, or designer could refer to our model to configure their feedback. Our model of AR feedback for motor rehabilitation has two complementary sides : **descriptive** and **qualitative**. Since an exhaustive evaluation of this model was not possible in the time available for this research, we have tested some of them in two studies (4.3 and 4.4). In addition, the feedback characteristics deployed in our final AVG are described in 5.3.5.

The first side of our model of AR feedback for motor rehabilitation, which we have called **descriptive** (Figure 4.1), is inspired by the model of biofeedback developed by Macintosh et al. [85].

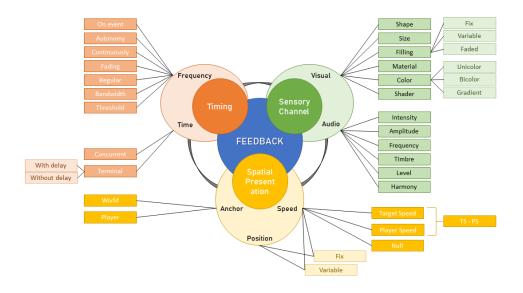


Figure 4.1 – **Descriptive Model of Feedback for VR/AR application.** Feedback is defined according three important notions - Spatial presentation, Sensory channel and Timing. Each notion includes several characteristics. For example, visual and audio for sensory channel. Then, each feature is subdivided in more specific features, such as color of the feedback. Our model could be used to describe more fully the feedback.

In their model, Macintosh et al. described biofeedback according to the following characteristics [85]:

- method of presentation audio, visual, haptic, reward, immersive
- movement variable accuracy, applied force, centre of pressure, electromyography, joint angle, movement coordination/execution
- focus of attention knowledge of results, knowledge of performance
- timing concurrent, terminal, summary
- frequency consistent, bandwidth, blocked, faded
- autonomy passive, active

The Macintosh's model is very detailed, but it is not completely adapted to AR environments. There is no notion relative to the real environment, the position and moving speed of the feedback, the reference frame. Through brainstorming sessions involving AR specialists, scientists, engineers, and therapists, but also through scientific and more general readings on the effect of AR environments on perception, and finally with the test of game/application in virtual/augmented/mixed reality, we highlighted new characteristics of feedback applicable to AR. Thus, it quickly appeared that the notion of spatial presentation of the "virtual object" (called hereafter hologram) serving as feedback was fundamental in AR. **Spatial presentation** (yellow section of the model - see Figure 4.1) has 3 characteristics :

## **Spatial Presentation**

# Spatial anchor, Position et Moving speed

The **spatial anchor** characteristic can take two values:

- 1. Feedback can be *body-locked*, that means attached to the gaze (i.e. when the user look at left, the feedback follows his gaze and go on the left of the screen)
- 2. Feedback can be *world-locked*, that means attached to the environment (i.e. the feedback is like a flower pot with predefined position in the room)

The characteristic of the **position** and **moving speed** of the feedback, relative to the player and the world, are linked. It could be variable or not. To illustrate this, let's take an example. In a scenario where the goal is to catch an hologram moving in front of the player at a predefined speed (called target speed), we could say that:

- Position of the feedback relative to the <u>world</u> is *variable* because its spatial coordinates change over time
- Position of the feedback relative to the <u>player</u> is *variable* and it depends on the position and relative speed of the player with respect to the position and speed of the feedback (in our example, the target speed)
- Speed of the feedback relative to the <u>world</u> is *invariable* et equal to the pre-defined target speed
- Speed of the feedback relative to the <u>player</u> is |*Targetspeed PlayerSpeed*|, the feedback is moving ahead at the target speed and the player has his own speed. The player perceives that the feedback is moving at target speed minus his own speed.

Next other important notion of our descriptive feedback model is **sensory channel** (green section of the model - see Figure 4.1. We share this notion with Macintosh et al, even if they used the term *method of presentation*. We estimated that this term is less precise than sensory channel to qualify **visual** and **auditory** characteristics of the feedback. We have also added sub-characteristics, notably for visual aspect because of the 3D vision in AR. So, we have to define sensory channel :

# Sensory Channel

# Visual, Audio

The Visual characteristics is characterized by the shape of the hologram, but also the size, the color (unicolor, bicolor, gradient), the material, the shader and the filling. All these characteristics influence the perception and recognition of the hologram, and allow to differentiate it from a real object [126]. The hologram could appear brighter, transparent, potentially badly bound to its environment, lacking shading and light reflections, depending on its configuration. In general, the realism of virtual objects is still an important issue in AR.

The Audio (or auditory) characteristics refer (here) to the subject of perception by the brain. There are, historically, six experimentally separable ways in which sound waves are analysed : pitch, duration, loudness, timbre, sonic texture and spatial location [127]. We removed the last one because it was redundant with spatial presentation. We added the "harmony" defined as the process by which the composition of individual sounds, or superpositions of sounds, is analysed by hearing (in other words, harmony is the fact that various sounds perceived together agree or go well together), that could influence the comprehension of the feedback.

Finally, last notion of our descriptive feedback model is **timing** (orange section of the model - see Figure 4.1), that have **time** and **frequency** characteristics.

Timing	
Time, Frequency	

The **time** is relative to the "when" component. Feedback can be displayed when the learner is performing the movement or after it has been executed.

Concerning **frequency**, the feedback can be displayed if a specific event occurs, or continuously, or if the user requests it, by regular interval, by bandwidth, if a threshold has been crossed, with fading.

The second side of our model of AR feedback for Motor Rehabilitation is called **qualitative** (Figure 4.2). It was inspired by an article from Martinez et al. [122], describing their learning model for skiing, based on literature and monitor's experience. We have completed this model by the notion of *descriptive* and *prescriptive* feedback described in [128, 129].

Our qualitative model has a total of six categories defining the quality of feedback:

- 1. **Precision**. Feedback focused on primary factors like the joint position correction and the effects they have on the movement over secondary factors like other joint position that did not influence the movement itself [130]
- 2. Comprehension. Asking the learners if they perceived the feedback and if they understood the information given as feedback (yes or no) [131]
- 3. Direction. Predominance of individual feedback over group feedback [132]
- 4. Affectivity. Predominance of positive feedback over negative feedback [133]
- Specificity. Predominance of Motivational, not linked with the user's behavior or performance, or Informative feedback, that can be descriptive (description of the error), prescriptive (advice to correct the error), corrective (instruction to correct the error and +/- immediate repetition) [129].
- 6. Focus of Attention. Knowledge of results (KR) presents feedback in relation to the outcome of a movement whereas knowledge of performance (KP) addresses qualities of the movement itself [75]

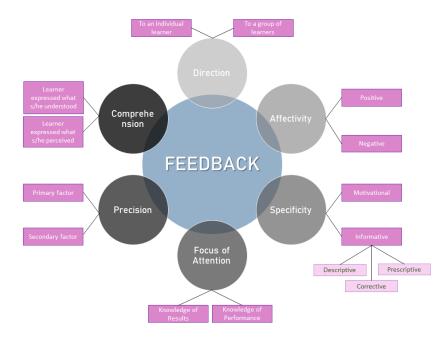


Figure 4.2 – Qualitative Model of Feedback for VR/AR application. Feedback is defined according six important notions - Precision, Comprehension, Direction, Affectivity, Specificity and Focus of Attention. Each notion includes several options that should be precised when describing a feedback. For example, positive and negative for affectivity notion. Our qualitative model could be used, in addition to the descriptive previously described model, to explicit more fully the feedback.

# 4.3 A prospective study on the impact of virtual feedback on walking speed on healthy adults (Article 4)

This study investigates the impact of different forms of visual AR feedback on gait in typically developing adults before future studies, including people with neurological disorders.

The primary research questions are: Can AR feedback be used to help individuals achieve or exceed a target walking speed (WS)? Does AR feedback result in increased variability in WS? We hypothesize that a game with AR feedback can help individuals maintain or exceed a target WS without significantly affecting WS variability.

The mini-games (MG) provided to participants were developed during my internship at the PEARL Lab in Toronto in October 2019. The PEARL Lab led by Professor Elaine Biddiss brings together a team of researchers and developers specializing in making and testing serious rehabilitation games for children. Based on the Macintosh feedback model (at that time, we had not fully developed our feedback model), we collectively thought within the PEARL Lab team to create five relevant mini-games to help individuals achieve or exceed a target walking speed. These mini-games had different visual characteristics and could be described by the Macintosh model: Focus of Attention (KR or KP), Method of Presentation. But as we were aware early on of the importance of spatial presentation in AR, we also described the feedback in terms of this specific feature. We used the Microsoft Hololens headset that we have already presented in chapter 3.

Participants were 15 adults (36.3 + - 9.8y, 11 women/4 men) with typical or corrected-to-typical vision. First, the participants were familiar with the AR environment with a scene

containing holograms. Then, they had to walk as fast as possible, over 15 meters. This average speed defined the target speed for the mini-games (Calibration). Finally, the five mini-games were presented in random order and a control scene without feedback. In each mini-game, participants had to walk 30 meters with the oral instruction of walking at their target speed.

After checking the stochastic assumptions, within-subjects ANOVA for repeated measures and the Dunnett test for multiple comparisons were performed to compare experimental values (i.e., during each mini-game) and a single control value. We have compared the mean WS and variability of WS during each trial. A questionnaire with a 5-point Likert scale from 1 to 5 (strongly disagree/agree) was completed to assess this experiment.

The mean WS and target speed were not statistically different during each mini-game and the control scene. All mini-games helped the participant to exceed the target WS. The Dunnett test revealed that the mean WS during MG4 was significantly higher than the target WS, but MG4 and MG5 resulted in high variability in WS compared to the control scene. Our hypothesis to explain these results was that the small field of view (30°) constrained some participants to turn their heads to follow holograms, disturbing their walking pattern. Another reason could be the aim of the mini-game. In MG4, some participants wanted to race with the butterfly, which may lead to speed variability as to when some reached it, they slowed down. So, changing the spatial representation of MG4 from world-locked to body-locked could be a way to decrease this high variability. Participants reported a high level of satisfaction with the game session. They rated the mini-games to be sufficiently efficient to reach the target speed. About the Hololens, they did not report any limitations for walking, but they highlighted discomfort due to the weight-bearing on the nose.

This investigation demonstrated that gait parameters and user experience could vary depending on the type of AR feedback presented. Some feedback modalities increased WS, whereas others had a more considerable impact on speed variability. Specific recommendations based on learnings in this study include using knowledge of results feedback to create a more challenging task that motivates participants to excel. These body-locked holograms are easier to track and clarify the game presentation. This first experimentation helped us to design better the feedback for the future test, including children with CP.

This paper is attached to this thesis manuscript as submitted in *JMIR Rehabilitation* and Assistive Technologies on October 2021. This work has been presented at the 29<sup>th</sup> European Society for Movement Analysis for Adults and Children Congress in October 2020.

# Development of a serious game for gait intervention in Augmented reality: a prospective study on the impact of virtual feedback on walking speed

JMIR Rehabilitation and Assistive Technologies

SUBMITTED ARTICLE



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# Abstract

*Background*. Improving walking speed can be a time consuming and tiresome process for people with disabilities. Integrating augmented reality (AR) games into gait training programs could increase motivation and engagement in rehabilitation sessions.

*Research question.* Does an AR game with visual feedback help to achieve or exceed target walking speed without increasing variability in walking speed?

*Methods.* This prospective study presents the game framework used to develop five AR mini-game activities for gait training. These mini-games combine different feedback modalities. 15 healthy participants walked on 30 m with the mini-games, as well as a control scene without feedback. Instruction was to walk at target walking speed (WS), recorded previously during calibration walking sprint. The mean and standard deviation of WS during each condition were recorded. ANOVA and multiple comparisons with Dunnett correction were performed. Additionally, participants' opinions on the game and device were collected via questionnaires.

*Results.* Mini-games did not cause any disturbance in the mean WS (ANOVA with Dunnett correction p¿0.05 for all mini-games vs. Control). All mini-games helped participants to largely exceed the target WS. The mini-games MG4 and MG5 resulted in a high variability in WS compared to the control scene. Participants reported a high level of satisfaction with the game session (5/5, IQR 1). Participants judged the game effective in helping them reach the target speed (4/5, IQR 1). Specific recommendations include using knowledge of results feedback, body-locked content, and clarify game objectives.

*Significance*. AR games supported increased walking speed. The choice of feedback modalities was important and influenced gait speed and variability. These results encouraged further development and evaluation of AR for gait training with careful consideration of the feedback modalities presented.

# **Keywords**

Gait Rehabilitation, Augmented Reality, Feedback, Serious Game, Walking Speed

# Introduction

Gait in patient with neurological disorders (ND) is characterized by a slower walking speed (WS), a shorterstride length, a lower cadence, and more time spent in double support (Armand, 2016). Gait quality, especially WS, are often hard to improve for people with ND.2 Improved walking ability has a positive impact on achievement of daily activities and motivating social engagement (Booth, 2018) Current motor learning theories recommend taskspecific, variable, and high intensity practice (Novak, 2013). New technologies, such as augmented reality (AR), appear to be a good way to combine repetition, practice specificity, motivation, feedback, and reward, both in a clinical context and at home (Cabo-de-la-cuerda, 2015). AR that delivers virtual elements superimposed on the person's view of the real-world could be used for gait training without a treadmill. Using a mixed-reality system combining an AR head-mounted display (HMD) and a sensor-based motion capture system, a case study showed that a patient adapted his gait performance (Held, 2020). However, AR promotes patients' engagement in the therapy and potentiates therapeutic gains by increasing motivation (Levin,2010,L). While the potential of AR feedback, particularly for enhancing motivation, has been demonstrated, few studies have delved into how different forms or presentations of AR feedback may impact gait parameters and performance.8 One concern to AR feedback for people with ND during gait training is the extent to which it may distract or increase cognitive load. Previous studies have shown that gait performance decreases for people with ND as dual task cognitive load increased (Carcreff, 2019). Moreover, several studies demonstrated that a high gait variability,

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including variability of WS, results in walking instability and a higher risk of falling (Sidoroff, 2020). In this study, we investigate the acceptability of an AR HMD and the impact of different forms of visual AR feedback on gait in typically developing adults prior to future studies including people with ND. The primary research questions are: Can AR feedback be used to help individuals achieve or exceed a target WS? Does AR feedback result in increased variability in WS? Our hypothesis is that a game with AR feedback can help individuals to maintain or exceed a target WS without significantly affecting WS variability.

# Methods

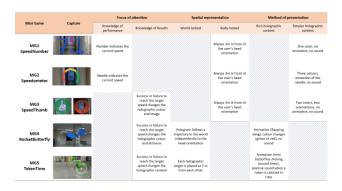
# AR game and feedback development framework

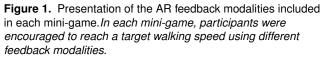
This work followed the development framework PROGame (Amengual, 2018): the team (therapists, researchers, engineers) identified the need for the serious game and user categories (users, experts), they clarified the requirements of the game (operational objectives and restrictions). These identifications were based upon prior experience and literature review (Booth, 2018,D,H). The identified need was to improve motivation and feedback during gait training. Operational objectives included: to be safe and adaptable to the motor capabilities of diverse patient; to provide efficient gait training in line with motor learning theory; to provide high quality feedback on WS; to contain gamified content to motivate the patient to achieve the rehabilitation goals; to be fun. Additionally, the team identified crucial technical requirements: people must be free to walk without restriction of movement (e.g., wireless, low-weight/comfortable, secure) and they must be able to use their usual walking aids (crutches, posterior walker). It should be a "ready-to-use" application both for patient and clinicians to avoid wasting valuable clinical time. The Hololens AR HMD was selected because its technical characteristics matched the identified requirements (Microsoft Company, 2020). It has also been demonstrated to be sufficiently accurate to evaluate the head pose and WS of the user without time drift in the global environment (Guinet, 2016). To investigate our hypothesis, 5 mini games using different feedback were developed (Figure 1). Feedback are characterized by modalities that define their functioning, their role or their form of presentation (MacIntosh, 2019):

- Focus of attention: knowledge of result (KR) focusing on the outcome of a movement or knowledge of performance (KP) addressing qualities of the movement itself,
- Method of presentation: simple visual content or richer effect (animation),
- Frame of reference: hologram can be world-locked (stationary frame of reference) or body-locked (attached frame of reference).

# Subjects & Protocol

Participants were 15 adults (36.3 +/- 9.8y, 11 women/4 men) with typical or corrected-to-typical vision. The study was approved by the National Ethical Committee and all





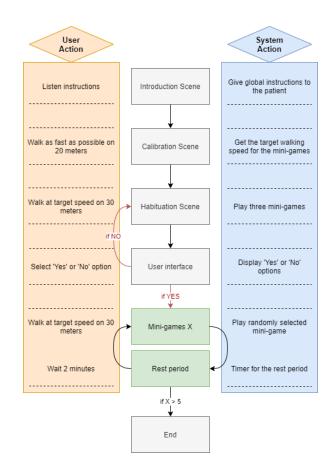


Figure 2. Protocol and flow of the application. Introduction scene - the global aim of the session was presented to the participant. This scene contained holograms (a 'Welcome to the game' message surrounded by a butterfly flying between trees) to familiarize participant with the hologram. Calibration scene the instruction given to the participant was to walk as fast as possible and this speed was averaged over 15 meters to define the target speed for the mini-games. Habituation scene - three mini-games were presented to test the comprehension of the participant. They were asked to walk at their target speed helped by the augmented feedback provided by the hologram. Interface user - a message was displayed in front of the user asking if they were ready to start the game session. Users said yes if they were ready to play, or no if they were not. Mini-game Scenes – 5 mini-games were presented once in random order, as well as a control scene without feedback. In each mini-game, participants had to walk 30 meters with the oral instruction of walking at their target speed.

participants completed written consent. The protocol is detailed in Figure 2.

# Evaluation Criteria

- 1. WS was calculated with the AR HMD (100Hz). The three first and last meters (acceleration/deceleration) were excluded from the analysis.
- 2. A questionnaire with a 5-point Likert scale from 1 to 5 (strongly disagree/agree) was completed to assess this experiment.

# Statistical analysis

After checking the stochastic assumptions, within-subjects' ANOVA for repeated measures and Dunnett test for multiple comparisons were performed for comparing experimental values (i.e., during each mini-game) and a single control value. Descriptive statistics were reported to describe participant feedback collected via questionnaires.

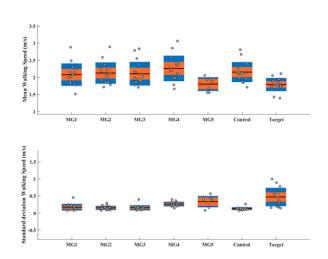
# **Results and Discussion**

# Walking speed

The mean WS during each mini-game and the control scene and target speed were not statistically different (F(5,76)=2.31,p=0.052) (Figure 3). These results are close to being significant, more participants are required to confirm the trend observed. Dunnett test revealed indeed that the mean WS during MG4 was significantly higher in comparison to the target WS. All mini-games helped the participant to exceed the target WS. There was a significant difference in the standard deviation of WS during each mini-game and the control scene (F(5,76)=9.39,p=5.37.10-7) (Figure 3). Dunnett test revealed that MG4 and MG5 resulted in a high variability in WS compared to the control scene (respectively 0.26 m/s, 0.33 m/s, 0.13 m/s). Of note, the frame of reference of feedback in MG4 and MG5 was world-locked as opposed to being presented relative to the user's head orientation. Due to the field of view  $(30^\circ)$ , some participants may have had to turn their head to follow holograms. This might have influenced speed variability. Other reason could be the aim of the mini-game. In MG4, some participants wanted to race with the butterfly, that may lead to speed variability as when some reached it, they slowed down. So, changing the spatial representation of MG4 from world-locked to body-locked could be a way to decrease this high variability.

# User questionnaire

Each participant successfully understood the instructions during mini-games (median 5/5,IQR 0). None had previous experience with AR. Participants reported a high level of satisfaction about the game session (5/5,IQR 1). They rated the mini-games to be sufficiently efficient to reach the target speed (4/5,IQR 1). About the Hololens HMD, they did not report any limitations for walking (5/5,IQR 0), but they highlighted discomfort due to the weight (579g) bearing on the nose. Participants also reported some confusion about the MG5. Of note, certain participants hesitated to catch the token, particularly when it appeared in virtual trees. For



**Figure 3.** Comparison of mean speed and variability during mini-games and control scene. The mean WS given in m/s was respectively 2.08 in MG1, 2.13 in MG2, 2.11 in MG3, 2.26 in MG4, 1.80 in MG5, 2.15 in the control scene and 1.79 for the target speed.

participants, the ranking of the best mini-games was MG2, MG4 and MG3 in a tie, MG1 and last MG5.

# Conclusion

This paper presented the collaborative design and evaluation of AR mini-games with visual feedback for gait training. This investigation demonstrated that gait parameters and user experience can vary depending on the type of AR feedback presented. Some feedback modalities increased WS whereas others had a larger impact on speed variability. Specific recommendations based on learnings in this study include using knowledge of results feedback to create a more challenging task that motivates participants to excel, bodylocked holograms that are easier to track, and clarify game presentation. Currently, a new version of the game based on the MG4 but with body-locked characteristics, is being developed that will be tested with people with ND.

# Acknowledgements

Authors would like to thank all participants and people involved in this work. This study is a part of the ARRoW CP project supported by the Ellen Poidatz Foundation and Association Nationale Recherche et Technologie (ANRT), in collaboration with the University of Paris-Saclay, University of Evry, IBISC, Team IRA2. The software development has benefited from the expertise of PEARL Lab team, Bloorview Research Institute, Holland Bloorview Kids Rehabilitation Hospital.

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# 4.4 A prospective study on the impact of virtual feedback on walking speed on children with CP (Articles 5 & 6)

This study was the next logical step from the previous research. The preliminary results on healthy adults helped us improve our classification of the different feedback characteristics that could affect walking rehabilitation efficiency and design more carefully the scenarios tested here. We have replaced the term "mini-games" with "scenario", because we have simplified the visual feedback to exclude the gamification effect (visual feedback was a round shape). The objective of this study was to define the most practical combination of visual feedback characteristics delivered in augmented reality to reach, maintain or exceed a target speed for people with cerebral palsy.

This study was a crossover trial with a repeated measures design in which each patient was assigned to a sequence of 7\*2 walking trials. Participants played seven scenarios, each twice. Each scenario presented visual feedback with different characteristics; one was control without visual feedback. Two trials by scenarios have been recorded, with two separate instructions: 1. "Walk as fast as possible for 30 m" (condition MAX) and 2. "Walk at your intermediate walking speed for 30 m" (condition INTER). The device used was the Microsoft Hololens augmented reality headset. For each condition (MAX and INTER), the feedback always displayed the current speed relative to the target speed. For example, during scenario 2, in condition INTER, the round shape appeared red to the user if his current walking speed is inferior to his INTER target speed (fail). It seemed green if his current walking speed was above the INTER target speed (success). For more details about the other scenarios, see the complete article below. For each condition (MAX and INTER), we assessed the following outcomes: Mean speed, Percentage of time spent above the target speed, time to get the target speed. Qualitative information was extracted from the Moizer user's questionnaire.

A total of 25 participants was included (mean age: 14.7 years, 14 girls/11 boys). In condition MAX, mean speed during control scenario was the lowest speed (1.430 +/- 0.317 m/s), and was inferior to target WS (1.482 +/- 0.361 m/s). Mean speed for all scenarios, including feedback, was superior to the target WS. The mean speed during scenario 6 was significantly higher (1.621 +/- 0.355 m/s) than in the control scenario. For time spent above target speed, participants succeeded significantly more during scenario 6 (81% +/- 15.5%) than in other scenarios. The qualitative analysis showed that scenarios 1, 3, 4, and 5 helped achieve speed goals compared to the control scenario. Scenarios 6 and 5 significantly decreased the time to reach target speed (respectively 1.28 s +/- 1.11 s and 1.75 s +/- 0.86 s) in comparison to the control scenario (control 3.31s +/- 1.35 s).

In conclusion, real-time visual feedback delivered through an AR headset is a feasible and acceptable intervention to provide immediate positive changes in walking speed for children with cerebral palsy. The feedback characteristics impact the results. A scenario combining a gradient color with a fixed spatial anchor, disappearing according to the time, demonstrated a significant increase in walking speed. Overall, the real-time feedback can provide many advantages as a gait training intervention: it could be implemented in an active and challenging video game; it received a positive evaluation from the participants as it is engaging and easy to understand; and most importantly, it provides unique concurrent information on walking performance that would otherwise be very difficult or even impossible for clinicians to deliver. We used these results to design our active video game: ARRoW-CP (See Chapter 5).

This was the second study conducted during this thesis work to test an AR application (BestOf ARRoW v2).

The preliminary results of this study have been published in *IEEE International Symposium on Mixed and Augmented Reality* on October 2020 [134] (Appendix 5.6). The overall results were synthesized into an article and submitted to *IEEE Transactions on Neural Systems and Rehabilitation Engineering* on October 18, 2021. This work has been presented at the 29<sup>th</sup> Société Française de Médecine Physique et de Réadaptation Congress in October 2021.

This paper is under review in *IEEE Transactions on Neural Systems and Rehabilitation Engineering.* We have attached the second version of the submitted article.



Anne-Laure Guinet, Guillaume Bouyer, Samir Otmane and Eric Desailly

Abstract-In an augmented reality environment, the range of possible real-time visual feedback is extensive. This study aimed to compare the impact of six scenarios in augmented reality combining four visual feedback characteristics on achieving a target walking speed. The six scenarios have been developed for Microsoft Hololens augmented reality headset. The four feedback characteristics that we have varied were: Color; Spatial anchoring; Speed of the feedback and Persistence. Each modality could had different values (for example, the color could be unicolor, bicolor or gradient). Participants had to walk for two consecutive walking trials for each scenario: at their maximal speed and at an intermediate speed. Mean speed, percentage of time spent above target speed and time to reach target speed were compared between scenarios using mixed linear models. A total of 25 children with disabilities have been included. Feasibility and user experience were excellent. Mean speed during scenario 6, which displayed a feedback with a gradient color, attached to the world, with a speed relative to the player equal to his own speed, and that disappeared over time, was significantly higher than other scenarios and control (p=0.003) for both target speeds. When the target speed was maximum speed, participants spent 80.98% of time above target speed during scenario 6. This scenario mixed the best combination of feedback characteristics to maintain the target walking speed (p=0.0058). Scenarios 5 and 6, which share the same feedback characteristics for spatial anchoring (world-locked) and feedback speed (equal to the player speed), decreased the time to reach the target speed (p=0.019). Delivering multi-modal feedback has been recognized to be more effective for improving motor performance. Our results therefore shown that not all visual feedback had the same impact on performance. Further studies, with more included people, are required to test the weight of each modality inside each scenario.

Index Terms—Augmented reality, Assistive technology, Feedback, Gait disorders, Patient rehabilitation

#### I. INTRODUCTION

Cerebral palsy (CP) describes "a group of permanent disorders of the development of movement and posture, causing activity limitation, which are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain" [37]. It is the most common cause of childhood disability, affecting 17 million people worldwide [18, 38]. Gait pattern functions are often altered (spastic, stiff or/and hemiplegic gait) causing mobility restriction. The natural

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history in people with CP is a gradual decline in ambulatory function as children grow and age [4, 33]. One effective approach to reverse this tendency is gait training (GT) showing beneficial effects on walking speed, endurance and other gait-related outcomes, with positive long-term effects [7, 43]. In order to optimize motor recovery, current motor learning theories recommend task-specific, variable and high intensity of practice but also the use of augmented feedback during therapy sessions [9, 34, 41, 31]. In paediatric rehabilitation, fun and motivation are also critical keys to successful therapy [35]. To this end, studies have demonstrated high level of interest, compliance and engagement with game-based intervention and virtual rehabilitation [28, 8, 23]. Virtual rehabilitation defined as "interventions that are built on virtual reality platforms to meet rehabilitation goals" are very efficient to provide concurrent feedback in real-time [26]. But active video games developed for these systems do not always integrate motor learning principles, including optimal feedback [13]. Feedback retraining paradigm is based on the conversion, the supplementation and augmentation of sensory information that are usually accessible only by an internal focus of attention, into accessible information [26, 32]. Augmented feedback is defined as augmented sensory information provided by an external resource (therapist or display) to the patient. The information provided to the user could refer to the movement's pattern itself or result on the environment or the outcome of a movement with respect to the goal [39]. Sensory channels usually used to deliver information are visual, auditory or haptic, in addition to the proprioceptive channel. The timing of feedback delivery is critical. Concurrent feedback is delivered while the skill is being performed, terminal feedback is delivered after the skill is performed with or without delay [36]. When using an augmented reality (AR) or virtual reality (VR) headset, there are many ways to provide the augmented feedback. In most studies, even if feedback is effective to improve motor activities, the characteristics applied during interventions were generally inconsistent with motor control feedback theory [29, 15]. Therefore, a recent systematic review highlighted that visual and auditory feedback were provided in all studies as a display of total score and/or reward sounds without any indication about movement characteristics (knowledge of performance) [14]. The objective of this study was to define the most effective combination of visual feedback characteristics delivered in augmented reality to reach, maintain or exceed a target speed for people with cerebral palsy.

## **II. PRELIMINARY WORK**

In a previous article, we proposed a model to describe the characteristics of feedback dedicated to the rehabilitation of walking in AR [21]. In an AR environment, feedback take the form of 3D holograms and sounds, whose spatial location has a considerable impact on their perception by users. The feedback can be attached to the world (i.e. the feedback has its own spatial coordinates in the world and stays in its place even if the player moves) or to the player (i.e. when the player moves, the feedback follows her/him). As a consequence, the position and speed of the feedback change: the feedback could translate at a predefined target speed, or at the player speed, or be stationary in the world. Our preliminary qualitative study on healthy adults showed that feedback characteristics displayed in an AR game influence the walking speed. Some feedback seemed to help to maintain or to exceed the target speed while others resulted in a high variability in walking speed compared to the control scene without feedback. These preliminary results helped us to improve our classification of the different feedback characteristics that could have an effect on walking rehabilitation efficiency [19] and to design more carefully the 6 scenarios tested here. Based on these previous results, we have also decided to test 2 different walking speed: a maximal walking speed and intermediate walking speed (mid-point between self-selected and maximal speed). The main reason is that our research on feedback characteristics is part of a global project aiming to improve walking rehabilitation in children with CP using Augmented Reality technology. In this context, therapists need to know what the best feedback characteristics are for both fast and sustained walk.

# III. METHODS

# A. Design study

This study was a crossover trial with a repeated measures design in which each patient was assigned to a sequence of  $7 \times 2$  walking trials. Participants played 7 scenarios, each twice. Each scenario presented a visual feedback with different characteristics, one of them was a control without visual feedback. Two trials by scenarios have been recorded, with two different instructions: 1. "Walk as fast as possible for 30 m" (condition MAX) and 2. "Walk at your intermediate walking speed for 30 m" (condition INTER). For each condition (MAX and INTER) the target speed was not the same. To calculate these 2 target speeds used after in the study, each session began with a calibration scene where the child had to walk as fast as possible for  $2 \times 15$  m (the MAX target speed was defined as the maximum average speed). Then, the child walked 30 m at self-selected walking speed (the INTER target speed was defined as the midpoint between the MAX target speed and the average speed during this self-selected trial). The explanation of "intermediate speed" was given to the participant before start the first scenario. To avoid the "order" effects, all participants walked during the same number of walking trials, in random order, and participated for the same number of periods (Figure 1). The instructions were standardized by playing a recorded human voice in the AR application.

#### B. AR Application: Best-Of ARRoW

*a) System characteristics:* Device used was Microsoft Hololens augmented reality headset (Microsoft, US). Microsoft HoloLens (1<sup>st</sup> generation) is the world's first fully untethered holographic computer, using Gaze tracking, Gesture input and Voice support to understand user actions (Figure 3). But also, Spatial sound to understand the environment. It's an Augmented Reality Head-Mounted Display of 579 grams, equipped with Processor Intel 32-bit architecture with TPM 2.0 with Custom-built Microsoft Holographic Processing Unit, connected with WiFi and Bluetooth. The Microsoft Hololens headset included also an inertial measurement unit (IMU), 4 environment understanding cameras, 1 depth camera and a photo/video camera, 4 microphones, 1 ambient light sensor. Best-Of ARRoW application has been developed with Unity 2019.2.8f1 (64-bit) using Mixed Reality Toolkit version 2. Microsoft Hololens tracking was accurate enough to measure the position of the user without time drift [20]. An

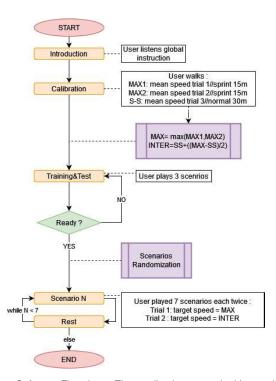


Fig. 1. Software Flowchart. The application started with a welcome message showing the holographic environment. The aim of the clinical study was explained to the patient through oral instruction.. Then, the calibration scene consisted of 2\*15 m walking sprints at maximal speed and 1\*30 m walking trial at self-selected speed, with no feedback except startup vocal instruction. The calibration gave 2 values that used as individualized target speed for the patient: MAX-the maximal walking speed and INTER-the intermediate walking speed. Three mini-games were then played for the warm-up and adaptation with the system and the holograms. When he/she was ready, the child decided to start the clinical study. The first scenario was displayed in random order. Each trial consisted of walking on 30 m in a straight line at the target speed (MAX and INTER), with one of the 6 scenarios including different feedback characteristics or without feedback (control). Each trial was followed by a rest period.

algorithm (called HoloStep) was developed specifically for measuring the real-time gait parameters from the head pose of children with CP: walking speed, cadence, step length and global distance travelled. Metrics such as sensitivity, specificity, accuracy and precision for step detection with HoloStep were above 96%. The Intra-Class Coefficient between steps length calculated with HoloStep and the reference was 0.92 for children with GMFCS I, and 0.86 for children with GMFCS II or III. [22].

b) Development framework: This work followed the serious game development framework PROGame, proposed by Amenguai Alcover et al [2]. The multidisciplinary team was composed by therapists (3 physiotherapists), researchers (2 in computer science, 1 in rehabilitation science, 1 in movement science) and a software engineer. The 6 scenarios that we have tested were (Table I):

- Scenario 1: A blue round shape moved ahead at the target speed;
- Scenario 2: A round shape moved ahead at the target speed, color changed if the user succeeded (green) or failed (red) to reach the target speed;
- Scenario 3: A round shape moved ahead at the target speed, color changed with a gradient (green-yellow-orange-red) depending on his own speed;
- Scenario 4: A round shape stayed 3 m in front of the user, color changed if the user succeeded (green) or failed (red) to reach

the target speed;

- Scenario 5: A round shape stayed at the end of the corridor, color changed if the user succeeded (green) or failed (red) to reach the target speed;
- Scenario 6: Five green round shapes were positioned every 5m, color changed with a gradient and the shape disappeared if the user failed to reach them on time.

For each condition (MAX and INTER), the feedback always displayed the current speed in relative to the target speed. For example, during scenario 2, in condition INTER, the round shape appeared red to the user if his current walking speed in bellow his INTER target speed (fail). It appeared green if his current walking speed is above the INTER target speed (success). The model of feedback used to develop these specific scenarios have been inspired by Macintosh et al. [29]. We have adapted their model to the AR environment. The visual sensory channel of the feedback could be characterised by the shape, the size, the persistence, the material, the shade and the color. In this study, we deliberately developed feedback with a simple design using elementary geometric shapes (round shape), with the same size, the same material and shade properties. We have introduced variations between scenarios to test the effect of color and persistence characteristics. Color could be unicolor (round shape was blue all the time), bicolor (round shape was green if the child exceeded his/her target speed, red if he/she didn't) or Gradient (round shape color changed from green to red, through orange and yellow depending on the distance from the target speed. The persistence characteristic was defined as the part of the shape that was visible for the user over time. Scenarios 1-5 were "full" that meant that the round shape was always visible. Scenario 6 was classified as "faded" because the round shape disappeared over time. The spatial representation of the feedback could be characterised by the position, the spatial anchor and the speed. For spatial anchor property, the feedback could be body-locked (i.e. attached to the gaze user) or world-locked (i.e. relative to the environment). In the Scenarios 1-3, feedback speed relative to the player was "Target speed - Player Speed", the feedback was moving ahead at the target speed and the player had his own speed. In this case, speed relative to the world was "target speed". In the Scenario 4, the feedback was placed 1 m in front of the user, so the user perceived null speed relative to him while he was walking, but speed relative to the world was "player speed" (a outside observer who was standing still could see that the feedback was moving ahead at target speed). In scenarios 5 and 6, the feedback was placed at fixed position in the world. Following the same logic, the speed relative to the player was "player speed" and the speed relative to the world was "null". So, feedback characteristics differed only by their color and filling variation, spatial anchoring, and relative moving speed. Figure 2 illustrated some feedback used. Multimedia Video is available online https://youtu.be/P\_fwBqIbqZU.

#### C. Participants and data collection

Participants were recruited from a pediatric rehabilitation center (Fondation Ellen Poidatz - Saint Fargeau). The inclusion criteria were: a clinical diagnosis of spastic CP, including hemiplegia, diplegia, and quadriplegia; age between 12 and 18 years; Global Motor Function Classification System (GMFCS) levels I-III; a minimum score of 2 on the Functional Mobility Scale 50m; ability to cooperate, understand and follow simple instructions to play the game; voluntary patient whose parents have given their free and informed written consent for their child's participation in the study. This study occurred between September 2020 and March 2021. Each participant wore the AR headset and followed the instructions given by the application.

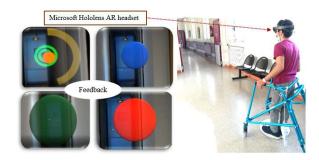


Fig. 2. Application Best-Of ARRoW. Example of 4 scenarios played in real-time with the Hololens during walking sprints. Top left: Scenario 6; Top right: Scenario 1; Bottom left: Scenario 2; Bottom right: Scenario 4

Sc	Color	Spatial anchor	Speed relative to the player	Speed relative to the world	Persistence
S1	unicolor	Body	TS - PS	TS	full
S2	bicolor	Body	TS - PS	TS	full
<b>S3</b>	gradient	Body	TS - PS	TS	full
S4	bicolor	Body	null	PS	full
S5	bicolor	World	PS	null	full
<b>S6</b>	gradient	World	PS	null	faded
<b>S7</b>	none	none	none	none	none

#### TABLE I

FEEDBACK CHARACTERISTICS FOR EACH SCENARIO. Feedback was given on current walking speed relative to the target speed. Unicolor: Round shape was blue all the time; Bicolor: Round shape was green if the child exceeded his/her target speed, red if he/she didn't; gradient: the color changed from green to red, through orange and yellow depending on the distance from the target speed; Body: Feedback attached to the player; World: Feedback attached to the world; Persistence: the part of the feedback that was visible for the user over time. Sc: Scenario; TS: target speed; PS: player speed; Scenario 7 (S7) was the control without any feedback

#### D. Outcome Assessment

During the session, raw data were logged (100Hz) in .txt format through the application and were available in the Windows Device Portal. Log file contained time (s), position x, y and z of the headset, step length and distance travelled calculated with HoloStep algorithm [22]. Outcome were: Mean speed (condition MAX and condition INTER), percentage of time spent above the target speed (condition MAX and condition INTER), time to get the target speed (condition MAX and condition INTER). The target speed was not the same for condition MAX and condition INTER (see Design study in section III.A. for more details). At the end, participants completed a questionnaire rating their experience (feasibility and user experience evaluation) [30].

#### E. Data Analysis

Data analysis was performed using MatLab version 9.6.0.1472908 (R2019a) Update 9. Raw data (time, position, step length) were filtered with Butterworth filter design (Filter order 2, Cutoff frequency 4 Hz) using *filtfilt* function. For each participant, trials were cut off three steps before the end to not consider deceleration. The instantaneous walking speed was calculated using position and re-filtered using polynomial curve fitting (order 1, framelen 701). Statistical analysis was performed using R version 4.0.5. Mean speed, percentage of time spent above the target speed and time to get the target speed were compared between *scenarios*. The different *scenarios* were compared using mixed linear models, including the patients as random effects

Sample Characteristics	Children $(n = 25)$		
Age (mean (SD))	14.7 (1.6 years))		
Sex (F/M)	14/11		
GMFCS	I:15 II:10		
Calibration Results	Children $(n = 25)$		
Speed MAX (mean (SD))	1.482 (0.361 m/s)		
Speed INTER (mean (SD))	1.295 (0.268 m/s)		
TABLE II			

PRELIMINARY INFORMATION. MAX: CONDITION MAX, INTER: CONDITION INTER

in order to take into account the repeated measures. To perform hypothesis testing on the fixed effects, parametric bootstrap tests based on the likelihood ratio test (LRT) statistic with 1000 iterations were used. This strategy was preferred to the classic as it introduce less bias. Tests were implement with the function PBmodcomp in the pbkrtest package (v.0.5.1). Statistical significance was determined at the 0.05 level throughout. Qualitative information was extracted to explore individual responses. Therefore, the percentage of time spent above the target speed was also analysed in function of the participant's target speed. Qualitative information was extracted from the user's questionnaire.

#### F. Ethical considerations

Ethics approval was granted by the Ethical Committee of Ile-de-France 1 in France (IRB/IORG : IORG0009918). Additionally, all parents and participants from 12 years of age, signed the informed consent prior to study initiation. All participants had a reflection period prior to the inclusion (minimum 15 days between information and consent). Confidentiality and data access were guaranteed by the National Commission for Data Protection. A Data Protection Officer has been designated for all research studies conducted in the rehabilitation center. He guaranteed that the data protection and the rights of the participants were respected according to the General Data Protection Regulation (European Union) 2016/679. This study was registered in the ClinicalTrials.gov database (NCT04460833).

#### **IV. RESULTS**

A total of 25 participants was included. Characteristics of population were detailed in Table II.

### A. Feasibility and user experience evaluation

All participants successfully completed the session. There was no missing data. No adverse effects such as difficulty in breathing, discomfort or cybersickness were observed during trials. None of the participants fell while walking and wearing the AR headset. The user experience questionnaire revealed that all participants properly understood the game instructions. All participants rated 5/5 on the items "I learned to use the game quickly" and "I understood the walking instructions easily". They thought that the game was immersive, fun and pleasant (items "The experience was challenging. I found the game stimulating", "The experience was immersive" and "The playing environment was visually appealing" rated 5/5). Some participants mentioned that in some scenarios it was less easy to move around because the feedback sometimes disappeared. All participants recognised the value of the game as a tool for learning (item "The game scenario had relevance to the issue of walking skills development" rated 5/5).

# B. Mean Speed

The average walking speed (WS) varies according to the *scenario* (Figure 3).

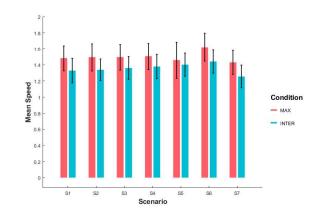
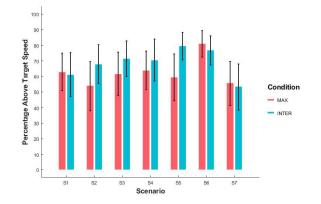
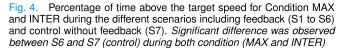


Fig. 3. Mean Speed in m/s for Condition MAX and INTER during the different scenarios including feedback (S1 to S6) and control without feedback (S7). *Significant difference was observed between S6 and S7 (control) during both condition (MAX and INTER)* 





a) Condition MAX: Mean speed during scenario 7 (control) was the lowest speed  $(1.430 \pm 0.317 \text{ m/s})$ , and was inferior to target WS  $(1.482 \pm 0.361 \text{ m/s})$ . Mean speed for all scenarios including feedback was superior to the target WS. The mean speed during scenario 6 was significantly higher  $(1.621 \pm 0.355 \text{ m/s})$  than in the scenario control (p; 0.05). The first quartile (Q1) for speed during scenarios 1-6 was higher than the Q1 for target speed and control. The median shown that 50% of people had a WS above 1.555 m/s with the help of feedback provided during scenario 6 while median target speed was 1.407 m/s.

b) Condition INTER: Mean speed during scenario 7 (control) was the lowest speed ( $1.256 \pm 0.286$  m/s), and was inferior to target WS ( $1.295 \pm 0.268$  m/s). All scenarios including feedback allowed to achieve the speed objective. The mean speed during scenario 6 was significantly higher ( $1.440 \pm 0.273$  m/s). The first quartile (Q1) for speed during scenarios 2-6 was higher than the Q1 for target speed and control. The median shown that 50% of people had a WS above 1.373 m/s with the help of feedback provided during scenario 6 while median target speed was 1.322 m/s.

#### C. Percentage of time above target speed

The percentage of time above target speed varies according to the *scenario* (Figure 4), and the participant target speed (Figure 5).

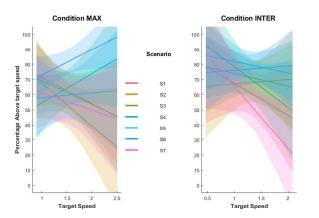


Fig. 5. Percentage of time above the target speed for Conditions MAX and INTER during the different scenarios including feedback (S1 to S6) and control without feedback (S7). *In condition MAX, faster walkers better performed with scenarios 5 and 6 whereas slower walkers spent more time above target speed with scenarios 1-2-3 and 6.* 

a) Condition MAX: Participants succeeded significantly more during scenario 6 ( $81\% \pm 15.5\%$  of time spent above target speed) than in other scenarios. Qualitative analysis shown a tendency that scenarios 1, 3, 4 and 5 helped to achieve speed goal in comparison to the scenario 7 (control without feedback). Particularly, 75% people spent at least 73.1% of time above target speed in scenario 6 whereas 75% people spent 20.9% of time above target speed without feedback. 'Faster walkers' (i.e. participants who had a higher target speed from the calibration) responses to scenarios were more heterogeneous (Figure 5): their performance were better with scenarios 4 and 6 (sup to 80%).

b) Condition INTER: Achieving the speed target was significantly impacted by the *scenario*. However, *scenario* 1 was the less effective (mean time spent above target speed was  $61.2\% \pm 29.6$ ) against *scenario* 5 (79.5\%  $\pm$  13.1) and *scenario* 6 (76.8\%  $\pm$  15.4). But, all *scenarios* helped to improve performance against *scenario* 7 without feedback (53.4%  $\pm$  30.7). Particularly, 75% people spent at least 76.6% of time above target speed (Q1) in *scenario* 5 whereas 75% people spent 7.8% of time above target speed without feedback. 'Slower walkers' (i.e. participants who had a lower target speed from the calibration) seemed to improve be less sensitive than 'faster walkers' to the feedback (Figure 5). 'Faster walkers' performances were better with feedback (20% against 50% and more).

#### D. Time to Reach Target Speed

The time to reach target speed varied according to the *scenario* (Figure 6).

a) Condition MAX: The scenarios 6 and 5 significantly decreased the time to reach target speed (respectively  $1.28 \text{ s} \pm 1.11$  s and  $1.75 \text{ s} \pm 0.86 \text{ s}$ ) in comparison to the scenario 7 (control 3.31 s  $\pm 1.35$  s). Qualitative analysis revealed that in the scenarios 2-6, 75% people reached the target speed in less than 2 s.

b) Condition INTER: The scenario 6 significantly decreased the time to reach target speed (1.86 s  $\pm$  0.76 s) in comparison to the scenario 7 (control 2.94 s  $\pm$  0.82 s). Qualitative analysis revealed that in the scenarios 2-6, 75% people reached the target speed in less than 2 s.

#### E. The Ranking of scenarios

Comparing the three outcomes (mean speed, percentage of time above target speed and time to reach target speed) across *scenarios* 

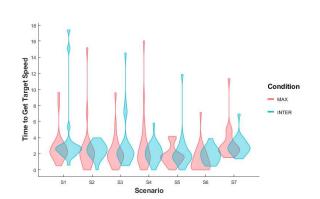


Fig. 6. Time to Reach the Target Speed for Condition MAX and INTER during the different scenarios including feedback (S1 to S6) and control without feedback (S7). *In condition MAX, Scenarios 5 and 6 significantly decreased the time to reach target speed. Outliers shown people who did not reach their target walking speed in a short time.* 

Scenario	Speed	% above TS	Time to get
S1	1.482 [5]	62.9 [3]	2.83 [5]
S2	1.494 [4]	53.9 [7]	2.69 [4]
<b>S</b> 3	1.495 [3]	61.7 [4]	2.25 [3]
<b>S4</b>	1.505 [2]	63.9 [2]	2.97 [6]
S5	1.459 [6]	59.5 [5]	1.75 [2]
S6	1.621 [1]	81.0 [1]	1.28 [1]
S7	1.430 [7]	55.6 [6]	3.31 [7]

TABLE III

THE RANKING OF SCENARIOS Mean Speed, Mean percentage of time above target speed and mean time to reach target speed for each scenarios: mean value and [ranks]. TS: target speed. Pink box : mean speed was superior to the mean target speed

during condition MAX, we established a ranking (Table III). All *scenarios* except S5 and control without feedback (S7) had a mean speed above the target speed. The "top 3" *scenarios* to spent more time above the target speed are S6, S4 and S1 (> 62% of time above target speed). All *scenarios* were better than the control S7 for the three outcomes.

#### V. DISCUSSION

This study explored the impact of augmented feedback characteristics to help people with motor disabilities reaching and maintaining a target walking speed using AR headset. Based on the theoretical model, several scenarios combining different feedback characteristics were tested.

## A. Feedback in AR to improve gait parameters

In augmented reality, real physical and digital objects co-exist in space and interact in real time [16]. In other words, users perceive the position and movement of the hologram in relation to their own movement and the real world around them. This option allows us to offer walking rehabilitation for disabled people in a real environment, they can walk with their natural walking pattern and their own walking aids. Augmented feedback characteristics could be multiple. In rehabilitation context, the content of the feedback could vary according to the aim of the therapy, for example improving specific kinematics or kinetics parameters. This study raises the question of the best combination of feedback modalities to help people reaching and maintaining a target walking speed. This question has not been sufficiently explored and the choice of the scenarios was based in particular on our previous results on healthy adults [19]. Some feedback modalities increased walking speed whereas others had a larger impact on speed variability. Specific recommendations from this previous study include using knowledge of results focusing on the outcome of a movement to create a more challenging task that motivates participants to excel, body-locked holograms that are easier to track, and clarify game presentation. Previously, Baram et al. trained 10 patients with CP with visual feedback. They used a display, attached to the eyeglasses frame, providing image of transverse lines, responding dynamically to the patient's own motion [3]. They showed that walking speed measured along a straight track of 10 meters was improved after 20 minutes of training using a this very simple visual feedback (+21.70  $\pm$  36.06%). This improvement was higher for participants with baseline walking speed below the median (+35.75  $\pm$  ±47.76%). Using our model of feedback, we could classify their feedback as {color = unicolor, spatial anchor = World-locked, speed relative to the player = player speed, speed relative to the world = null, filling = full  $\}$  which is very close to the scenario 6 characteristics. Our results showed that success depends on children gait speed during calibration and feedback characteristics. For percentage spent above target speed, even if it was only qualitative analysis, we observed on Figure 5 that children with baseline walking speed below the median ('slower walkers') presented more homogeneous results than faster walkers. Scenarios with feedback helped them, specially in condition MAX, but there were no scenario that surpassed the others. By contrast, for faster walkers, the feedback characteristics had an impact on their performance. In scenario 6, percentage spent above target speed is near 100% for these children which barely reached 20% of that amount in scenario 3. Providing a relevant feedback to improve performance, and specifically walking speed, is a key point for rehabilitation purposes. In stroke patients, prescriptive feedback (describing the errors and suggesting how to correct them) was found to be more effective than descriptive feedback (just describing the errors) [42]. Feedback delivered in real-time with AR headset could be implicitly prescriptive, by playing on challenges, rewards, motivation and conviviality. In scenario 6, if the user doesn't catch the round shape ("the coin") in time, it disappears. So he can easily understand that he is too slow (description of the error) and if he wants to win he should speed up (suggestion to correct the error). Our results highlighted that the modification of visual aspect ({color = *gradient*}) helped to improve walking speed. The feedback attached to the world with fixed position ({speed relative to the world = *null*}) seemed to be better to minimize visual discomfort and by extension fatigue. In fact, with movement there were some loss of tracking, conduced to unstable holograms, when the device cannot readily locate itself with respect to the world. A faded filling of the feedback acted as magnets for young people because of the games aspect and the challenge involved.

#### B. Individual responses to the feedback

The results of this study confirmed that people with CP could adapt their walking speed and positively responded to the real-time AR feedback [24]. However, we observed that not all patients performed equally well with the *scenarios*. If we looked at individual responses of each participants and during each *scenario*, the results were very different. Some people did not performed better with the feedback, other was helped by 1 *scenario* but disturbed in another *scenario*. Some authors highlighted these inter-individual differences. Slaboda et al. shown that the effect of continuous visual flow on the ability to regain and maintain postural orientation differed according the age

of participants [40]. Recently, Liu et al. have underscored different patient profiles: non-responders and responders to the feedback. In their study, patients were people after stroke. They were instructed to walk on a treadmill while visualizing on a large screen an avatar replicating their exact walking pattern in real-time. Overall, patient improved both step length and walking speed when the avatar is displayed on a side view. But results were not the same for all participants, authors distinguished non-responders and responders to the feedback. They hypothesized that the initial step length ratio could influence the result, because patients with a larger paretic step length better respond [27]. Booth et al. added that self-perception of walking, preference, cognitive ability and previous experience with feedback could be other factors that influenced the results [6]. These studies have shown that certain populations are more susceptible to the virtual environment and, therefore, will respond differently than predicted. The potential of VR/AR systems and feedback are encouraging, and more detail on individual response could be interesting to optimize VR programs for individual people.

#### C. Hololens Limitations

The Hololens AR headset presents some technical limitations [11]. First, during trials, children mentioned the restricted field of view (FOV) of the screen used to display the holograms (approximately 30°x 17°). Human's have general static view of about 135 to 180 degrees horizontally, with about 120 degrees of binocular vision. With eyeball rotation (about 90 degrees) the field of view extends to 270 degrees. In addition, vertical field of vision for humans is about 50 degrees in the upper visual field and 70 degrees in the lower visual field. Hololens FOV is much smaller and estimated to be about 30 degrees wide and 17.5 degrees high. So, the Hololens occupies a small portion of human vision, and there is a specific point where objects reach the end of the screen and disappear. Secondly, Coolen et al. compared the obstacle-avoidance maneuvers of participants stepping over either real or holographic obstacles of different heights and depths. They showed that the number of observed obstacle collisions were much higher for holographic than for real obstacles. They noticed for a participant, large unnatural head adjustments in order to view the holographic obstacle while crossing, he or she had to make[12]. Moreover, wearing a head-mounted display while walking has an impact on kinematics and performance on a standard clinical test of dynamic balance (Time Up and Go Test) [1]. The headset can limit peripheral vision and increase head movement to explore the environment [25]. Finally, children reported that the HoloLens is not very comfortable with a weight of 579 g.

#### D. Clinical Application

In recent years, new technologies have been introduced in rehabilitation practice, both for upper and lower limbs therapy. To 'actively practice the task of walking', systems combining treadmill training and exergame delivered through a television screen in a semiimmersive environment has been tested. Results on patient with CP showed good adherence and improvement both in walking speed on 10 meter walk test and distance travelled in the 2-minute walk test [10]. Recently, an innovative treadmill platform based on immersive virtual reality through a 180° semi-cylindrical screen (GRAIL from Motek) provided promising results both on Gross motor function, endurance and walking speed on children with CP [5, 17]. Both these systems offer a possibility to provide high-intensity training in a multi-modal training environment and variable practice but also to increase motivation level [39]. However, motor learning principles are not always fully integrated into VR/AR systems because of the lack of knowledge about which feedback modality and which

intensity level should be provided in rehabilitation settings [14]. Only 42% of custom VR systems for rehabilitation delivered multi-sensory feedback that combined visual and auditory and/or haptic feedback [14] whereas a combination of multi-modal feedback is recommended to be more effective for improving motor performance [39]. Our results bring information about effective visual feedback improving gait parameters. Next step of our project is to develop and test an active video game in AR for gait rehabilitation based on these first results including visual and auditory feedback.

#### E. Study Limitations

Each session had a limited number of walking trial to ensure maximum quality, so there were a limited number of *scenarios* that have been tested. All participants performed 14\*30 meters. This was chosen because of the restricted endurance of the patient with CP. Moreover, the long-term effect of feedback intervention has not been tested in this study. But, a randomized control trial (4-week protocol) with an active video game in AR based on these findings is in progress.

#### VI. CONCLUSION

Real-time visual feedback delivered through an AR headset is a feasible and acceptable intervention to provide immediate positive changes in walking speed for children with cerebral palsy. The feedback characteristics have an impact on the results, a *scenario* combining a gradient color, with fixed spatial anchor, disappearing according the time demonstrated significant increase in walking speed. Overall, the real-time feedback can provide many advantages as a gait training intervention: it could be implemented in an active and challenging video game; it received positive evaluation from the participants as it is engaging and easy to understand; and most importantly, it provides unique concurrent information on walking performance that would otherwise be very difficult or even impossible for clinicians to deliver. In the future, clinical trials with multiple training sessions are needed to test the applicability of real-time feedback in the clinical setting.

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#### VIII. DISCLOSURE

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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# 4.5 Profile of Responders to AR Feedback

# Introduction

In our two previous studies (Articles 5 & 6 in Section 4.4), we have confirmed that children with CP can adapt their walking speed, and they can positively respond to the realtime AR feedback [135]. However, we have observed that not all patients performed equally well with the *scenarios*. When we have looked at the individual responses of each participant for each *scenario*, we have observed some essential differences. Some people did not perform better with the feedback; others were helped by a particular *scenario* but disturbed in another *scenario*.

Some authors highlighted these inter-individual differences. Recently, Liu et al. have underscored different patient profiles: "non-responders" and "responders" to the feedback. In their study, patients were people after stroke. They were instructed to walk on a treadmill while visualizing an avatar replicating their exact walking pattern in real-time on a large screen. Overall, patients improved step length and walking speed when the avatar was displayed on a side view. But results were not the same for all participants; the authors distinguished non-responders and responders to the feedback. They hypothesized that the initial step length ratio could influence the result because patients with a larger paretic step length better responded [136]. This study has shown that specific populations are more sensitive to the virtual environment.

We followed the same scientific approach as Liu et al. to explore if children with CP had the same responses regardless of the AR feedback presented. We can establish different profiles of individuals in response to AR feedback.

# Methods

To define the individual response to AR feedback, we have conducted a posthoc analysis with data from the experiment detailed in Article 6 (See Section 4.4). In this article 6, we have described six different *scenarios*. Given that our work was focused on the AR environment, we have used our model of feedback in AR (Section 4.2) to group the *scenarios* according to "Spatial Presentation  $\rightarrow$  Moving Speed" that was a particular characteristic of AR.

We have simplified the six scenarios into three categories of AR feedback :

- <u>FOLLOW</u>: Scenarios 1 to 3 were grouped the speed relative to the world was the target speed (*patient followed the round shape*);
- <u>HEAD</u>: Scenario 4 the speed relative to the world was the player speed (round shape was attached to the patient's head);
- <u>WORLD</u>: Scenarios 5 to 6 were grouped the speed relative to the world was null (round shape was fixed in the world).

The variable included in this posthoc analysis were mean speed and percentage of time above target speed (after this INDEX) and time to reach target speed (after this TIME). We had focused our analysis on the condition MAX when the target speed was the maximal speed of the participant (See Section 4.4). This choice was related to the gait rehabilitation protocol, including walking sprints in the ARRoW-CP study (See Chapter 5). The variables INDEX and TIME, for each feedback (FOLLOW or HEAD or WORLD) and without feedback (CONTROL) was calculated as follow:

<ol> <li>(Mean Speed FEEDBACK/Target Speed)*100</li> <li>(Mean Speed CONTROL/Target Speed)*100</li> </ol>	Ind $A = [1] - [2]$	Index_FEEDBACK = mean (Ind A, Ind B)
<ul><li>[3] Percentage of time above Target Speed FEEDBACK</li><li>[4] Percentage of time above Target Speed CONTROL</li></ul>	Ind $B = [3] - [4]$	$\operatorname{Index}_{\operatorname{FEEDBACK}} = \operatorname{Inean} (\operatorname{Ind} \operatorname{A}, \operatorname{Ind} \operatorname{B})$
[5] Time to reach Target Speed FEEDBACK	[5]	Time_FEEDBACK
[6] Time to reach Target Speed CONTROL	[6]	Time_CONTROL

We have performed a **Correlation Analysis** using Pearson's coefficient to measure the linear correlation between variables, then a **Principal Component Analysis** (PCA) in order to see the variability between the individuals and if there were linear relationships between variables. Finally, we have performed a **Hierarchical Clustering on Principal Components** (HCPC) of a factorial analysis to gather the 25 individuals of our dataset into a couple of clusters which would correspond to different responders profiles. Data analysis was conducted in October 2021 using RStudio 2021.09.0 Build 351 with R version 4.1.0. Main package used was FactoMineR [137]\*. For vizualisation plot, we used GGally and ggplot2 packages.

#### Results

**Correlation Analysis.** Correlation matrix plot (Figure 4.3) showed a strong correlation for performance between feedback FOLLOW and HEAD (r = 0.74) but also between feedback HEAD and WORLD (r = 0.73). A lower correlation was found for performance between feedback FOLLOW and WORLD (r = 0.62). Otherwise, performance was higher in WORLD feedback condition (mean Index\_Follow = 10.44), then in HEAD feedback (mean Index\_Follow = 6.42), and FOLLOW (mean Index\_Follow = 3.70). It was interesting to observe that whatever the feedback, the time to reach the target speed was not related to the performance during the trial. In other words, reaching the target speed quickly is not correlated with performance (better walking speed and better time spent above the target speed). Moreover, time to reach the target speed according the feedback was not linked. We observed that mean time to reach the target speed was lower in WORLD feedback condition (mean Time\_World= 1.707 s), and higher in CONTROL condition (mean time\_Control = 3.307 s) [mean Time\_Follow = 2.632 s and mean Time\_Head = 3.400 s].

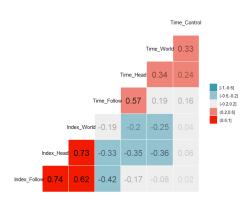


Figure 4.3 – Correlation matrix plot

<sup>\*.</sup> FactoMineR is an R package dedicated to multivariate Exploratory Data Analysis. It is developed and maintained by François Husson, Julie Josse, Sébastien Lê, d'Agrocampus Rennes, and J. Mazet.

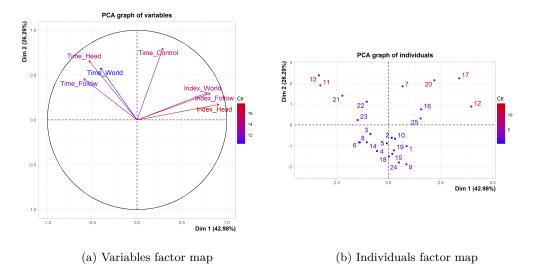


Figure 4.4 – Principal Component Analysis - Graph of Variables and Individuals

**Principal Component Analysis.** Handling missing values impute with 2-dimensional PCAmodel The first two dimensions of analyse expressed 69.27% of the total dataset inertia i.e. 69.27% of the individuals (or variables) cloud total variability was explained by the plane. This percentage was relatively high and thus the first plane well represented the data variability (Figure 4.4).

Correlation coefficient and significance test between each variable in the model and the first two dimensions of the PCA indicated that the variables Index\_Head (r = 0.91, p = 1.97e-10), Index\_Follow (r = 0.81, p = 6.69e-07) and Index\_World (r = 0.78, p = 3.70e-06) were the most correlated to the first dimension and that the variables Time\_Control (r = 0.80, p = 1.44e-06), Time\_Head (r = 0.66, p = 3.05e-04) and Time\_World (r = 0.58, p = 2.36e-03) were the most correlated to the second dimension.

The dimension 1 opposed individuals such as 12, 17 and 20 (to the right of the graph, characterized by a strongly positive coordinate on the axis) to individuals such as 21 and 13 (to the left of the graph, characterized by a strongly negative coordinate on the axis) (Figure 4.4).

The group in which the individuals 12, 17 and 20 stand was sharing the following characteristics:

 High values for the variables Index\_World, Index\_Follow, Index\_Head and Time\_Control (from the strongest to the weakest)

The group in which the individuals 21 and 13 stand was sharing the following characteristics:

- High values for the variables Time\_Head and Time\_Follow
- Low values for the variable Index\_Head.

The dimension 2 opposed individuals such as 12, 21, 17, 20 and 13 (to the top of the graph, characterized by a strongly positive coordinate on the axis, already described) to individuals such as 9, 24 and 15 (to the bottom of the graph, characterized by a strongly negative coordinate on the axis) (Figure 4.4).

The group in which the individuals 9, 24 and 15 stand was sharing the following characteristics:

 Low values for the variables Time\_Control, Time\_World and Time\_Follow (from the weakest to strongest) **Hierarchical Clustering on Principal Components.** Hierarchical Clustering (HCPC) as well as partitional clustering was performed on the principal components of the PCA using the Ward's method. PCA is thus considered as a prepossessing step before performing clustering methods. The Ward's method consisted in aggregating two clusters such that the growth of within-inertia was minimum at each step of the algorithm. The within inertia characterised the homogeneity of a cluster. The aim of the HCPC was to better highlight the main feature of the data set. Here, the

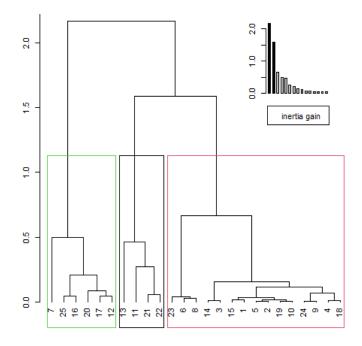


Figure 4.5 – **Hierarchical clustering on Principal Components.** The individuals are sorted according to their coordinate on the first principal component. Cluster 1: Black; Cluster 2: Red; Cluster 3: Green

PCA function kept the first two dimensions and thus the HCPC only used these two dimensions. The shape of the dendrogram (see Figure 4.5) suggested partitioning the individuals into three clusters.

The cluster 1 was made of individuals such as 11, 13 and 21. This group was characterized by :

- High values for the variables Time\_Head, Time\_World and Time\_Follow (variables were sorted from the strongest).
- Low values for the variable Index\_Head.

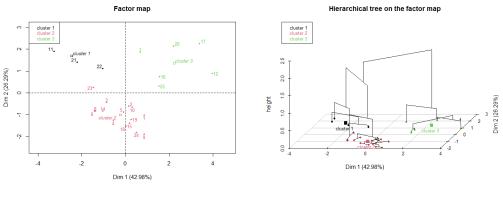
The cluster 2 was made of individuals such as 9, 15, 18 and 24. And was characterized by :

— Low values for the variables Time\_Control, Time\_Head, Time\_World, Index\_World and Index\_Follow (variables were sorted from the weakest).

The cluster 3 was made of individuals such as 7, 12, 17 and 20. And was characterized by :

 High values for the variables Index\_Head, Time\_Control, Index\_Follow and Index\_World (variables were sorted from the strongest).

The Hierarchical classification graphs (Figure 4.6) showed that the three clusters were wellseparated on the first two principal components. The individuals from cluster 1 were characterised by more time to reach target speed whatever the FEEDBACK was, but particularly for HEAD



(a) Factor Map

(b) Hierarchical Tree on the Factor Map

Figure 4.6 – **Hierarchical Classification of Individuals.** The classification made on individuals revealed 3 clusters. The hierarchical tree was drawn on the factorial map with the individuals colored according to their clusters. The barycentre of each cluster was also represented by a square.

(9.12 s on average for the individuals in this cluster compared with 3.15 s for all of the individuals) and large decrease in performance for HEAD feedback (-17.40 on average for the individuals in this cluster compared with 6.42 for all of the individuals). The individuals in cluster 2 were characterized by less time to reach target speed for HEAD and WORLD FEEDBACK (example for HEAD, 1.72 s on average for the individuals in this cluster compared with 3.15 s for all of the individuals) but also in CONTROL condition. The individuals in cluster 3 performed better with all FEEDBACK since the average INDEX\_HEAD of this cluster (33.41) was much higher than the average for all the individuals (6.42), the average INDEX\_FOLLOW of this cluster (26.27) was much higher than the average for all the individuals (3.70), the average INDEX\_WORLD of this cluster (40.99) was much higher than the average for all the individuals (10.44). This clustering has allowed the following refinements of the analysis helping us to define three categories of individuals.

#### Discussion & Conclusion.

A distinction has been made between three profiles of participants when confronted with AR feedback. They could be defined as follows :

- "Responders" who had a high performance i.e. high mean speed and high percentage of time above target speed, and who reached the target speed faster with feedback than in the control condition without feedback
- "Non Responders" who equally performed whatever the feedback was
- "Disturbed" who had a lower performance and longer time to reach target speed when the feedback was HEAD, and longer time to reach target speed for the other feedback condition.

Our results were following the literature. Liu et al. [136] have shown that particular stroke patients were more susceptible to the virtual environment and, therefore, responded differently to the feedback than others. Slaboda et al. showed that the effect of continuous visual flow on the ability to regain and maintain postural orientation differed according to the age of participants [138]. Booth et al. suggested that self-perception of walking, preference, cognitive ability, and previous experience with feedback could be other factors that influenced the results [139]. These studies have shown that specific populations are more susceptible to the virtual environment and respond differently than predicted. We can only formulate some hypotheses about our results. After the session, a particular patient reported some bug in *scenario* 5 (WORLD), that could explain the "Disturbed" profile. Moreover, we observed that some patients did not perform at their best during the calibration, making the task too easy to realize during walking sprints. This too easy task made the feedback useless to perform, which could explain the "Non Responders" profile. These parameters need to be further explored in future studies evaluating feedback to explain these different profiles of responders to AR feedback. Although this is an exploratory analysis, we have observed a low proportion of "disturbed" patients and a high proportion of "responders" patients that is very encouraging to develop the potential of AR systems and feedback for gait rehabilitation in children with CP.

# 4.6 Conclusion

Defining the term feedback was an essential part of our work. We provided a synthesis of feedback definition based on the literature, usable in a motor rehabilitation context. Augmented feedback is defined as augmented sensory information (visual, auditory, haptic, proprioceptive) provided by an external resource (therapist or display) to the patient. This definition allowed us to develop a feedback model in AR for motor rehabilitation with two sides: descriptive and qualitative. Our model is a practical extension of the theory of feedback, helping us formalize their characteristics. We have used this model to design better the feedback deployed in the ARRoW-CP game. Considering the complexity of the model and time constraint, we have not been able to test all the characteristics of our model. The choice of the tested modalities was made after multidisciplinary consultation, taking into account the results of the preliminary study on adults, but also across test campaigns with children. The results with children with cerebral palsy showed us that the choice of modalities, particularly spatial representation, was necessary since the feedback influenced the gait performance during the session. Three individual profiles emerged: the "responders" who improved their performance with the feedback, the "non-responders" who had an equivalent performance in the control situation and with the feedback, and the "disturbed" who presented a decrease in their performance with the feedback. The small number of "disturbers" patients and the high number of "responders" patients encouraged us to develop AR feedback in our AVG ARRoW-CP.

# Chapter 5

# ARRoW-CP: Active Video Game for Gait Training in children with Cerebral Palsy

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Chapter 5 describes our final active video game, called ARRoW-CP, which proposes a 4-weeks protocol with a series of walking sprints to be performed at high intensity. We precise the game development framework used to develop ARRoW-CP. Finally, we expose the clinical study protocol designed to test the efficacy of ARRoW-CP (*Article 7*). Today, we evaluate ARRoW-CP with children with CP at the Ellen Poidatz Rehabilitation Center. We present the preliminary results of this clinical study.

# 5.1 Introduction

The previous chapter has shown how essential it is to incorporate elements of motor learning theory into the rehabilitation strategies. In the active video game area, this implies designing the system with the users (therapists and patients) to adapt the objectives, constraints, levels of difficulty, feedback, and so on. In this chapter, we will justify the choice of the rehabilitation protocol that has been implemented in the active video game ARRoW-CP (Section 5.2). Then we will precise the game development strategy adopted with the users. We will describe precisely the mechanics and characteristics of the feedback deployed in the application (Section 5.3), by referring to our model of feedback in AR. We will present the protocol of the randomized controlled trial to evaluate the impact of our innovative AR rehabilitation strategy (Section 5.4). Finally, we will present the preliminary results of this study that is underway at the Fondation Poidatz (Section 5.5).

# 5.2 Gait Training Intervention

The intervention consisted of 4 weeks of overground gait training (OGT) with three sessions per week, including a series of walking sprints, through the active video game ARRoW-CP. This is an adaptation of the validated protocol from Zwinkels et al. showing strong effects in youth with CP (Section 2.3.2.1). The original protocol occurred on eight weeks and twice a week [48]. Every training session consisted of a series of 30 s walking sprints achieved at the prescribed intensity, volume, and time: Week 1-2: 8 sprints, work/rest ratio 1:4; Week 3-4: 10 sprints, work/rest ratio 1:4; Week 5-8: 12 sprints, work/rest ratio 1:3. Following their program, there were significant improvements in (MD: mean difference post-pre, d: effect size) : Anaerobic performance (Muscle Power Sprint Test - MPST) : MD = 19.2 Watts, d = 0.40; Agility (10x5 m sprint test) : MD = -1.9 s, d = 0.54; Aerobic performance (Shuttle Run Test - SRT) : MD = 1.4 shuttles, d = 0.94.

In their discussion, the authors suggested that future research should increase training frequency to 3 times a week and/or increase time per interval (i.e., training volume). Because of the post-operative context, therapists preferred the first option. At the beginning of the fourth step of the rehabilitation process, children have not yet recovered their pre-operative level; specifically, some children still need crutches or k-walkers. Moreover, a further aspect not to be underestimated after surgery: fatigue and pain [140]. Patients are physically deconditioned after a sometimes long-time of cast immobilization. Therapists agreed that increasing training volume in this context was not suitable. In the ARRoW-CP protocol, the choice of intensity, volume, and time were based on practical experience from expert clinicians and from literature [49,141,142]. Every training session consisted of a prescribed intensity, volume, and time as described in Table 5.1.

Before the first session of each week, the target velocity was calculated during a Muscle Power Sprint Test (MPST). This test was made through the ARRoW-CP game and was presented as a "calibration" to the participant. The target velocity was defined as the highest velocity of 6 sprints (see Section 5.4.2 for more details about MPST). This test was repeated every week to adapt the game's difficulty to the child's progress.

CHAPTER 5. ARROW-CP: ACTIVE VIDEO GAME FOR GAIT TRAINING IN CHILDREN 104 WITH CEREBRAL PALSY

	WEEK 1	WEEK 2	WEEK 3	WEEK 4
Frequency (session/week)	3	3	3	3
Sprint Time (s)	30	30	30	30
Rest Time (min:s)	2:00	2:00	1:30	1:30
Sprint Repetition	4	6	8	10
Total Time (min)	10	15	18	22

Table 5.1 – Details of the OGT implemented in the serious game ARRoW-CP

The control group intervention for the RCT is detailed in Section 5.4. Briefly, the control group protocol consisted of 4 weeks of treadmill gait training (3 sessions per week), with a maximal duration of 30 minutes. This protocol was an adaptation of Grecco et al. [142]. They observed an average augmentation of 83% (range 80-85%) in 6MWT following 12-weeks of treadmill training in postoperative rehabilitation protocol. The distance travelled during the 6MWT increased significantly (before:  $166.4 \pm 39.1$  m; after:  $304.7 \pm 75.8$  m; p = .00), with an effect size: d = 2.29. The large effect size justified the choice of this protocol.

# 5.3 User-centered Game Development & Design

# 5.3.1 Game Development Framework

This work followed the process framework for AVG for motor rehabilitation therapy PROGame proposed by Amenguai Alcover et al (Figure 5.1) [143].

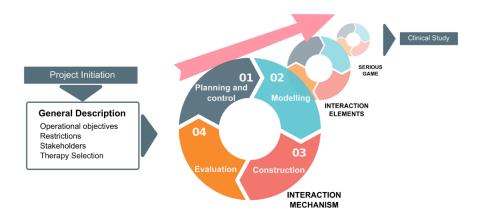


Figure 5.1 – Process flow for Active Video Game development. From Amenguai et al. [143]

The development team was composed of therapists (3 physiotherapists), researchers (2 in computer science, 1 in rehabilitation science, 1 in movement science), and a software engineer. From project initiation to the start of the clinical study, the process development framework occurred between January 2020 and June 2021.

The first step was the **project initiation**. The team identified the need for an active video game, the stakeholders, and user categories (users and experts). We also clarified the game functionality and constraints. We selected the therapy to transfer into the active video game (see section 5.2). The team used communication tools like an oral presentation of preliminary results,

open debates, surveys, and meeting reports. The aim was to support incremental development between team members and share knowledge. At this stage, the project proposed a general description:

- 1. The project's goal was to develop an active video game for walking rehabilitation of children with motor disabilities and particularly cerebral palsy.
- 2. The active video game should fulfill the following operational objectives: to be safe; to provide efficient gait training; to improve walking speed; to motivate the patient; to be fun.
- 3. The active video game must include the principles of motor learning, which are task-specific practice, variable practice, high practice intensity, progressive difficulty, augmented feedback, and adaptability to user abilities. Furthermore, the active video game should include motivational elements to increase engagement.
- 4. The team identified crucial technical aspects. First, children must be free to move in the global environment without the restriction of movement. Second, children must use their usual walking aids (crutches or posterior walkers).
- 5. The team's members agreed on making walking speed the main variable input of the serious game. Mean reason is that intensive gait training focused on walking speed has shown their clinical efficacy. The therapy to transfer into the active video game should include walking sprints.

The second stage of the development framework was the **interaction mechanism**. This stage was subdivided into four steps: Planning; Modelling; Construction, and Evaluation. The Microsoft Hololens AR headset was selected during this phase because its technical characteristics matched all identified specifications. Version 1 of the headset is used in this study (see Section 3.3). ARRoW-CP game was developed using Unity software (version 2019.4.8f1) and Microsoft Visual Studio 2019. The Mixed Reality ToolKit for Unity (MRTK version 4.2.3), a Microsoft-driven project that provides a set of components and features used to accelerate cross-platform MR app development in Unity, was used. We have developed a reliable algorithm, called HoloStep, for measuring the real-time gait parameters with an AR headset system (see Section 3.4 and 3.5). We demonstrated that 1. The accuracy of the AR headset's sensors was sufficiently high to evaluate the position of the user without time drift in the global environment [120]; 2. HoloStep was reliable for measuring and calculating walking speed, cadence, step length, and total distance travelled in comparison to a reference motion analysis tracking system (MOCAP) [121].

The third stage of the development framework was the **interactive elements**: What is the design of interactive features that force patients to perform the therapy correctly? This stage was also subdivided into four steps: Planning; Modelling; Construction, and Evaluation. During this phase, the team investigated actual commercial or serious games or active video games to get inspired. Team exploration and discussion conducted to Pokemon Go or Zelda, in which the gamer must explore the world and accomplish missions. These games appear very popular among the young generation. Then, to find the best feedback modalities for our active video game, we have conducted preliminary studies exploring the impact of feedback modalities on walking speed, both in healthy adults and in children with CP (see Chapter 4.3 and 4.4). The study on healthy adults showed that specific feedback helped increase walking speed, provided that the game instruction was explicit. A second study has occurred with children with CP. Results showed that a bodylocked hologram that advances in front of the user at the target speed was better to control the patient's walking speed. These studies allowed us to modify and adapt feedback modalities. The final increment was the **active video game design** to encourage the patient to perform the therapy regularly to reach a therapeutic effect. Experts and end users have unanimously validated the choice of the game universe (Samurai world).

### 5.3.2 User Test Campaign

The project team met regularly to discuss and make decisions throughout the construction process. When a full version of ARRoW CP has been considered testable, an extensive user test campaign has been organized in the rehabilitation center Ellen Poidatz to collect views and experiences from therapists (December 2020). A total of 23 participants were included (physiotherapist, 32%, occupational therapist 9%, speech-language pathologist 9%, neuropsychologist, doctor). The team retrieved valuable data from a Google Forms questionnaire, specifically created to gather stakeholders' views and observational analysis of the participants' live reactions and spontaneous behavior. The global score for ARRoW-CP experience was high (78.5 % rated the game at least 8/10). More than 60% felt the game stimulating. There was no adverse effect, like stress (more 90% evaluated their stress level under 2/5) or distraction (87% evaluated their concentration level above 4/5). The motivation was strong (95.7% above 4/5) and directly related to the feeling of having achieved the game's objectives. The sense of immersion is more divided ("I felt immersed into the game universe" was ranged from 1 to 5/5) (ISSUE 1). The overall understanding and consistency of the game mechanics seemed to be successful. However, some participants questioned the assessor about the therapeutic objectives during the game, which means that they were not understood by everyone (ISSUE 2). Real-time feedback helped the vast majority of participants to reach their objective (91.3%) noted above 4/5). However, diversification of feedback was requested without increasing the frequency (feedback on step length 54.4%, and other feedback modalities on walking speed 54.4%) (ISSUE 3). The informative feedback during rest helped the user. However, more prescriptive and visual feedback was requested to facilitate the comprehension of the "good movement" to perform (ISSUE 4). All the therapists wanted access to data summarizing the child's performance in previous sessions. The arguments put forward were to be able to follow the child's progress and to give him/her therapeutic advice in addition to the ones delivered in the application (ISSUE 5).

These results led us to improve the ARRoW-CP game. Thus, in order to solve or improve the FIVE ISSUES raised, we have proposed and developed FIVE ENHANCEMENTS (Table 5.2).

# CHAPTER 5. ARROW-CP: ACTIVE VIDEO GAME FOR GAIT TRAINING IN CHILDREN 107 WITH CEREBRAL PALSY

ISSUES	ENHANCEMENTS	
	Add new mixed reality content (particle, an-	
Default of Immersion	imation, long term reward system) and Add	
	narrative/story content	
	Add more instructions and information about	
Misunderstood Therapeutic Goals	The rapeutic Goals at the beginning of each	
	game session	
	Add negative/positive feedback in real time,	
More real time feedback on performance	add new audio content congrats/encouraging	
More real time reedback on performance	and Provide autonomous feedback on gait pa-	
	rameters (a customer choice system)	
More prescriptive and more visual feedback	Add interactive and visual feedback using Mo-	
during rest time	tion Capture System	
Summary of the child's results and perfor-	Add a therapist HUB displaying previous per-	
mance	formance of the child	

# Table 5.2 – Issues raised and enhancements developed after the User-Test Campaign

# 5.3.3 Communication Campaign

To facilitate the use of ARRoW-CP, we have adopted 3 strategies:

# 1. Information Booklet about Serious/Active video Game

For both family and therapists, we have shared in open access the information booklet "Jeux vidéo sérieux, késako ?" (in french https://www.fondationpoidatz.com/arrow-cp/). In this booklet, people have access to general information about serious games and active video games (definition, strength, recommendations, frequently asked questions) (Appendix 5.6).

# 2. User Guide & Tutorial in AR

We designed this tutorial for the therapist. First, we edited a PDF guide. Then, Léa Laffitte, who completed a one-month internship at the Fondation Ellen Poidatz in July 2021, developed an interactive application for the Hololens. This ARRoW-CP interactive tutorial included the general presentation of the ARRoW CP project (with the therapeutic objectives and the rehabilitation protocol implemented), the basic operations and interaction techniques of the Hololens, and a demonstration to perform the walking sprint.

# 3. Training session

For children, a preliminary session was scheduled for all of them before engaging in the gait training protocol with ARRoW-CP. It was a one-to-one interview between therapist and child. The aim was to explain Augmented Reality and ARRoW-CP. The child experimented with the Hololens with different games available from Windows App Store (RobotRaid, Young Counker, Origami), and she/he tested a demo of ARRoW-CP. He asked questions he wanted. For therapists, a training session was organized for all of them before proposing the gait training protocol with ARRoW-CP. It was a one-to-one interview between one executive team member and the therapist. The aim was to explain Augmented Reality and ARRoW-CP. The therapist played the ARRoW-CP Tutorial, received the user guide's PDF version, and asked questions he wanted.

# 5.3.4 Game Architecture & Content

ARRoW-CP immerses the player in a samurai world (Figure 5.2). She/He meets the village chief who can no longer protect its population. The natural elements are unleashed, causing famine and various damages. The player's role is to develop her/his energy through ninja training to build a protective totem for the village's inhabitants. The trailer of ARRoW-CP is available here https://youtu.be/BbmniiiJuaA.

All packages used to design the game were available in the Unity Asset Store: Polygon Samurai low poly 3D Art by Synty, Tiny Dragon by Suriyun, the GUI Kit - The Stone, and the Particle FX. Voice of game characters were recorded with volunteers in a professional radio studio attached to the Fondation Ellen Poidatz (HandiFM – France 107.3 FM).

At the beginning of the game, the therapist wears the AR headset. When this is a new patient, the therapist creates the player profile by giving her/his first name, age, height, and weight (*Profile Creation*). For the next sessions, the therapist only needs to load the existing user profile (*Profile Selection*). The therapist accesses the *Physio Hub*, which contains global information about the player profile, performance during the previous sessions, and also the current game level (i.e., number of sessions remaining). This control panel has been designed with the medical and rehabilitation team to match the information required to monitor the child. The therapist adjusts the AR headset on the child's head.

During the first game session (Week 1 - Session 1), the child is introduced to the ARRoW-CP game universe with narrative and presentation of the characters and the game objectives (*Game Presentation*). Then, after a vocal validation by the child, the calibration starts (*Calibration Presentation*). This calibration is required at each first session of a week. When the calibration is completed, the gait training protocol starts (*Game Training Presentation*). The number of sprints to be performed and the resting times differ from week to week as described in Section 5.2. When the gait training is completed, the child can choose if she/he wants to visualize his/her results by looking at the chest. Finally, Sensei Keito says goodbye to the child, encouraging him/her to continue the efforts between the sessions. The complete flow diagram is presented in Figure 5.3.



Figure 5.2 – **Image capture from the ARRoW CP game**. On the left, this is Yuki, the little dragon that children must follow during walking sprints and Sensei Keito who oversees Ninja training.

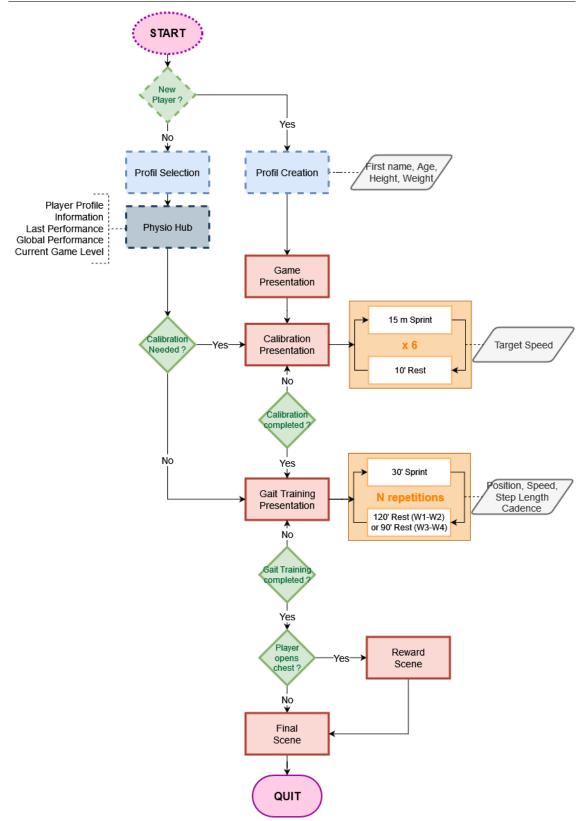


Figure 5.3 – Flow Diagram of the ARRoW-CP Active Video Game. Color code : Dotted line: AR headset is worn by the therapist ; Full line: AR headset is worn by the child ; Green box: decision ; Blue box: Air Tape Input is required ; Red box: Narrative Scene ; Yellow box: Gait training protocol - child performs sprints; Grey box: data recorded.

# 5.3.5 Feedback Characteristics in ARRoW-CP

The ARRoW-CP AVG combines gait training and motor learning ingredients: high intensity, difficulty progression, motivation, context-focused and goal-directed, task-specific practice, with augmented feedback. The choice of these feedbacks was based according to our previous experimental results (Article 5 in 4.3 & Article 6 in 4.4). These studies have shown that feedback helped to increase walking speed. In healthy adults, feedback combining a focus of attention with knowledge of results, a spatial representation with world-locked holograms, and a method of presentation with rich holographic content (animation, color changes) increased walking speed. In children with CP, feedback combining world-locked holograms that disappeared over time allowed a significant overshoot of the target speed. On the other hand, a body-locked hologram moving in front of the user at the target speed was better to control the patient's walking speed. After a debate between team development members, taking into account both efficacy of each feedback characteristic and their acceptability, the choice was made to develop the game around a "speed chase" scenario, using a body-locked hologram to follow. The main reasons were technical constraints (Microsoft Azure credits were required to build fixed and high-quality world-locked holograms) and experts' points of view (therapists judged that the world-locked holograms disappeared over time decreased the quality of walking). Several game test campaigns with children and professionals have been conducted to validate this final choice. This section will describe the feedback deployed in ARRoW-CP during the walking sprint, rest period, and at the end of each session.

#### 5.3.5.1 Feedback during walking sprint

During each walking sprint, Yuki the dragon, is flying in front of the player. His speed is the target speed extracted from the calibration scene. The patient has to follow Yuki. This feedback is called "Rocket Yuki". Moreover, from the 5<sup>th</sup> second, if the player exceeds the target speed for three consecutive seconds, a green particle highlights Yuki and a human voice congratulates the player. In the same way, if the player's speed is under the target speed, a red particle highlights Yuki and a human voice encourages the player. This feedback is called "Congrats/Encourage" (Figure 5.4). If we refer to our model of feedback in AR described in Section 4.2, we can categorize these two feedback mechanisms as illustrated in Figure 5.5.

#### 5.3.5.2 Feedback during rest period

At the end of each sprint, Yuki the dragon provides audio feedback (human voice) about the child's performance on three criteria: walking speed, step length, and head level. This feedback is called *"Evaluation System"*. Moreover, according to these results, an island appears to show a good way to walk faster, and/or with equal step length and/or with the back and knees well straight, the head straight. This animation was realized with Yona Rutkowski during his internship at the Fondation Ellen Poidatz using VICON Motion Capture analysis sytem. This feedback is called *"Advise System"* (Figure 5.6).

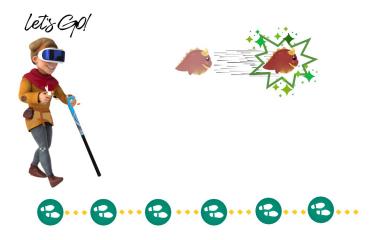


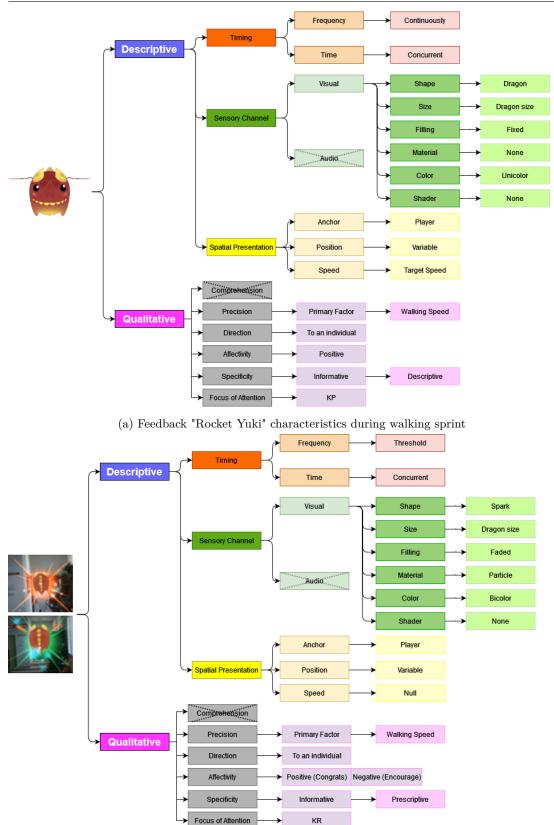
Figure 5.4 – **Feedback during walking sprint - Schematic representation.** "Rocket Yuki" is flying in front of the user at the target speed, and "Encourage" green color is displayed when the user's walking speed is correct.

# 5.3.5.3 Feedback at the end of the session

At the end of each session, the child can choose if she/he opens the chest to visualize her/his results in the form of a table with stars (feedback on demand). This feedback is called *"Score System"*. The main game quest to build a totem to protect the village continues, and the totem appears with more or fewer decorations according to the child's results. This feedback is called *"Totem"* (Figure 5.7).

To summarize, based on our model of feedback in AR for rehabilitation and our previous experimental studies but also through the multidisciplinary game development framework, we have developed these feedback for ARRoW-CP AVG :

Period	Name	Expected Effect	
	Rocket Yuki	Maintaining walking speed up	
Sprint	Congrats	Maintaining walking speed up	
	Encourage	Increasing walking speed	
Rest	Evaluation System	Correction of gait parameters	
nest	Advise System	Correction of gait parameters	
End of the Session	Score System	Improving or maintaining global performance	
End of the Session	Totem	Maintaining motivation an enjoyment	



(b) Feedback "Congrats/Encourage" characteristics during walking sprint

Figure 5.5 – **Feedback characteristics during walking sprint.** The feedback is categorized according our descriptive & qualitative model of feedback in AR.

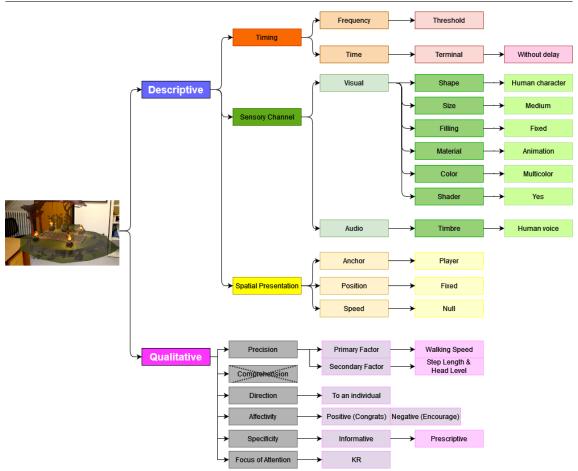


Figure 5.6 – Feedback "Advise System" appears after each sprint if the patient failed to achieve walking speed, and/or step length, and/or head level objectives. The feedback "Advise System" is categorized according our descriptive & qualitative model of feedback in AR.



Figure 5.7 – Feedback "Score System" and "Totem" at the end of each session. On the left, the reward panel appears if the patient opens the chest (feedback on demand) delivering stars. On the right, the totem construction is improving week to week.

# 5.4 ARRoW-CP: Effect of an Augmented Reality Active Video Game for Gait Training in Children with Cerebral Palsy following Single-Event Multilevel Surgery

# 5.4.1 Protocol for a Randomized Controlled Trial

The ARRoW-CP study is a randomized clinical controlled trial, currently in progress. First results are presented in Section 5.5. A total of 14 children and adolescents in the age of 12–18 years with CP will be included. The minimum time between surgery and inclusion is 7 weeks and participants have a Functional Mobility Scale 50 meters rating superior or equal to 2. Both groups follow a GT program of four weeks to improve their gait performance. The intervention group participate in the overground GT protocol with the ARRoW-CP game (See Section 5.2). The control group consist of GT on a treadmill with a maximal duration of 30 minutes. Measurements take place before the GT, directly after, and six months later. The primary objective is anaerobic performance. Secondary objectives are aerobic performance and enjoyment (See Section 5.4.2).

This paper is attached to this manuscript as submitted in BMJ Open in December 2021.

# Strengths of this study

- This is the first randomized-control trial to evaluate the impact of an active video game in AR on gait performance in children with cerebral palsy after surgery
- The active video game ARRoW-CP game has been specially designed for children with cerebral palsy using the PROGame design framework including a multidisciplinary team
- The addition of gait training to the conventional care may improve the aerobic and anaerobic gait performance in children with CP after surgery
- The enjoyment could decrease over time because of the relative limitation of game elements and game mechanics deployed in the ARRoW-CP game

# **OPEN ACCESS PROTOCOL**

# Effect of an Augmented Reality Active Video Game for Gait Training in Children with Cerebral Palsy following Single-Event Multilevel Surgery : Protocol for a Randomized Controlled Trial

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#### Abstract

**Introduction:** In paediatric rehabilitation, fun and motivation are also critical keys to successful therapy. A variety of interventions have shown positive effects, high level of interest, compliance and engagement with active video game (AVG). This seems to be an interesting approach for the postoperative gait rehabilitation of children with CP. In this study, we will investigate if an overground gait training (GT) delivered through an AVG can improve gait performance.

**Methods and analysis:** The ARRoW-CP study is a randomized clinical controlled trial. A total of 14 children and adolescents in the age of 12–18 years with CP will be included. The minimum time between surgery and inclusion will be 7 weeks. The test group will participate in the GT program with ARRoW-CP AVG, control group will receive GT on a treadmill. The primary objective is anaerobic performance. Secondary objectives are aerobic performance and enjoyment. This study has been registered in ClinicalTrials.gov (NCT04837105). The findings will be disseminated by publications in peer-reviewed journals and conferences.

**Keywords:** cerebral palsy; surgery; active video game; augmented reality; gait performance

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#### Introduction

Cerebral palsy is commonly defined as a "group of permanent disorders of the development of movement and posture, causing activity limitation" [1]. The overall prevalence of CP remains constant (2.11 per 1000 births) [2] with an estimated prevalence of 17 million people worldwide. [3] Individuals with CP present various clinical symptoms including a non-exhaustive list of neurological, orthopaedic, movement, cognitive, vision/hearing, aero digestive disorders.

Musculoskeletal disorders are considered as a secondary impairment contributing to restricted mobility in childhood and adulthood. [4, 5, 6] Since 1985, therapeutic interventions to correct orthopaedic disorders include single-event multilevel surgery (SEMLS). This surgery proposes, during one operative period, to realign the musculoskeletal system, practicing tendon transfer, muscle lengthening, derotation and/or deflexion osteotomy and joint stabilization. In a recent systematic review presenting state of the evidence on effective interventions for children with CP, SEMLS has been classified as effective ('probably do it W+') for improving both Gross Motor, walking speed and walking capacity but also contracture and alignment deformities [7]. To date, systematic reviews on the effect of SEMLS reported improvement in passive range of motion, kinematics and kinematics gait parameters, overall gait index and energy efficiency [8, 9]. Results were more disputed about the long-term effect on temporospatial gait parameters, gross motor function and the activity and participation domain [10].

A recent review has proposed a model of 5-step that could guide clinicians during the postoperative rehabilitation [11]. The authors suggested that the fourth phase, which included more intensive exercises, functional gait training and resistive muscle strengthening should be optimized to improve the gross motor function and walking speed after surgery. Functional gait training has been defined as 'actively practice the

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task of walking, to improve walking ability' [12]. Intervention could be overground gait training (OGT), or treadmill gait training (TT), with or without body support.

Previously, Grecco et al. demonstrated the efficacy of treadmill gait training program including both on functional mobility and gross motor function on children with CP after SEMLS [13]. Recently, a systematic review showed that gait training was a safe and effective intervention to improve walking capacity in children with CP, outside postoperative context [12]. In particular, the minimal clinically important difference (MCID) for increase in walking speed (0.1 m/s)was achieved after intervention in 12 studies/14 (studies level between II and III). Authors discussed two points: OGT could provide greater effect on locomotor abilities than TT because OGT is more representative of the natural walking [], and the addition of feedback could enhance the patient outcomes []. These points were important to consider after SEMLS, because the overall gait pattern of children was modified by the bone and muscle gestures. Novak et al. highlighted the importance of the context focused therapy and goal-directed training for children with CP [7]. Functional gait training should therefore take into account those recommendations and involve motor learning strategies: task-specific, variable practice, high intensity, augmented feedback during therapy sessions and motivation of the patient [14].

In recent years, new technologies have been introduced in rehabilitation practice, both for upper and lower limbs therapy. These systems include a large type of technology ranging from fully immersive virtual reality (VR) or augmented reality (AR) using commercially available head-mounted displays (HMD) (e.g., Oculus Quest; HTC VIVE, Microsoft HoloLens...), Cave Automatic Virtual Environment where video is projected on the walls and floor20 to video game console on television screen (e.g., Nintendo Switch, PlayStation...).

To 'actively practice the task of walking', systems combining treadmill training and exergame delivered through a screen in a semi-immersive environment have been tested with good results [15]. However, motor learning principles are not always fully integrated into VR/AR systems because of the lack of knowledge about which feedback modality and which intensity level should be provided in the rehabilitation settings [16].

To our knowledge, even if OGT was recommended for functional gait training, no AR system with serious game exists to provide high-intensity, with progressive difficulty, and variable modalities including feedback. To this end, we have developed the serious game ARRoW-CP combining OGT based on previous results [12] and literature [17] and motor learning theory [14]. In this serious game, continuous feedback as well as terminal feedback, both with different audio and visual modalities, are combined. Details of ARRoW-CP exergame, architecture, framework development and feedback characteristics, are available in the Additional File 1.

The current study, denoted as the ARRoW-CP study, will investigate whether a gait training protocol through a serious game in AR can increase the physical fitness levels of children with CP following surgery. Our hypothesis is that the serious game ARRoW-CP is at least as effective as treadmill training and more enjoyable for children.

#### Methods and analysis

#### Study Design

This study is a randomized control trial with two groups: OGT using the serious game ARRoW-CP in AR (OGT-AR) and Treadmill Training control group (TT). All children and adolescents participate in a four-week gait training intervention to improve their walking function in one of this two groups. During this period, children continue their usual physical therapy program (5 weekly 45-minutes session). This postoperative protocol has been standardized following a 5-step framework [11]. These usual rehabilitation sessions include muscle stretching exercises, muscle strengthening exercises (active resistance exercises), functional exercises (sit-to-stand, transfer, balance, walk, stairs).

#### Description of the two gait training interventions

To standardize the session content as much as possible, the therapists involved in the study participate in the training sessions before the start of the study. During these sessions, a member of the project team presents them the process and objectives of this study, as well as the gait training protocol proposed for the two groups. A session is dedicated to the familiarization with Microsoft Hololens and the ARRoW-CP game via a tutorial application.

#### ARRoW-CP: Overground Gait Training in AR

Intervention group receives the OGT-AR protocol through the serious game ARRoW-CP using the Microsoft Hololens headset (Figure 1). ARRoW-CP sessions are monitored by physiotherapist, assistant physiotherapist, or research assistant.

The intervention consists of 4 weeks of OGT (3 sessions per week), including a series of walking sprints. This protocol is an adaptation from Zwinkels et al. [17]. Every training session consists of a prescribed intensity, volume and time (Table I). Before the first session of each week, the target velocity is calculated during a Muscle Power Sprint Test (MPST) [18]. This test is made through the ARRoW-CP game and is presented as a "calibration" to the participant. During this test, no feedback is presented to the player. The target velocity is defined as the highest velocity of 6 sprints (see Intervention – Outcomes section below for more details about MPST). This test is repeated every week to adapt the difficulty of the game to the child's progress.

#### Treadmill Gait Training

The control group protocol consists of 4 weeks of GT on a treadmill (3 sessions per week), with a maximal duration of 30 minutes. This protocol is an adaptation from Greeco et al.[13]

Before the first session, the target velocity is estimated during a treadmill speed test: participants are instructed to walk on a treadmill with increasing speed (initially 0.5 km/h and increased 0.5 km/h each minute). Each minute, children are asked about shortness of breath and the subjective responses are classified using the 1-10 Borg Rating of Perceived Exertion Scale. The test is stopped if the score is higher than 5. The target velocity is defined as 80% of the maximum speed achieved during the test.

The first 5 minutes is a warm-up time, the speed is gradually increased until reaching the target velocity. The child walks for a maximal 20 minutes at their target velocity. Then, the treadmill speed is gradually diminished over the final 5 minutes. Training could be interrupted at any time at the child's request or physical therapist judgement. Treadmill sessions are monitored by physiotherapist, assistant-physiotherapist, or a member of the research staff.

#### Randomization Procedure

After baseline measures, eligible participants are randomized to the intervention or control group based on a computerized randomization program. Blocks randomization are calculated in block sizes of fours and six. The randomization procedure is only available to an independent researcher who will not be involved in the delivery of the interventions or the performance of the measurements.

#### Participants

All participants are recruited from the XXX Rehabilitation Centre. The children are operated in several hospitals in Paris: Necker Enfants Malades University Hospital, Trousseau University Hospital or Robert Debré University Hospital. Ethics approval is granted by the French Ethical Committee Sud-Est. Additionally, all parents, and participants from 12 years of age, should provide informed consent prior to study initiation. All participants should have a cooling off period prior to the inclusion (minimum 15 days between information and consent). Confidentiality and data access are guaranteed by the National Commission of Informatic (CNIL). A Data Protection Officer has been designated for all research studies conducted in this rehabilitation centre. He guarantees that the data protection and the rights of the subject are respected according to the General Data Protection Regulation (EU) 2016/679 (GDPR). The study has been registered in ClinicalTrials.gov (Identifier: NCT04837105).

Inclusion criteria are children with CP admitted for inpatient rehabilitation following SEMLS, 12–18 years of age, functioning preoperatively at GMFCS I–III. The minimum time between surgery and inclusion in the study is 7 weeks (step 4 of post-SEMLS rehabilitation process), they should have a Functional Mobility Scale 50 meters rating superior or equal to 2 (ability to walk on 50m using a walker or frame without help from another person).

All children should be able to cooperate, understand and follow simple instructions in French to practice the game. Only voluntary patients whose parents give their consent for their child's participation in the study are included. Criteria for non-inclusion include a diagnosis of photosensitive epilepsy in the medical record and/or patient's case history mentioning seizures that occurred while playing a video game, visual cognitive or auditory impairment that would interfere with playing the game. The patient should have normal or corrected vision and hearing.

#### Patient and Public Involvement

Patient involved as described above. No public involved.

#### Outcomes

Outcome measures take place at baseline (T0), immediately after four weeks of GT (T1), and six months later (T2). See Supplementary File 2 for outcomes details and criterion validity.

#### The 6 minutes walk test (6MWT)

The 6MWT is increasingly used in paediatrics, in clinics to monitor patients' abilities or in research as a criterion for evaluating the effectiveness of a rehabilitation protocol [19]. The 6MWT assesses distance walked over 6 minutes as a submaximal test of aerobic capacity/endurance. The reference guideline detailing the recommendations and instructions has been updated in 2013 [20, 21].

#### Muscle power sprint test (MPST)

The MPST evaluates anaerobic performance of youth with CP over a 6x15 meters at their maximal speed [18]. Velocity (m/s), acceleration (m/s2), force (kg/s2) and power (watts) are calculated. Anaerobic performance is defined as peak and mean power. Peak power (PP) is the highest power of 6 sprints and mean power (MP) is the average over 6 sprints

#### Shuttle Run Test (SRT)

The 10-meters shuttle run test is an adapted version of the 20-metre shuttle run test33 to accommodate children with cerebral palsy (CP) classified at Level I or Level II on the Gross Motor Function Classification System (GMFCS) [22]. In this study, the SRT-II will be used because of postoperative context. The SRT-II starts at 2 km/h. Speed is increased 0.25 km/h every level (minute). The test is over when the child cannot go to the next cone in time.

#### Question naire

To assess enjoyment for both control and test group, the 16-items of the Physical Activity Enjoyment Scale (PACES) will be used. The PACES is a valid and reliable measure of physical activity enjoyment.36,37 It has been used in many studies assessing the effectiveness of VR therapy [23]. This questionnaire will be presented to the participant at the end of the last session.

#### 0.1 Sample size and statistical analysis

According to a study of Grecco et al. an average augmentation of 83% (range 80-85%) in 6MWT was calculated after following 12-weeks of treadmill training in postoperative context [13]. The distance travelled during the 6MWT increases from before:  $166.4 \pm 39.1$  m to after:  $304.7 \pm 75.8$  m. The effect size was calculated: d= 2.29. It has been hypothesized that participants following the ARRoW CP protocol will show the same effect on the 6MWT.

With an alpha = 0.05 and beta = 0.20 (power = 0.80), a sample size of 6 subjects per group will be required. When taking a failure rate of 10% into account, 14 subjects should be included.

The required sample size was calculated with G Power 3.1.9.7. Parameter was t-tests – Means; difference between two independent means (two groups) with a priori analysis.

The effect of the GT protocol will be analysed using a multivariate repeated measures ANOVA. The possible differences between and within T0, T1 and T2 for the intervention group and control group will be calculated with a statistical significance level of p =0.05. If there is a significant difference, a post-hoc test will be executed to further investigate group differences. Quantitative descriptive statistics will be used to present patient characteristics and global results. All statistical analyses will be performed using R with a statistical significance level of p = 0.05.

#### Discussion

This approach has evolved from two directions: interest to improve walking capacity after SEMLS for children with CP, and from concern that the usual postoperative rehabilitation approach has not produced sustainable improvements in participation and activity in daily life for these children [8].

#### Serious game development framework

This work followed the serious game development framework PROGame, proposed by Amenguai Alcover et al. [24]. A participate process including both professional healthcare and patients has been conducted. The first step was the project initiation. The team identified the need for a serious game, the stakeholders, and user categories (users and experts). They also clarified the game functionality and constraints. They selected the therapy to transfer into the serious game. The operational objectives were to be safe; to provide efficient gait training; to improve walking speed; to motivate the patient; to be fun. These identifications were based upon prior experience and literature review [12, 25]. The team used communication tools like oral presentation of preliminary results, open debates, surveys and meeting reports. The aim was to support incremental development between team's members (that has their own specialty) and to share knowledge. At this stage, the project proposed a general description. The second step was the interaction mechanism, all technical solutions were explored and an algorithm for gait parameters detection was developed and tested [26, 27]. During the third step, the interactive elements, the team investigated actual commercial or serious games to get inspired. Team exploration and discussion conducted to games like Pokémon Go or Zelda, in which the gamer must explore the world and accomplish missions. These games appear very popular with the younger generation. The development team was composed of therapists (3 physiotherapists), researchers (2 in computer science, 1 in rehabilitation science, 1 in movement science) and a software engineer. These steps occurred between January 2020 and June 2021. All details are available in Additional File 3.

The aim was to think together about the best solution for improving postoperative results, especially walking capacity. New technologies were young people's preferred solution. These solutions seem to be a very promising tools for rehabilitation purposes, allowing to manipulate the environment, to offer interaction, to optimize feedback and many other potentialities [28, 29]

#### Feedback, a key point for motor learning

Feedback retraining paradigm is based on the conversion, the supplementation and augmentation of sensory information that are usually accessible only by an internal focus of attention, to accessible information [29, 30]. In this paradigm, augmented feedback is defined as augmented sensory information provided by an external resource (therapist or display) to the patient [31]. The information provided to the user could be relative to the movement's pattern or result on the environment or the outcome of a movement with respect to the goal. Sensory channels used to deliver information are visual, auditory, or haptic, linked to the proprioception properties of humans. The timing of feedback delivery is critical. Concurrent feedback is delivered while the skill is being performed, terminal feedback is delivered after the skill is performed with or without delay [32]. In most studies, even if feedback is effective to improve motor activities, the characteristics applied during interventions were generally inconsistent with motor control feedback theory. Authors suggest that timing, frequency and autonomy should be adjusted to optimize long-term effect [28]. A strategy that provides feedback to the user on demand promotes learning. Then, by reducing the frequency and timing of the feedback, the user can develop a sense of self-regulation.

#### Serious aspect of ARRoW-CP game

ARRoW-CP serious game combines many of the motor learning theories: context focused therapy and goaldirected training, task-specific, variable practice, high intensity, augmented feedback during therapy sessions and motivation of the patient [16]. To find the best feedback modalities for our serious game, we have conducted preliminary studies exploring the impact of feedback modalities on walking speed, both in healthy adults and in children with CP. Study on healthy adults showed that certain feedback helped to increase walking speed, provided that the game instruction was clear. Typically, feedback combining a focus of attention with knowledge of results, a spatial representation with world-locked holograms and a method of presentation with rich holographic content (like animation, colour changes) increased walking speed in healthy subject [33]. This step allowed us to modify and adapt feedback modalities. A second study has occurred with children with CP. Results showed that scenarios combining world-locked holograms that disappeared over the time helped children with CP to reach their target speed. On the other hand, a bodylocked hologram that advances in front of the user at the target speed was better able to control the walking speed of the patient[34].

ARRoW-CP is an adaptation of the validated protocol from Zwinkels et al. The original protocol consists of 8 weeks, twice a week [17]. Every training session consisted of a 30s walking sprints following the prescribed intensity, volume, and time: Week 1 to week 2: 8 sprints, work/rest ratio 1:4; Week 3 to week 4: 10 sprints, work/rest ratio 1:4; Week 5 to week 8: 12 sprints, work/rest ratio 1:3. Because of post-operative context, the intensity should be reduced. At the beginning of the fourth step of rehabilitation process, even if gait training is recommended, children did not recover to their pre-operative level, specifically some children needed crutches or k-walkers. Moreover, a further aspect not to be underestimated: fatigue and pain [35]. The choice of intensity, volume and time in the ARRoW CP protocol is based on practical experience from expert clinician and from literature [12, 17].

#### Conclusion

This article presents in detail the gait training protocol tested through a RCT. Both control group and experimental group have an evidence-based physical therapy training. This article also presents the game development framework of the ARRoW-CP serious game. This game is based on the most recent motor learning approach. This is the first study assessing the efficacy of postoperative gait rehabilitation using an active video game. If our hypothesis is validated, ARRoW-CP game will make possible to intensify gait training. This innovative strategy will have significant clinical impact by improving walking capacity for children after SEMLS. Publishing study protocol of the RCT offers the opportunity to collaborate with other teams and to give more details about the study. Results will be available in 2022.

# Figures



Figure 1 Image capture from ARRoW-CP active video game. On the left, this is Yuki, the little dragon that children must follow during walking sprints and Master Keito who oversees providing Ninja gait training. In the middle, a child wearing the Microsoft Hololens AR headset to see holograms. On the right, game elements encourage participants, increase motivation and improve adherence to the therapy (game score board).

# Tables

	Week 1	Week 2	Week 3	Week 4
Session Number	3	3	3	3
Sprint Time (s)	30	30	30	30
RestTime (min:s)	2:00	2:00	1:30	1:30
Sprint Number	4	6	8	10
Total Time (min)	10	15	18	22

Additional Files

Additional file 1 — ARRoW-CP Active Video Game

Additional file 2 — Main Outcomes details and criterion validity.

Additional file 3 — Active Video game development framework

Competing interests Not applicable.

# Author's contributions

ALG, GB, SO and ED conceived of the study, participated in its design and helped to draft the manuscript. SPT and MB participated in the coordination of the study. All authors read and approved the final manuscript.

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# 5.4.2 Outcomes to assess the efficacy of Gait Training Intervention

#### 5.4.2.1 6 Minutes Walk Test

The six-minute walk test (6MWT) is increasingly used in paediatrics, in clinics to monitor patients' walking capacity or in research as a criterion for evaluating the effectiveness of a rehabilitation protocol [144,145]. The 6MWT assesses distance walked over 6 minutes as a sub-maximal test of aerobic capacity/endurance. The reference guideline detailing the recommendations and instructions has been updated in 2013 [146,147]. Recently, we have proposed an adaptation of the 6MWT instructions for children with CP [148].

To summarize, patients are instructed to walk, not run, as far as they could along a 20-m level surface track during a 6-minute period. This shorter distance has been validated for children in order to be more focus on the task [145]. They could use their usual walking aids. After each minute, participants are told the elapsed time and standardized encouragement are provided. Patients are allowed to stop and rest during the test but are instructed to resume walking as soon as they feel able to do so. The stopwatch is not stopped during this time. The 6MWT distance (in meters) is registered. Measured 6MWT distance could be compared with normative values for children with CP [149]. It is also recommended to monitor heart rate during the 6MWT.

In population of children with CP, test/retest reliability is excellent for distance output (ICC = 0.98) [149]. The 6MWT is poorly related to VO2 peak in ambulatory adolescents and young adults with CP. The 6MWT is a more suitable measure of walking capacity than peak cardiopulmonary fitness in children with CP [150, 151]. The 6MWT outcome appears to be more strongly influenced by potential limits to walking speed rather than cardiopulmonary fitness.

#### 5.4.2.2 Muscle Power Sprint Test

The Muscle Power Sprint Test (MPST) evaluates anaerobic performance of youth with CP. The 15-m distance is marked by 2 lines taped to the floor. Cones are placed at the end of each lines. Participants are instructed to walk as fast as possible from one line to the other. Between each run, participants are allowed to rest for 10 seconds before turning around for the following sprint. Children are encouraged to give maximal effort. The following variables are calculated for each of the 6 sprints: velocity (m/s) = distance/time, acceleration (m/s<sup>2</sup>) = velocity/time, force  $(kg/s^2) = body mass \times acceleration, and power (watts) = force \times velocity.$ 

Anaerobic performance is defined as peak and mean power. Peak power (PP) is the highest power of 6 sprints and mean power (MP) is the average over 6 sprints [152]. The MPST is a valid test to assess the anaerobic performance in children with CP, significant correlations between the performance on these tests for both PP and MP were found. (PP: r = 0.731; MP: r = 0.903) [153]. Standard error of measurement, minimal detectable change and normative data are available in [152, 153]. Children with CP had impaired anaerobic performance [154, 155].

The MPST pre-post intervention was assessed outside ARRoW-CP game. Only the MPST which defined the target walking speed was calculated into the game.

### 5.4.2.3 Shuttle Run Test

The 10-metre shuttle run test (SRT) is an adapted version of the 20-metre shuttle run test [156] to accommodate children with cerebral palsy (CP) classified at Level I or Level II on the Gross Motor Function Classification System (GMFCS) [157]. Description of the test is the following [158] : "The course is 10 metres long; the end is marked with 2 cones and measuring tape. Subjects should wear regular sports clothing and shoes, and orthoses, if applicable. Each child should also wear a heart rate monitor. Children walk or run between the 2 markers at a set incremental speed. These runs are synchronised with a pre-recorded sound. As the test proceeds, the interval between each successive beep reduces, forcing the child to increase speed over the course of the test, until it is impossible to keep in sync with the recording." To facilitate the Shuttle Run Test passe for therapist, we have developed an Android application that beeps at pre-defined regular intervals, indicating the time spent (and the time remaining to reach the next marker) and allows the therapist to increment the number of shuttles made by the child (Figure 5.8) [159].

In this study, the SRT-II will be used because of the postoperative context. The SRT-II starts at 2 km/h. Speed is increased 0.25 km/h every level (minute). The SRT-II is a valid and reliable test. Test-retest is excellent (ICC = 0.99) and high correlations were found for the relationship between data for both shuttle run tests and data for the treadmill test (r = 0.96) [157].



Figure 5.8 – Shuttle-Run App for Android. This application facilitates the Shuttle Run Test passe for therapist.

### 5.4.2.4 PACES : Evaluation of the patient satisfaction and Enjoyment

This section is the fruit of a work conducted jointly with Paul Tirel. He did his research internship in rehabilitation at the Ellen Poidatz Foundation between January and March 2021 on the following subject: "Evaluation of satisfaction and enjoyment after a therapy using an active video game in augmented reality".

Satisfaction and enjoyment play a key role in the motivation of people engaged in rehabilitation, particularly among young people. ARRoW-CP required a questionnaire to assess both criteria. The method chosen to address this issue involved three steps. First, a systematic review to overview existing questionnaires was conducted in IEEE, PubMed and Google Scholar databases. Second, a Delphi process including multidisciplinary experts to select the best questionnaire for ARRoW-CP was organized. Third, a translation process was conducted to propose the selected questionnaire in French language. The inclusion criteria for the systematic review were:

- The questionnaire was used in a SG, AVG, exergame
- The questionnaire assessed satisfaction and/or enjoyment of users
- The questionnaire presented psychometric evaluation
- Article published after 2000, in English and/or French language

The research equation was: (Satisfaction OR Motivation OR Enjoyment) AND (Score OR Evaluation OR Questionnaire) AND (Serious Game OR Active Video Game OR Exergame). The questionnaires were classified according to their purpose, context of use and validation, and psychometric qualities (reliability, validity, sensitivity). Experts involved in the Delphi process discussed to select the more adequate questionnaire. From the 26 questionnaires extracted after the systematic review, 14 were judged inadequate regarding the study objectives, 2 could not be used without significant modifications, 6 did not have sufficient psychometric qualities. After three rounds of voting and discussion, the choice was made to use the PACES questionnaire, which initially aims to evaluate pleasure and satisfaction during physical activity [160]. As this questionnaire had several versions, it was decided to use the shortest one: the M-PACES (16 items). A rigorous translation process based on several guidelines [161–163] was conducted. A content and construct validity study of the PACES-French version was conducted including children with CP (Appendix 5.6). Today, the PACES-French version has been adopted as a secondary outcome in the ARRoW-CP study.

# 5.5 Preliminary results of the ARRoW-CP study

# 5.5.1 Patients Characteristics

On January, 7<sup>th</sup> 2022, we have included 11 patients. Five patients in the ARRoW-CP group, and four patients in the treadmill group. The characteristics of the patients are detailed in Table 5.3. Sex distribution between groups was not equal, 4 girls in treadmill group and 4 boys in ARRoW-CP group. Mean age between groups was equivalent. There were patient with GMFCS I-III, and they had soft tissue and bone surgery or only soft tissue.

In both groups, all participants successfully completed the 12 sessions. There was no missing data. No adverse effects such as difficulty in breathing, discomfort or cybersickness were observed during session. None of the participants fell while walking and wearing the AR headset or on the treadmill.

		ARRoW-CP Group	Treadmill Group
	N	5	4
Sex (girl:boy)		1:4	4:0
A	ge (mean $+/-$ sd)	16.2 + - 1.04	14.0 + - 1.0
	Hemiplegic Spastic	1	0
<b>T</b>	Diplegic Spastic	2	3
Туре	Triplegic Spastic	1	1
	Quadriplegic Spastic	1	0
GMFCS	Ι	3	2
Before Surgery	II	0	1
	III	2	1
SEMLS	Soft Tissue and Bone Surgery	5	2
SEMILS	Soft Tissue	0	1

Table 5.3 – Characteristics of the patients included.

# 5.5.2 Preliminary Results

We present statistical analysis of the data available for both the 6MWT and MPST. The 6MWT data is the distance performed during 6 minutes, the MPST data are the mean speed of the 6 sprints and the maximal speed of the best sprint (Details in 5.4.2).

First, a Student Test has been realized to test the comparability of subjects between groups BEFORE intervention. For the 6MWT, the initial difference between groups was equal to 236.60-239.67 = -3.07 m (in favour of treadmill group) with IC 95% = [-95.34 m; 89.20 m]. This difference was not statistically significant (t = -0.083, p-value = 0.94) meaning that the 2 groups were homogeneous. For the MPST mean, the initial difference between groups was equal to 0.97-0.77 = 0.20 (in favour of ARRoW-CP group) with IC 95% = [-0.37; 0.77]. This difference was not statistically significant (t = 1.0618, p-value = 0.3597). For the MPST max, the main difference between groups was equal to 1.05-0.81 = 0.23 (in favour of ARRoW-CP group) with IC 95% = [-0.48; 0.95]. This difference was not statistically significant (t = 1.0127, p-value = 0.3835). The two groups were comparable in 6MWT and MPST BEFORE intervention.

To compare the impact of ARRoW-CP intervention on the main outcomes, we have compared the difference AFTER-BEFORE between groups with a Student Test.

For the 6MWT (Figure 5.9), the difference between groups was equal to 46.67 (in favour of ARRoW-CP group) with IC 95% = [-50.79; 138.13]. There was no significant difference in the difference AFTER-BEFORE between groups (t = 1.15, p-value = 0.30). To interpret these results, we have calculated the mean difference AFTER-BEFORE and the confidence interval in each group. For ARROW-CP group, mean difference AFTER-BEFORE was 122.00 m, IC 95% = [27.87; 216.13]. This difference was statistically significant (t = 3.60, p-value = 0.023) meaning that the 6MWT in the ARRoW-CP has been improved. For Treadmill group, mean difference AFTER-BEFORE was 78.33 m, IC 95% = [6; 150]. This difference was statistically significant (t = 4.67, p-value = 0.043) meaning that the 6MWT in the treadmill group has been improved.

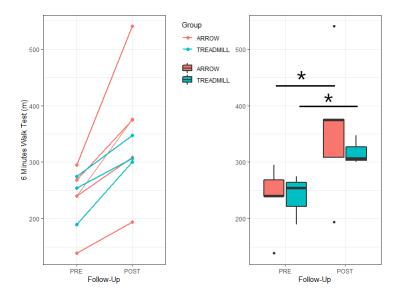


Figure 5.9 – **Results on the 6MWT.** All patients improve the 6MWT after the intervention. In both groups, significant difference in AFTER and BEFORE condition was observed.

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For the MPST mean (Figure 5.10), the difference between groups was equal to 0.087 (in favour of ARRoW-CP group) with IC 95% = [-0.19; 0.36]. There was no significant difference in the difference AFTER-BEFORE between groups (t = 1.16, p-value = 0.35). For ARROW-CP group, mean difference AFTER-BEFORE was 0.24 m/s, IC 95% = [0.16; 0.29]. This difference was statistically significant (t = 9.77, p-value = 0.00062) meaning that the MPST mean in the ARRoW-CP has been improved. For Treadmill group, mean difference AFTER-BEFORE was 0.14 m/s, IC 95% = [-0.17; 0.44]. This difference was not statistically significant (t = 1.92, p-value = 0.20) meaning that the MPST mean in the Treadmill group has not been improved.

For the MPST max (Figure 5.10), the difference between groups was equal to -0.046 (in favour of treadmill group) with IC 95% = [-0.78; 0.69]. There was no significant difference in the difference AFTER-BEFORE between groups (t = -0.26, p-value = 0.82). For ARROW-CP group, mean difference AFTER-BEFORE was 0.25 m/s, IC 95% = [0.17; 0.32]. This difference was statistically significant (t = 9.39, p-value = 0.00072) meaning that the MPST max in the ARRoW-CP has been improved. For Treadmill group, mean difference AFTER-BEFORE is 0.29 m/s, IC 95% = [-0.46; 1.05]. This difference was not statistically significant (t = 0.24) meaning that the MPST max in the Treadmill group has not been improved. These preliminary results must be considered with prudence because of the low number of patient.

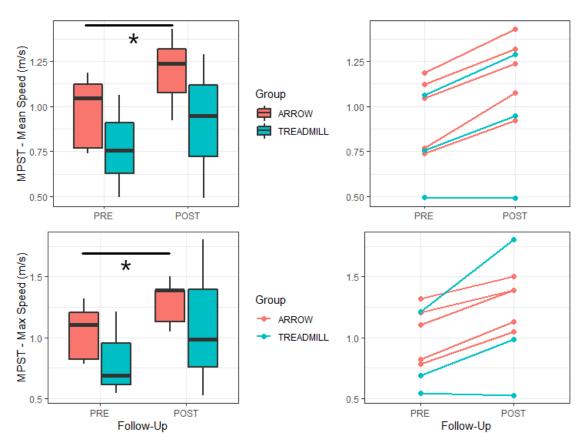


Figure 5.10 – Results on the MPST. Maximal speed and mean speed

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The enjoyment and satisfaction evaluated with the PACES questionnaire was in favour of ARRoW-CP (Figure 5.11). Items such as "I enjoy it", "It's very pleasant", "It's very exciting" were higher in ARRoW-CP group than in the treadmill group. Items such as "I feel bored", "I dislike it", "feel as though I would rather be doing something else" were higher in the treadmill group. Interestingly, item "It gives me a strong feeling of success" was very higher in the ARRoW-CP group. Moreover, there were homogeneous and good rated for "My body feels good" and "I get something out of it", meaning that both intervention seemed to be adapted for children with CP.

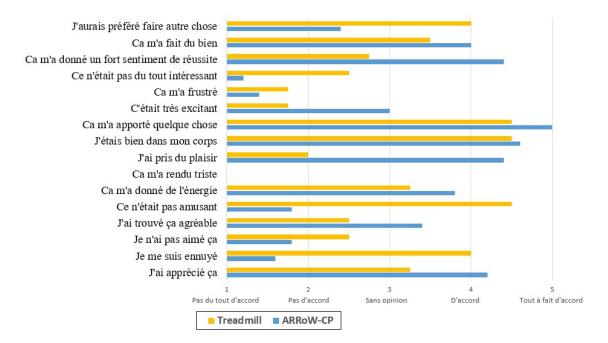


Figure 5.11 – Enjoyment assessment with the PACES.

To summarize, there were no statically difference between groups BEFORE and AFTER the intervention for 6MWT, MPST mean and MPST max. But we have observed a tendency in favour of ARRoW-CP group, patients in this group have improved all the main outcomes. In addition, the enjoyment and satisfaction are higher in the ARRoW-CP group. Even if more patients are required to confirm this trend, these results are very encouraging for the whole project.

# 5.5.3 Zoom on a patient : Results and Feedback

As we have seen previously, ARRoW-CP seems to impact the primary outcomes. Our research questions about feedback developed in Chapter 4 lead us to explore the impact of the feedback mechanisms deployed in ARRoW-CP described in Section 5.3.5.

We have chosen to present these results across a patient in this report. Milena \* is a 14 years-old girl (Patient 10). She has diagnosed with hemiplegia spastic CP, GMFCS I before surgery. She had a bone and soft tissues surgery in June 2021. She started the ARRoW-CP intervention in October 2021. Her results on the 6MWT (+ 106 m i.e + 0.30 m/s), MPST mean (+ 0.31 m/s) and MPST max (+ 0.31 m/s) AFTER the ARRoW-CP intervention were very positive. We recall that evolution of 0.1 m/s is considered clinically relevant.

<sup>\*.</sup> Her name has been changed

The full report of the ARRoW-CP intervention for Milena, as shared with the therapists and her parents, is presented in the Appendix 5.6. Milena completed the entire ARRoW-CP protocol, which equals 12 sessions and 84 walking sprints in 4-weeks. The PACES questionnaire revealed that she was very enthusiastic about this experience (Figure 5.12).

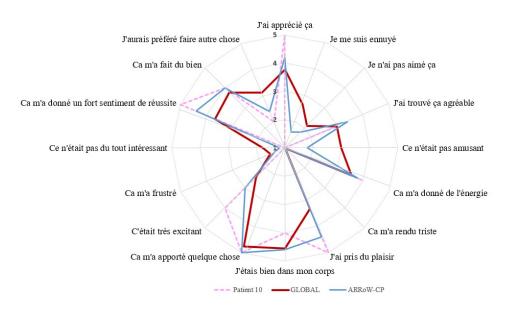


Figure 5.12 – **Enjoyment assessment with the PACES for Patient 10.** Comparison of the results of Patient 10 (pink dotted) and patients of ARRoW-CP group only (blue) and all patients of the study (red)

# 5.5.3.1 Feedback during walking sprint for Patient 10

During each walking sprint, Yuki the dragon, is flying in front of the player. The patient has to follow Yuki (Feedback "Rocket Yuki"). A green/red particle highlights Yuki and an human voice congratulates/encourages the player according to the success to reach the target speed (Feedback "Congrats/Encourage") (Section 5.3.5.1 for more details). Milena said she understood this feedback well and did not report any bugs during sprints. We observed his reactions/adaptations when the Yuki was flying and highlighted in green or red. Milena had three different behaviors: she accelerated, slowed down, and kept the same speed. We have enumerated the occurrences of the Yuki Feedback "Congrats/Encourage" and the patient walking speed over the 84 sprints (Table 5.4) :

	Acceleration	Slow down	Same Speed	Total
Green Yuki	33	43	237	313
Red Yuki	55	11	42	105
Total	87	54	279	420

Table 5.4 – Gait speed behaviors according Yuki Feedback "Congrats/Encourage" characteristics during Sprints for Patient 10. For the 84 sprints, the three different walking speed behaviors are counted.

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These results are promising, and they are very much in line with our expectations. We have designed the "Congrats" feedback (when the Yuki is highlighted in green to congratulate the patient - Green Yuki in Table 5.4) to help the patient **to maintain** their walking speed above the target speed. It is the case 76% of the time. We have designed the "Encourage" feedback (when the Yuki is highlighted in red to encourage the patient - Red Yuki in Table 5.4) to help the patient **to increase** her walking speed above the target speed. It is the case 52% of the time. But, we have to note that the feedback "Congrats/Encourage" did not always provide the same reactions.

For example, for the sprint represented in Figure 5.13, the patient did not reach her target speed between the  $2^{nd}$  and  $5^{th}$  second. So, Yuki highlighted in red. Then, the patient increased her WS immediately. Thanks to this acceleration, the second feedback was green. She continued to expand her WS (boosting effect). The third feedback was green. After that, she decreased and increased her WS but not significantly; WS stayed above the target speed. We hypothesized that the "Rocket Yuki" feedback has worked; when she saw Yuki pulling away, she decided to get closer to him. So, there was a mixed positive and combined effect with the spatial representation, the timing, and the visual characteristics of the feedback. Next, Yuki is highlighted in green. Her WS increased a little and decreased. Then, between the  $15^{th}$  and  $18^{th}$  second, her WS was under the target speed, so Yuki highlighted in red. The reaction of the patient was immediate; she accelerated. The effect of this last "boost" disappeared around the  $25^{th}$  second. We observed that the "Congrats" feedback had a boosting effect that faded with time. The hypotheses that we put forward at this study stage are of two kinds. First, the "Congrats" feedback is redundant, and it has been shown that motivation fades over time if the incentives are delivered repeatedly. And secondly, these incentives are not associated with "immediate" rewards. Indeed, congratulating people is good, but rewarding allows them to "materialize" their efforts.

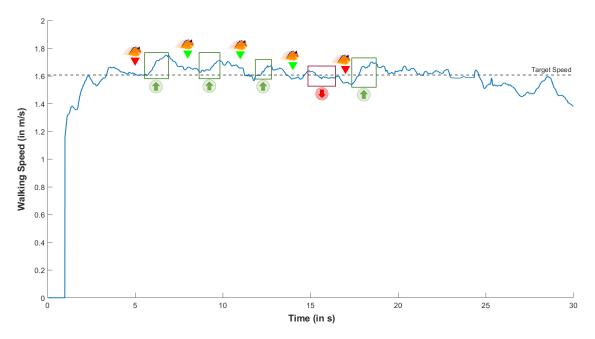


Figure 5.13 – Impact of Yuki Feedback during walking sprint for Patient 10. Participant walking speed during the Week 4 - session 2 - Sprint 6/10. Yuki highlights in red when speed in last 3 s < target speed, and in green when speed > target speed. Icon code : The "green arrow" represents the acceleration of the patient, the "red arrow" the slowing down of the patient.

#### 5.5.3.2 Feedback during rest period for Patient 10

During each rest time, between sprints, Yuki provides audio feedback (human voice) about the performance of the child on three criteria: walking speed, step length and head vertical position (Feedback "Evaluation System"). Then, when the patient failed to reach her/his objectives, a visual corrective feedback appears. The Ninja animated character shows the correct walking pattern (Feedback "Advise System") (Section 5.3.5.2 for more details).

For Walking Speed, Milena received the feedback "Advise System" during rest period 11 times on 84 sprints, including a number of times (8 times) in just the  $11^{th}$  session. Her case report forms revealed that she mentioned to be not motivated and very tired this day. However, this feedback seemed to be effective during the  $9^{th}$  session. After the first two sprints where she did not succeed for the WS goal, she achieved her goal for the next sprints (Figure 5.14).

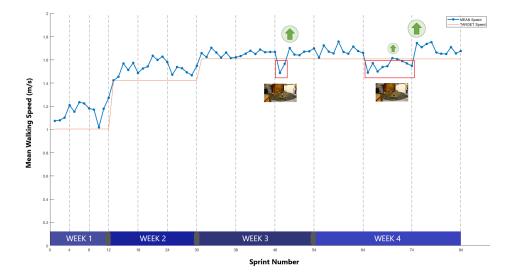


Figure 5.14 – Impact of the "Evaluation System" and "Advise System" Feedback during rest period on Walking Speed for Patient 10. The target speed calculated during the weekly calibration increases over the weeks. Patient 10 failed to reach her objective 11 times on 84 sprints. The feedback during rest period helped her to increase her WS 5 times. Vertical dotted line separate each session.

For **Head Vertical Position**, feedback "*Evaluation System*" and "*Advise System*" helped Milena to improve her head vertical position between Sprint 8 and 9 (Figure 5.15). But this feedback was only the reflect of head position, not body posture. We can not say that she walked upright with knee extended for example.

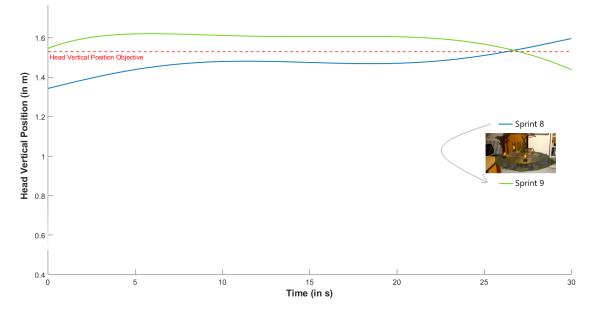
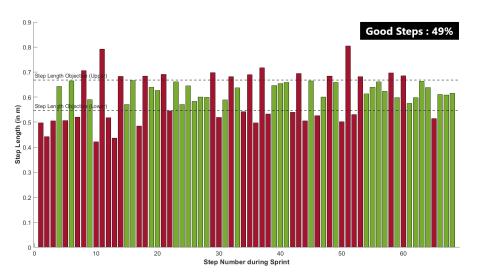


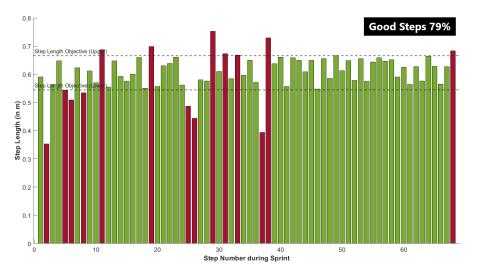
Figure 5.15 – Head vertical position during the Week 4 - session 3 - Sprints 8 and 9 for Patient 10. During the rest period, the *"Evaluation System"* and *"Advise System"* feedback are provided to the patient, resulting in higher head position between sprint 8 (in blue) and 9 (in green).

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For **Step Length**, she never reached the objective of more than 70% of "good steps". Hence, she always received the *"Evaluation System"* feedback indicating that their steps were not at the same length, and *"Advise System"* feedback showing the Ninja who was walking with symmetrical steps. This patient had asymmetric of step lengths. Her best score for this variable was 52.7% of "good steps", but on average, over all the 84 sprints, she had 15.1% of "good steps". Even if feedback did not improve her result, it allowed quantifying this information useful for the therapist. On the contrary, for another patient, the *"Advise System"* feedback permitted an improvement between sprints (Figure 5.16a and Figure 5.16b). We hypothesized that this patient could more easily adjust their step lengths.



(a) Step length during the Week 2 - Session 2 - Sprint 3 for Patient 5. The percentage of good steps for this sprint is 49 %.



(b) Participant Step length during the Week 2 - session 2 - Sprint 4 for Patient 5. The percentage of good steps for this sprint is 79 %. Patient has improved his performance.

Figure 5.16 – Impact on the Feedback during rest for Step Length. The "Evaluation System" and "Advise System" feedback are provided to the patient, resulting in higher percentage of good step length between sprints. In green, step length is within the objective interval (good step). In red, step length is outside the objective.

#### 5.5.3.3 Feedback at the end of each session for Patient 10

The "Score System" feedback provides stars according to the performance in the three parameters (WS, step length, and Head position). This feedback is delivered on-demand, the child can choose if he wants to see her/his score (Section 5.3.5.3 for more details). Milena wanted this feedback in every session. It isn't easy to analyze the impact of this feedback between sessions, but she reported that it helped evaluate her performance. For the "Totem" feedback, her opinion is mixed. She understood the symbolism of the village's protective totem, but she did not understand how to build a more powerful "Totem". A link should be made between the performance during the sprints and the construction of the "Totem" is not motivating enough (the children did not feel the interest of its construction), or the link between collecting the maximum of "Congrats" and the evolution of the "Totem" was not clear enough. Perhaps, the player needed another feedback during the sprints that made it clear that he was collecting points for his totem.

### 5.6 Conclusion

The objective of the ARRoW-CP AVG is to improve gait rehabilitation after SEMLS. It appeared very early that the choice of the rehabilitation protocol implemented in the AVG was essential. Indeed, we have highlighted in our state of the art that an efficient therapy must integrate the ingredients of motor learning, including task-specific and goal-directed therapy. Thus, we looked for gait training protocols that were ideally based on those principles but, even more importantly, proved their ability to improve patients' walking parameters. We also insisted that an AVG had to propose a playful approach of already validated rehabilitative practices.

Regarding existing gait rehabilitation protocols, we have chosen the High Intensive Training protocol proposed by Zwinkels et al. [48]. This intensive gait training benefited from multidisciplinary expertise to adapt the efficient dose, and it has shown its feasibility and effectiveness on a population of children with motor disabilities. Our control group benefited from treadmill gait training following the protocol of Grecco et al. [142]. This protocol has been validated in children with CP after SEMLS and has shown significant effects on walking ability. Both protocols were adapted in terms of duration to consider post-operative constraints. Our objective was that this protocol could be applied without altering the time of the rehabilitation center stay for children, a criterion considered essential to ensure the feasibility of the clinical study and the adoption by professionals in case of success of this intervention. Thus, the two protocols tested in the ARROW clinical study were conducted over four weeks, within a minimum of 7 weeks after surgery.

To develop ARRoW-CP AVG, we adopted the process framework for AVG for motor rehabilitation proposed by Amenguai Alcover et al. [143]. Through brainstorming sessions and test campaigns, the different steps of this framework allowed us to elaborate the ARRoW-CP game that we have described mainly in this chapter. The game's universe, architecture, and content were discussed in a multidisciplinary team, which is its strength.

As a follow-up to our work on feedback reported in chapter 4, we have developed the feedback delivered in ARRoW-CP at three moments of the game. During walking sprints, the main character Yuki trains the patient to follow him by moving at the target speed and delivering congratulation or encouragement based on his/her performance in real-time. During the rest

#### CHAPTER 5. ARROW-CP: ACTIVE VIDEO GAME FOR GAIT TRAINING IN CHILDREN 134 WITH CEREBRAL PALSY

periods between two sprints, the patient receives an evaluation and advice according to her/his previous performance on walking speed, step lengths, and head position. And finally at the end of each session, the patient receives stars based on the performance during the completed session on these three walking parameters. She/He also sees the construction of her/his protective village *Totem*, therefore materializing the progress in the narrative line of the game.

The ARRoW-CP clinical study protocol is fully described in a submitted paper. The primary and secondary outcomes are tests measuring aerobic and anaerobic walking capacity (6MWT, MPST) and a questionnaire assessing the person's motivation and satisfaction (PACES). The first results are very encouraging. They show a significant effect on improving all walking parameters, equally for the 6MWT in both groups and favor of the ARRoW-CP group for the MPST. Greater satisfaction is found in the ARRoW-CP group, with higher scores on items such as "I enjoy it" and "It gives me a strong feeling of success".

An analysis of the impact of the feedback was conducted on one patient. We have observed exciting effects that formulate hypotheses and new research questions. We have designed the "Congrats" feedback (when the Yuki is highlighted in green to congratulate the patient) to help the patient maintain their walking speed above the target speed. It is the case 76% of the time for this patient. However, we have to note that the feedback "Congrats/Encourage" did not always provide the same reactions. For example, the positive effect of the "Congrats" feedback delivered during the walking sprints seems to fade over time. The question about its redundancy and the link between the different feedback delivered in the ARRoW-CP AVG and thus maximize the impact of this therapeutic strategy. This clinical study is currently in progress. The inclusion of new patients will allow us to confirm these observed trends.

# Conclusion

This thesis investigates the feasibility and effectiveness of using an active video game in augmented reality to improve the gait rehabilitation of children with cerebral palsy after orthopedic surgery of the lower limbs.

This chapter concludes our work by answering our initial research questions:

- Is post-operative rehabilitation standardized ?
- $\square$  Do the spatiotemporal parameters of walking measured with an AR headset are reliable ?
- Show What are the model and characteristics of feedback in motor rehabilitation ? Are they compatible with AR ?
- S Can walking speed be controlled using virtual feedback delivered in AR ?
- Does an AVG in AR improve walking capacity in children with CP ? Does an AVG in AR improve children's motivation for gait rehabilitation ?

Finally, we present the limitations and perspectives of our research work.

### Is post-operative rehabilitation standardized ?

This first research question has led to new ones. We expanded it by asking exploring therapeutic strategies used for postoperative gait rehabilitation. Our first task was to examine the literature. The community unanimously agrees that postoperative rehabilitation is crucial to SEMLS success [62]. However, we quickly realized that no clinical study had evaluated the impact of rehabilitation after surgery. We, therefore, conducted a literature review to introduce a description and a qualitative analysis of the rehabilitation protocols described in the scientific literature [66] (Article 1). This work allowed us to highlight the common points, the differences, and the limits of each of them. Thus, our first contribution was to propose a common five-step framework for this specific rehabilitation. This framework can be used in future studies to standardize the protocol description in terms of objective, content, and intensity. From this literature review, two main axes are described as essential to maximize the fourth step, the intensification phase, from the seventh week to the sixth postoperative month: muscular strengthening and gait training [142, 164]. In this work, we are focused on gait training. The main reasons were the low speed in children with CP in comparison to typically developed children in preoperative context and the stagnation or even degradation of this walking speed after SEMLS [63].

Next step was to explore the means to improve gait rehabilitation protocols. Even if SEMLS was recommended in the systematic review of Novak et al. [43], the authors did not provide any details on the rehabilitation protocols following surgery. We could not duplicate any existing - and validated - postoperative protocol at this stage. The chosen approach was to investigate the fundamental ingredients of motor learning theory that should be integrated into motor rehabilitation to develop an efficient therapy [77]. We explored the recent literature on critical factors that would influence motor learning in the rehabilitation setting for people with motor disorders to answer this question. We notably included articles and references from the last international clinical practice guideline focusing on the interventions to improve physical function for children and young people with CP published in 2021 [79].

Based on these articles, we have synthesized the ten ingredients that are effective for motor recovery:

- High intensity practice, defined as a minimum training dose of 5 hours per week, can accelerate functional recovery [46,67,80]
- Difficulty progression, which consists in progressively increasing the difficulty of the task while adapting it to the learner, avoids boredom [81]. Moreover, it exists a strong association between challenge (appropriate difficulty level) and motivation [73]
- Motivation is a key factor for learning. A high level of motivation both increases activity capacities and children's participation, thus keeping adherence to treatment [82]. Main elements of motivation include appropriately challenging tasks, variable practice, setting realistic goals transferable to daily life, and aspects of competition such as a reward system [83]
- Task-specific, context focus and goal-directed therapy, in which tasks incorporated functionally meaningful movements that promoted activity and participation in their daily lives, maximizes the learning process and facilitated their generalization and transfer [84]
- Family support has been recently introduced in the conception of the rehabilitation program, strongly encouraged by the "F-Words": Fitness, Function, Family, Friends, Fun, Future. Family support has not been evaluated but the international clinical practice guideline recommended as an important supplement to clinicians-delivered intervention, especially when interventions were home-based [79]
- Finally, Feedback plays a crucial role to enhance motor learning and motivation level [75].

At this stage, and after brainstorming sessions, we have decided to develop an active video game (AVG) to improve gait rehabilitation. We hypothesize that gamification for rehabilitation using technologies, such as virtual or augmented reality systems, could be a suitable method for improving treatment efficiency, providing the user with goals, challenges, interactive and funny experience [100]. AVG is defined as video games used for sensorimotor (upper and lower limb use, posture, gait) and cognitive rehabilitation across the ICF's body function structure, activity, and participation domains. They are used to motivate, engage the patient in the therapy, to increase the treatment adherence, to improve well-being, and to promote activity [112].

To explore the impact of AVG for motor rehabilitation in Pediatric Neurology, we have conducted a depth overview of the literature. The majority of the studies reported the potential benefits of AVG, such as:

— increase intensity by repetition and consistent practice of the same task,

- individual difficulty progression,
- record and analysis of performance outcomes,
- provide a safe environment to undertake tasks which may be difficult/unsafe in real life,
- offer appealing games that may make therapy tasks more fun and engaging,
- increase compliance with therapy,
- enhance motivation which may lead to longer practice duration and more practice,
- movement repetitions,
- provide augmented feedback about task performance or task results,
- expand therapy at home including family.

However, the authors observed mixed results related to the efficacy of the AVG in improving the targeted skill [44,118]. Still, the majority reported some therapeutic gains, but the results were not always significant against the control group. We analyzed that the key components to promote motor learning were not always integrated into active video game interventions [73]. The potential benefits of AVG are significant, but motor learning ingredients should not be ignored in Motor Rehabilitation using AVG. The game interest, which raises the child's level of motivation for rehabilitation, is not sufficient to improve motor skills.

Before rushing towards the game development, we selected the best device. The involvement of the multidisciplinary team has led to a discussion on the technological device to be used. The team identified four crucial technical aspects. First, children must be free to move in the global environment, without the restriction of movement, to perform overground gait training [49]. Second, children must use their usual walking aids (crutches or posterior walkers). Thirdly, the device must be accurate enough to calculate spatiotemporal gait parameters (speed, cadence, step length). Fourth point, more technical constraint, this device should offer ample possibilities in software customization, providing technical documentation, SDKs, and dedicated development support. These four considerations impacted the choice of the technological device. To summarize, the device should be adaptable for overground gait training rehabilitation, hands-free, be compatible with exercises including variable intensity, and be well-documented. The Microsoft Hololens AR Headset was selected because its technical characteristics matched the specification.

# Do the spatiotemporal parameters of walking measured with an AR headset are reliable ?

To answer, we have investigated the feasibility of using an AR headset as a unique sensor and device for gait recognition in a real (overground) environment through two experimental studies.

The first one assessed the reliability of the headset tracking in comparison with a reference motion analysis tracking system during different walking conditions. Minimal errors between signals were 6 to 27 mm. Maximal errors were 55 to 250 mm and concerned the anteroposterior axis. Speed variation between the different sets did not influence the accuracy of the Hololens tracking. There was no signal drift over time. We have demonstrated that the accuracy of the Hololens is sufficiently high to evaluate the position and the velocity of the AR headset, and the user by extension, without time drift [120] (Article 2).

The next step of our work was to develop an algorithm to extract the spatiotemporal parameters from the AR headset. The objective was to develop and validate a new algorithm, HoloStep, calculating in real-time gait spatiotemporal parameters with the head's position of the user. The principle of detection used by HoloStep was based on the fact that during walking, the body displacement is pseudo-periodic. The body slightly leaned to the left/right side. Moreover, at each initial contact (when the foot was touching the ground), the body was lower on the vertical axis. Therefore, when the user was leaving to the left  $(x \min)$  and the body was at a lower position (y min), the user's left foot started to touch the ground. A similar situation happened on the other side (x max and y min) [165]. HoloStep was developed using a combination of locking distance, locking time, and peak amplitude detection with custom thresholds for children with CP. At each frame, HoloStep calculates: Time, Position, Filtered position, step detected (Boolean), Step length, and Walking distance from the beginning of the trial. In our second experimental study [121] (Article 3), we have shown that HoloStep is accurate enough, compared to the gold standard, to be used in clinical practice. In comparison to the control, [166], sensitivity, specificity, accuracy, and precision of the HoloStep algorithm were excellent. HoloStep can also provide feedback to patients on their spatiotemporal parameters through the AR headset. HoloStep algorithm is freely available, and it can be implemented in other active video games for motor rehabilitation. These two essential results encourage us to go further with the Hololens and use this device for our gait rehabilitation active video game.

# What are the model and characteristics of feedback in motor rehabilitation ? Are they compatible with Augmented Reality ?

Among the ingredients of motor learning, feedback is widely recognized as an efficient means to modify specific motor behaviors, make the learning process more explicit, enhance the practice environment, facilitate learning of complex tasks, and speed up the learning process. We conducted a literature review to propose a standard definition of feedback applied in (and applicable to) rehabilitation. We included publications related to the effectiveness of interventions in individuals with motor disabilities, including feedback. We proposed this synthetic definition of feedback that can be applied in the motor rehabilitation context: *"Feedback is based on the conversion, the supplementation, and augmentation of sensory information that are usually accessible only by an internal focus of attention, to access information. In this paradigm, augmented feedback is defined as augmented sensory information (visual, auditory, haptic, proprioceptive) provided by an external resource (therapist or device) to the patient".* 

The potential of augmented feedback has been demonstrated. Still, few studies have examined how different forms, presentations, timing, contents of augmentative feedback can impact motor rehabilitation [85]. In other words, define the "best" characteristics to apply to feedback to maximize motor learning.

Thus, as pointed out in the definition, augmented feedback characteristics are multiple. When using an AR headset, there are many ways to provide the augmented feedback. In most studies, even if feedback is effective in improving motor activities, the characteristics applied during interventions were generally inconsistent with motor control feedback theory [73]. The next step was developing a feedback model suitable for both motor rehabilitation and the augmented reality context. It is a practical extension of the theory of feedback to formalize their characteristics better. To create our model, we have extended and adapted the biofeedback model described by Macintosh et al. [75], and the qualitative model from Martinez et al. [122], to the AR context. Our model of AR feedback for motor rehabilitation has two complementary sides: descriptive and qualitative (Section 4.2). Our descriptive model is defined according to three essential notions: spatial presentation, sensory channel, and timing. Each idea includes several characteristics—for example, spatial anchor, position, and moving speed of the feedback for spatial representation. Our qualitative model has six categories defining the quality of feedback: precision, comprehension, direction, affectivity, specificity, and focus of attention. Thus, it would be possible to determine the impact of each feedback characteristic on motor rehabilitation by testing different combinations of our model. In a more practical sense, when creating a future AR application including feedback, a researcher, developer, or designer could refer to our model to configure their feedback.

# Can walking speed be controlled using virtual feedback delivered in AR ?

Since a comprehensive evaluation of our model of AR feedback for motor rehabilitation was not possible in the time available for this research, we have tested some of the characteristics of AR feedback in two studies.

In the first one, we investigated the impact of different forms of visual AR feedback on gait in typically developing adults. The primary research questions were: "Can AR feedback be used to help individuals achieve or exceed a target walking speed? Does AR feedback result in increased variability in walking speed?" we hypothesized that a game with AR feedback could help individuals to maintain or exceed a target walking speed without significantly affecting walking speed variability [167]. Our results demonstrated that gait parameters and user experience could vary depending on the type of AR feedback presented. Some feedback modalities increased walking speed, whereas others had a more considerable impact on speed variability. Specific recommendations based on learnings in this study include using knowledge of results feedback to create a more challenging task that motivates participants to excel, body-locked holograms that are easier to track, and clarify game presentation (Article 4).

In the second experiment, the objective was to define the most practical combination of visual feedback characteristics delivered in augmented reality to reach, maintain or exceed a target speed for children with cerebral palsy. We have demonstrated that the feedback characteristics have an impact on the results. A scenario combining a gradient color, with fixed spatial anchor, disappearing according to the time shown significant increase in walking speed [134] (Article 5 and Article 6).

The main result from these studies is that real-time visual feedback delivered through an AR headset is a feasible and acceptable intervention to provide immediate positive changes in walking speed for children with cerebral palsy. Moreover, the real-time feedback can give many advantages as a gait training intervention: it could be implemented in an active and challenging video game; it received a positive evaluation from the participants as it is engaging and easy to understand; and most importantly, it provides unique concurrent information on walking performance that would otherwise be very difficult or even impossible for clinicians to deliver.

In our previous study, including children with CP, we have confirmed that they can adapt their walking speed and positively respond to the real-time AR feedback. However, we have observed that not all patients performed equally well with the scenarios. When we looked at each participant's responses for each scenario, we observed some critical differences. Some people did not achieve better with the feedback; others were helped by a particular scenario but disturbed in another scenario. To define the individual response to AR feedback, we have conducted a posthoc analysis with data from the experiment detailed in Article 6. Our analysis showed three profiles of individuals: the "responders" who improved their performance with the feedback, the "nonresponders" who had an equivalent performance in the control situation and with the feedback and, the "disturbed" who presented a decrease in their performance with the feedback. Our results were the following literature. Liu et al. [136] have shown that particular stroke patients were more susceptible to the virtual environment and, therefore, responded differently to the feedback than others. Although this is an exploratory analysis, we have observed a low proportion of "disturbed" patients and a high proportion of "responders", which is very encouraging to develop the potential of AR systems and feedback for gait rehabilitation in children with CP.

# Does an AVG in AR improve walking capacity in children with CP? Does an AVG in AR improve children's motivation for gait rehabilitation ?

As mentioned previously, the objective of the ARRoW-CP AVG is to improve gait rehabilitation after SEMLS. It appeared very early that the choice of the rehabilitation protocol implemented in the AVG was essential. Indeed, we have highlighted in our state of the art that an efficient therapy must integrate the ingredients of motor learning, including task-specific and goal-directed therapy. Thus, we looked for gait training protocols that were ideally based on those principles but, even more importantly, proved their ability to improve patients' walking parameters. Regarding existing gait rehabilitation protocols, we have chosen the High Intensive Training protocol proposed by Zwinkels et al. [48]. This intensive gait training benefited from multidisciplinary expertise to adapt the efficient dose, and it has shown its feasibility and effectiveness on a population of children with motor disabilities.

To develop ARRoW-CP AVG, we adopted the process framework for AVG for motor rehabilitation proposed by Amenguai Alcover et al. [143]. Through brainstorming sessions and test campaigns, the different steps of this framework allowed us to elaborate on the ARRoW-CP AVG, including most ingredients of the motor learning principle. Based on our work about the characteristics of AR feedback, we have developed the feedback delivered in ARRoW-CP at three moments of the game. During walking sprints, the main character trains the patient to follow him by moving at the target speed, and delivering congratulation or encouragement based on his/her performance in real-time. During the rest periods between two sprints, where the patient receives an evaluation and advice according to her/his previous performance on walking speed, step lengths, and head position. And finally, at the end of each session, the patient receives stars based on the performance during the completed session on these three walking parameters. She/He also sees the construction of a protecting village Totem, therefore materializing the progress in the narrative line of the game (Section 5.3.5).

The ARRoW-CP study is a randomized clinical controlled trial (Article 7). A total of 14 children and adolescents between the age of 12–18 years with CP will be included. The minimum time between surgery and inclusion will be seven weeks, and participants will have a Functional Mobility Scale 50 meters rating superior or equal to 2. Both groups will follow a gait training program of four weeks to improve their walking capacity. Measurements will occur before the intervention, directly after, and six months later. The primary and secondary outcomes are tests measuring aerobic and anaerobic walking capacity (6MWT, MPST) and a questionnaire assessing the person's motivation and satisfaction (PACES).

In January,  $7^{th}$  2022, we have included nine patients. The first results are very encouraging. They show a significant effect on improving all the walking parameters, equally for the 6MWT in both groups and favor of the ARRoW-CP group for the MPST. Greater satisfaction is found in the ARRoW-CP group, with higher scores on items such as "I enjoy it" and "It gives me strong feeling of success". This clinical study is currently in progress. The inclusion of new patients will allow us to confirm these observed trends.

### Limitations

Our work has certain limitations, particularly concerning the technology chosen. Our first idea was to propose an active video game taking place inside the whole rehabilitation center compound, with, for example, points to be reached within a given time. We thought that this would have allowed the children to walk more between their rehabilitation sessions and thus favor acquiring their new walking pattern. Indeed, we observed after surgery, for many children, a low level of physical activity, which does not favor the development of their walking capacities. Our desire for unlimited exploration of the environment came up against the constraints of the Microsoft Hololens headset. Indeed, at the beginning of the project, the Hololens headset had some limitations. Thus, the physical persistence of the holograms in the real world between two game sessions was initially not stable enough to allow this development. Gradually, Microsoft has extended the use of Microsoft Azure points, which improves this temporal stability. We discovered this possibility too late in our game development process. Moreover, these options are not free, which will imply additional production and usage costs to be taken into account if the project evolves towards the implementation of these features.

On the other hand, the rapid evolution of technological tools proves to be as much an asset as a potential limitation. For example, the Microsoft Hololens version 2 was released in early 2021, making version 1 with which we started the project obsolete. Following some minor modifications, we have made our ARRoW-CP game compatible with the Hololens 2 (special attention will be needed to ensure compatibility with later versions of the Hololens headset). Beyond the technological aspects, software development is also an important issue. The exponential number of video games currently available on the market underlines that children and teenagers change games very regularly. Today, the Samurai and Ninja universe of ARRoW-CP has met great success among young people and their interest in Japanese culture. As fashions evolve, the continued attractiveness for future generations is questioned. However, in our context of post-operative rehabilitation, the ARRoW-CP game is intended to be used for a limited time, preventing early weariness. We will talk about the evolution of the content and the universe of the ARRoW-CP game from the perspectives.

We must address our research on feedback here the limitations of our theoretical model. This feedback model has been specifically designed and developed for augmented reality and motor rehabilitation. If other researchers wish to use this model in another context, this may require adaptations. On the other hand, we could not experiment with all the feedback modalities proposed in our model for the duration of this thesis.

Regarding the clinical study evaluating the impact of ARRoW-CP, we had many questions about the control group. We did not want to minimize the chances of improvement in walking capacity for the children included in this group. The ideal control group would have offered the same intensive training protocol as implemented in ARRoW-CP. However, the changing health situation made this option impossible. We experienced a staffing restriction, requiring rehabilitation assistants rather than therapists to manage the control group. These staffs are trained in treadmill rehabilitation but not high-intensity gait training sessions. We, therefore, chose a treadmill protocol for the control group that had been validated in the literature and then adapted it so that it could be performed over four weeks. Despite this, if the ARRoW-CP group achieves the same results in terms of walking capacity as those observed in the current control group, but with more satisfaction and adherence from the young people, this would be a very positive result.

Another point we wish to address in the limitations is using other spatiotemporal gait parameters and vertical head position, which are recorded in the game but moderately integrated. Indeed, step length and vertical head position are not used in ARRoW-CP as a game element during sprints; the child does not receive real-time feedback on these parameters. They are delivered as information at the end of the sprint and are linked to a terminal feedback allowing to earn stars at the end of the session. This choice was made to focus on the walking speed parameter and not overwhelm the child with several pieces of information that could be difficult to process in parallel during the sprint. Therefore, we chose to allow the therapist to access this information so that he could work specifically on step length and head vertical position outside the ARRoW-CP sessions according to her/his results. The walking cadence is transmitted neither to the child nor the therapist during the game. This choice was made following the test campaign, during which the therapists did not consider this information useful. However, we will come back to this in perspective, as new research questions this choice.

Furthermore, we chose to improve walking capacity through intensive gait training. However, other strategies to achieve this objective include muscle strengthening, proprioceptive work, or varied sports activities. However, our choice is based on a multidisciplinary reflexive process justified during our work. Finally, not all of the ingredients that we have highlighted on motor learning could be explored. In the following section, we will discuss ways to involve families more in the rehabilitation process.

### Perspectives

The first perspective is to complete the randomized controlled trial evaluating the impact of our active video game ARRoW-CP on improving the walking capacity of children with CP after surgery. Thus, as soon as all patients are included, we will measure its overall clinical efficacy compared to the control group.

Through the analysis of the data collected by the ARRoW-CP game, we will also investigate whether the children with the best performance in the game are the ones who improve the most on their primary outcomes. Similarly, continuing our focus on feedback, we will evaluate the effect of the terminal feedback (the *Score System* and the *Totem*) on the spatiotemporal gait parameters. We will also study the link between increased walking speed, step length, and head vertical position during sprints.

A comprehensive analysis of the PACES satisfaction questionnaire items will provide us with information on the appreciation of the game, which should lead to avenues for improvement for the ARRoW-CP game. Indeed, as noted in the limitations of our research, the rapid disinterest in the game world could have a long-term impact on young people's motivation and adherence to the therapy. Therefore, it is essential to update the content to keep up with the times regularly. We are not short of ideas. Thus, we would like to extend the application to other gait parameters, such as kinematic. For that, we would consider coupling the Hololens headset with inertial measurement units to measure the joint movements during the walk finely.

Another track of interest is to involve more families in the rehabilitation process and to develop a version of ARRoW-CP that can be played at home. This "mobile" version would encourage young people to continue their walking efforts during the weekends through a motivating mobile application. This application would incorporate the motor learning principles explored in ARRoW-CP and quantitative gait data to inform the child, family, and therapists of the youth's progress.

In pursuing our study on feedback, a new development avenue was born following a meeting with the NaturalPad team. We want to investigate audio feedback related to the walking cadence. Indeed, Dr Tallon's team (PhD) is experimenting with a mobile application synchronizing the steps and the rhythm of the music in the context of the gait rehabilitation of a patient with Parkinson's disease. Contacts have been made with this research team to develop this idea further.

All these reflections indirectly raise the question of the continuation of this project. Avenues are already being considered within the consortium that supported this thesis, the Ellen Poidatz Foundation and the University of Evry-Paris-Saclay. The support of the SATT of Paris-Saclay or the call for proposals such as PHRIP and ANR are preferred avenues. The will to pursue this project is substantial. If the clinical results are significant, the question of sharing and disseminating the ARRoW-CP active video game will be raised. First, we will need to find a way to ensure its adoption into the clinical routine in the Fondation Ellen Poidatz rehabilitation center. This will require communication of the results and new training sessions for the therapists. Finally, external communication of the results could allow its deployment in other pediatric rehabilitation centers. Testing ARRoW-CP with other populations in different therapeutic contexts would be an exciting prospect.

Finally, during our work, the question of the profiles of "responders", "non-responders" and "disturbed" patients to the feedback was of great interest to us. The desire to pursue this research question led us to contact Professor Lamontagne of McGill University in Canada. Her team is currently evaluating additional feedback in virtual reality in terms of presentation and sensory modality on the spatial and temporal asymmetry of walking in post-stroke patients.

This thesis results from a cross between several scientific fields sharing the ambition to be part of "tomorrow's medicine", which is participatory, predictive, preventive, customizable, and evidence-based. Thus, we have contributed to rehabilitation sciences by developing, in collaboration with stakeholders, an active video game for gait rehabilitation. We have investigated more fundamental notions in motor learning, notably on the impact of feedback. We have explored the field of information and communication sciences and technologies by developing tools and content usable in augmented reality. Our research over these three years and our many perspectives are a testament to the promising and exciting future of digital health to ensure the best care for our patients.

# Appendix

The GMFCS Instructions



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# GMFCS – E & R Gross Motor Function Classification System Expanded and Revised

GMFCS - E & R © Robert Palisano, Peter Rosenbaum, Doreen Bartlett, Michael Livingston, 2007 CanChild Centre for Childhood Disability Research, McMaster University

GMFCS © Robert Palisano, Peter Rosenbaum, Stephen Walter, Dianne Russell, Ellen Wood, Barbara Galuppi, 1997 CanChild Centre for Childhood Disability Research, McMaster University (Reference: Dev Med Child Neurol 1997;39:214-223)

## INTRODUCTION & USER INSTRUCTIONS

The Gross Motor Function Classification System (GMFCS) for cerebral palsy is based on self-initiated movement, with emphasis on sitting, transfers, and mobility. When defining a five-level classification system, our primary criterion has been that the distinctions between levels must be meaningful in daily life. Distinctions are based on functional limitations, the need for hand-held mobility devices (such as walkers, crutches, or canes) or wheeled mobility, and to a much lesser extent, quality of movement. The distinctions between Levels I and II are not as pronounced as the distinctions between the other levels, particularly for infants less than 2 years of age.

The expanded GMFCS (2007) includes an age band for youth 12 to 18 years of age and emphasizes the concepts inherent in the World Health Organization's International Classification of Functioning, Disability and Health (ICF). We encourage users to be aware of the impact that **environmental** and **personal** factors may have on what children and youth are observed or reported to do. The focus of the GMFCS is on determining which level best represents the **child's or youth's present abilities and limitations in gross motor function**. Emphasis is on usual **performance** in home, school, and community settings (i.e., what they do), rather than what they are known to be able to do at their best (capability). It is therefore important to classify current performance in gross motor function and not to include judgments about the quality of movement or prognosis for improvement.

The title for each level is the method of mobility that is most characteristic of performance after 6 years of age. The descriptions of functional abilities and limitations for each age band are broad and are not intended to describe all aspects of the function of individual children/youth. For example, an infant with hemiplegia who is unable to crawl on his or her hands and knees, but otherwise fits the description of Level I (i.e., can pull to stand and walk), would be classified in Level I. The scale is ordinal, with no intent that the distances between levels be considered equal or that children and youth with cerebral palsy are equally distributed across the five levels. A summary of the distinctions between each pair of levels is provided to assist in determining the level that most closely resembles a child's/youth's current gross motor function.

We recognize that the manifestations of gross motor function are dependent on age, especially during infancy and early childhood. For each level, separate descriptions are provided in several age bands. Children below age 2 should be considered at their corrected age if they were premature. The descriptions for the 6 to 12 year and 12 to18 year age bands reflect the potential impact of environment factors (e.g., distances in school and community) and personal factors (e.g., energy demands and social preferences) on methods of mobility.

An effort has been made to emphasize abilities rather than limitations. Thus, as a general principle, the gross motor function of children and youth who are able to perform the functions described in any particular level will probably be classified at or above that level of function; in contrast, the gross motor function of children and youth who cannot perform the functions of a particular level should be classified below that level of function.

## **OPERATIONAL DEFINITIONS**

**Body support walker** – A mobility device that supports the pelvis and trunk. The child/youth is physically positioned in the walker by another person.

Hand-held mobility device – Canes, crutches, and anterior and posterior walkers that do not support the trunk during walking.

**Physical assistance –** Another person manually assists the child/youth to move.

**Powered mobility** – The child/youth actively controls the joystick or electrical switch that enables independent mobility. The mobility base may be a wheelchair, scooter or other type of powered mobility device.

Self-propels manual wheelchair – The child/youth actively uses arms and hands or feet to propel the wheels and move.

**Transported** – A person manually pushes a mobility device (e.g., wheelchair, stroller, or pram) to move the child/youth from one place to another.

Walks – Unless otherwise specified indicates no physical assistance from another person or any use of a hand-held mobility device. An orthosis (i.e., brace or splint) may be worn.

Wheeled mobility – Refers to any type of device with wheels that enables movement (e.g., stroller, manual wheelchair, or powered wheelchair).

## **GENERAL HEADINGS FOR EACH LEVEL**

- LEVEL I Walks without Limitations
- LEVEL II Walks with Limitations
- LEVEL III Walks Using a Hand-Held Mobility Device
- LEVEL IV Self-Mobility with Limitations; May Use Powered Mobility
- LEVEL V Transported in a Manual Wheelchair

## DISTINCTIONS BETWEEN LEVELS

**Distinctions Between Levels I and II** - Compared with children and youth in Level I, children and youth in Level II have limitations walking long distances and balancing; may need a hand-held mobility device when first learning to walk; may use wheeled mobility when traveling long distances outdoors and in the community; require the use of a railing to walk up and down stairs; and are not as capable of running and jumping.

**Distinctions Between Levels II and III** - Children and youth in Level II are capable of walking without a hand-held mobility device after age 4 (although they may choose to use one at times). Children and youth in Level III need a hand-held mobility device to walk indoors and use wheeled mobility outdoors and in the community.

**Distinctions Between Levels III and IV** - Children and youth in Level III sit on their own or require at most limited external support to sit, are more independent in standing transfers, and walk with a hand-held mobility device. Children and youth in Level IV function in sitting (usually supported) but self-mobility is limited. Children and youth in Level IV are more likely to be transported in a manual wheelchair or use powered mobility.

**Distinctions Between Levels IV and V** - Children and youth in Level V have severe limitations in head and trunk control and require extensive assisted technology and physical assistance. Self-mobility is achieved only if the child/youth can learn how to operate a powered wheelchair.

# Gross Motor Function Classification System – Expanded and Revised (GMFCS – E & R) BEFORE 2<sup>ND</sup> BIRTHDAY

**LEVEL I:** Infants move in and out of sitting and floor sit with both hands free to manipulate objects. Infants crawl on hands and knees, pull to stand and take steps holding on to furniture. Infants walk between 18 months and 2 years of age without the need for any assistive mobility device.

**LEVEL II:** Infants maintain floor sitting but may need to use their hands for support to maintain balance. Infants creep on their stomach or crawl on hands and knees. Infants may pull to stand and take steps holding on to furniture.

**LEVEL III:** Infants maintain floor sitting when the low back is supported. Infants roll and creep forward on their stomachs.

**LEVEL IV:** Infants have head control but trunk support is required for floor sitting. Infants can roll to supine and may roll to prone.

**LEVEL V:** Physical impairments limit voluntary control of movement. Infants are unable to maintain antigravity head and trunk postures in prone and sitting. Infants require adult assistance to roll.

### BETWEEN 2<sup>ND</sup> AND 4<sup>TH</sup> BIRTHDAY

**LEVEL I:** Children floor sit with both hands free to manipulate objects. Movements in and out of floor sitting and standing are performed without adult assistance. Children walk as the preferred method of mobility without the need for any assistive mobility device.

**LEVEL II:** Children floor sit but may have difficulty with balance when both hands are free to manipulate objects. Movements in and out of sitting are performed without adult assistance. Children pull to stand on a stable surface. Children crawl on hands and knees with a reciprocal pattern, cruise holding onto furniture and walk using an assistive mobility device as preferred methods of mobility.

**LEVEL III:** Children maintain floor sitting often by "W-sitting" (sitting between flexed and internally rotated hips and knees) and may require adult assistance to assume sitting. Children creep on their stomach or crawl on hands and knees (often without reciprocal leg movements) as their primary methods of self-mobility. Children may pull to stand on a stable surface and cruise short distances. Children may walk short distances indoors using a hand-held mobility device (walker) and adult assistance for steering and turning.

**LEVEL IV:** Children floor sit when placed, but are unable to maintain alignment and balance without use of their hands for support. Children frequently require adaptive equipment for sitting and standing. Self-mobility for short distances (within a room) is achieved through rolling, creeping on stomach, or crawling on hands and knees without reciprocal leg movement.

**LEVEL V:** Physical impairments restrict voluntary control of movement and the ability to maintain antigravity head and trunk postures. All areas of motor function are limited. Functional limitations in sitting and standing are not fully compensated for through the use of adaptive equipment and assistive technology. At Level V, children have no means of independent movement and are transported. Some children achieve self-mobility using a powered wheelchair with extensive adaptations.

## BETWEEN 4<sup>TH</sup> AND 6<sup>TH</sup> BIRTHDAY

**LEVEL I:** Children get into and out of, and sit in, a chair without the need for hand support. Children move from the floor and from chair sitting to standing without the need for objects for support. Children walk indoors and outdoors, and climb stairs. Emerging ability to run and jump.

**LEVEL II:** Children sit in a chair with both hands free to manipulate objects. Children move from the floor to standing and from chair sitting to standing but often require a stable surface to push or pull up on with their arms. Children walk without the need for a hand-held mobility device indoors and for short distances on level surfaces outdoors. Children climb stairs holding onto a railing but are unable to run or jump.

**LEVEL III:** Children sit on a regular chair but may require pelvic or trunk support to maximize hand function. Children move in and out of chair sitting using a stable surface to push on or pull up with their arms. Children walk with a hand-held mobility device on level surfaces and climb stairs with assistance from an adult. Children frequently are transported when traveling for long distances or outdoors on uneven terrain.

**LEVEL IV:** Children sit on a chair but need adaptive seating for trunk control and to maximize hand function. Children move in and out of chair sitting with assistance from an adult or a stable surface to push or pull up on with their arms. Children may at best walk short distances with a walker and adult supervision but have difficulty turning and maintaining balance on uneven surfaces. Children are transported in the community. Children may achieve self-mobility using a powered wheelchair.

**LEVEL V:** Physical impairments restrict voluntary control of movement and the ability to maintain antigravity head and trunk postures. All areas of motor function are limited. Functional limitations in sitting and standing are not fully compensated for through the use of adaptive equipment and assistive technology. At Level V, children have no means of independent movement and are transported. Some children achieve self-mobility using a powered wheelchair with extensive adaptations. Palisano, Rosenbaum, Bartlett & Livingston, 2007 Page 3 of 4

### BETWEEN 6TH AND 12TH BIRTHDAY

Level I: Children walk at home, school, outdoors, and in the community. Children are able to walk up and down curbs without physical assistance and stairs without the use of a railing. Children perform gross motor skills such as running and jumping but speed, balance, and coordination are limited. Children may participate in physical activities and sports depending on personal choices and environmental factors.

Level II: Children walk in most settings. Children may experience difficulty walking long distances and balancing on uneven terrain, inclines, in crowded areas, confined spaces or when carrying objects. Children walk up and down stairs holding onto a railing or with physical assistance if there is no railing. Outdoors and in the community, children may walk with physical assistance, a hand-held mobility device, or use wheeled mobility when traveling long distances. Children have at best only minimal ability to perform gross motor skills such as running and jumping. Limitations in performance of gross motor skills may necessitate adaptations to enable participation in physical activities and sports.

Level III: Children walk using a hand-held mobility device in most indoor settings. When seated, children may require a seat belt for pelvic alignment and balance. Sit-to-stand and floor-to-stand transfers require physical assistance of a person or support surface. When traveling long distances, children use some form of wheeled mobility. Children may walk up and down stairs holding onto a railing with supervision or physical assistance. Limitations in walking may necessitate adaptations to enable participation in physical activities and sports including self-propelling a manual wheelchair or powered mobility.

Level IV: Children use methods of mobility that require physical assistance or powered mobility in most settings. Children require adaptive seating for trunk and pelvic control and physical assistance for most transfers. At home, children use floor mobility (roll, creep, or crawl), walk short distances with physical assistance, or use powered mobility. When positioned, children may use a body support walker at home or school. At school, outdoors, and in the community, children are transported in a manual wheelchair or use powered mobility. Limitations in mobility necessitate adaptations to enable participation in physical activities and sports, including physical assistance and/or powered mobility.

Level V: Children are transported in a manual wheelchair in all settings. Children are limited in their ability to maintain antigravity head and trunk postures and control arm and leg movements. Assistive technology is used to improve head alignment, seating, standing, and and/or mobility but limitations are not fully compensated by equipment. Transfers require complete physical assistance of an adult. At home, children may move short distances on the floor or may be carried by an adult. Children may achieve self-mobility using powered mobility with extensive adaptations for seating and control access. Limitations in mobility necessitate adaptations to enable participation in physical activities and sports including physical assistance and using powered mobility.

### BETWEEN 12<sup>TH</sup> AND 18<sup>TH</sup> BIRTHDAY

Level I: Youth walk at home, school, outdoors, and in the community. Youth are able to walk up and down curbs without physical assistance and stairs without the use of a railing. Youth perform gross motor skills such as running and jumping but speed, balance, and coordination are limited. Youth may participate in physical activities and sports depending on personal choices and environmental factors.

Level II: Youth walk in most settings. Environmental factors (such as uneven terrain, inclines, long distances, time demands, weather, and peer acceptability) and personal preference influence mobility choices. At school or work, youth may walk using a handheld mobility device for safety. Outdoors and in the community, youth may use wheeled mobility when traveling long distances. Youth walk up and down stairs holding a railing or with physical assistance if there is no railing. Limitations in performance of gross motor skills may necessitate adaptations to enable participation in physical activities and sports.

Level III: Youth are capable of walking using a hand-held mobility device. Compared to individuals in other levels, youth in Level III demonstrate more variability in methods of mobility depending on physical ability and environmental and personal factors. When seated, youth may require a seat belt for pelvic alignment and balance. Sit-to-stand and floor-to-stand transfers require physical assistance from a person or support surface. At school, youth may self-propel a manual wheelchair or use powered mobility. Outdoors and in the community, youth are transported in a wheelchair or use powered mobility. Youth may walk up and down stairs holding onto a railing with supervision or physical assistance. Limitations in walking may necessitate adaptations to enable participation in physical activities and sports including self-propelling a manual wheelchair or powered mobility.

Level IV: Youth use wheeled mobility in most settings. Youth require adaptive seating for pelvic and trunk control. Physical assistance from 1 or 2 persons is required for transfers. Youth may support weight with their legs to assist with standing transfers. Indoors, youth may walk short distances with physical assistance, use wheeled mobility, or, when positioned, use a body support walker. Youth are physically capable of operating a powered wheelchair. When a powered wheelchair is not feasible or available, youth are transported in a manual wheelchair. Limitations in mobility necessitate adaptations to enable participation in physical activities and sports, including physical assistance and/or powered mobility.

Level V: Youth are transported in a manual wheelchair in all settings. Youth are limited in their ability to maintain antigravity head and trunk postures and control arm and leg movements. Assistive technology is used to improve head alignment, seating, standing, and mobility but limitations are not fully compensated by equipment. Physical assistance from 1 or 2 persons or a mechanical lift is required for transfers. Youth may achieve self-mobility using powered mobility with extensive adaptations for seating and control access. Limitations in mobility necessitate adaptations to enable participation in physical activities and sports including physical assistance and using powered mobility.

Definition of feedback in the field of rehabilitation science

Author	Year	Word used for feedback	Qualifica tif	input	Action verb	output	characteristics	aim	Supplementary elements
Kassover M	1986	biofeedback	procedure	information about an aspect of body function	fed back	visual or auditory signal	a non-invasive technique	increase strength and improve motor control in patients	
Dursun E	2009	biofeedback	method of treatment	information about their normal and/or abnormal neuromuscular or automatic activity	to properly measure, process and feedback to individuals	auditory and/or visual feedback	uses electronic or electromechanical instruments		
Alahakone	2010	Biofeedback	a set of therapeutic procedures		to measure, to process and provide feedback ; such implementations (input) are augmented ; to provide responses to subjects on their movements	visual, auditory or tactile signals	uses electronic or electromechanical instruments ; incorporates a sensory device, a restitution device that can display the biofeedback information to the subjects, and a processing system that can perform computations, decision making, and control of the input/output devices in the system	to aid patients during their therapy sessions to augment and improve motor performance faster and prevent injuries	
		feedback	informatio n	patient's neuromuscular behavior, a corrective measure to be followed, or an indication of an improper		patient's neuromuscular behavior, a corrective measure to be followed, or an indication of an improper movement.			
Ronsse R	2010	augmented feedback		movement kinematics or kinetics	to adding extrinsic feedback	visual display of movement kinematics or kinetics to the task-intrinsic feedback that is normally available		to boost performance	
Sigrist R	2012	augmented feedback (= extrinsic feedback)	informatio n		it is provided	information that cannot be elaborated without an external source	provided by a trainer or a display	can relate the learner's individual performance to a desired performance or to an instruction.	teacher competency
Chiviacowsky	2014	feedback	informatio		information provided to learners	referring to their movement's pattern or result on the environment	after each trial or group of trials	one of the most important and studied factors affecting the learning of motor skills.	
		feedback	informatio	performance	received				
		intrinsic feedback	informatio	within a person because of movement	provided				
Gilmore PE	2014	extrinsic feedback	informatio n		be given		from an external source, such as theapist; during or at the end of task performance		
		Knowledge of results and knowledge of performance	type of extrinsic feedback		given	KP = the outcome of a movement with respect to the goal $KP$ = the movement patterns used for goal achievement.			
Hemayattalab	2014	feedback	sensory informatio	the actual state of a person's movement	indicates something	something			
R		intrinsic feedback	sensory informatio	natural consequences of producing a movement	arises	natural consequences of producing a movement			
Lee BH	2015	augmented feedback	informatio n	regarding their performance of functional activities	provides to patients	movement nature or quality, and it identifies both correct and incorrect components of functional activities			
Levin I	2017	augmented feedback ; Virtual environment- based feedback		gait abnormalities	augments the feedback; converts information that is usually accessible only by an internal focus of attention, to accessible information	such as attention to gait kinematios and asymmetry		learning a complex task	
Kearney E	2018	augmented feedback	extrinsio feedback	the outcome of movement and movement characteristics, termed knowledge of results and knowledge of performance, respectively	provided to a learner that supplements the information that is naturally available.	visual information		engages visual sensory ohannels and oan make the learning process more explicit	
		intrinsic feedback		natural perceptions within the individual					
		extrinsic feedback	informatio		offered to the individual		from external source		
Macintosh A	2018	biofeedback	extrinsic feedback ; informatio	motor performance	collected and communicated back to the individual		added to interventions	to enhance the practice environment.	timing, frequency
Oliveira N	2019	Biofeedback retraining paradigms		gait pattern deviations	augments native sensory and visual feedback.	real-time visual, auditory or haptic information	based on the patient's active interpretation and response to feedback cues of gait pattern deviations. They require sensing, processing and output (e.g. visual, haptio or auditory) technologies that tend to be less expensive than the hardware required to generate and control assistive		
Alwhaibi R.	2019	augmented feedback	informatio	performance	supplements sensory feedback		comes from a source external to the performer	to improve motor learning in the rehabilitation of persons with neurological disorders.	
Frikha M	2019	augmented feedback	informatio n	behaviors and performances	conveyed to athletes; provides	the extent to which their behaviors and performances correspond to expectations.	the effectiveness of this feedback depends on teacher competency, augmented feedback frequency, and timing.		
SYNTHESIS	2020	Augmented feedback	Sensory informatio n; (extrinsic feeback)	movement kinematics or kinetics motor performance gait pattern	sugniements and augments intrinsio feedback; converts information that is usually accessible only by an internal rocus of attention, to accessible information; grouvided' to the patient	visual, auditory or haptic information; referring to the movement's pattern or <i>result on the environment</i> or the outcome of a movement with respect to the goal	real-time or at the end of the task; information that cannot be elaborated without an external source (therapist or display); added to interventions	relies on the sensory integration of visual signals to modify specific motor behaviors; engages visual sensory channels and can make the learning process more exploit; to improve motor learning in the rehabilitation of persons with neurological disorders. to enhance the practice environment; learning a complex task; one of the most important and studied factors affecting the learning of motor skills.	the effectiveness of this feedback depends on teacher competency, display parameters, augmented feedback frequency, and timing.

# Article 5

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# Towards an AR game for walking rehabilitation: Preliminary study of the impact of augmented feedback modalities on walking speed

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#### ABSTRACT

Designing a serious game for walking rehabilitation requires compliance with the theory of motor learning. Motivation, repetition, variability and feedback are key elements in improving and relearning a new walking pattern. As a preamble to the development of an AR rehabilitation game, and in order to choose the most effective feedback to provide to the patient, this article presents a preliminary study on the impact of presentation modalities on walking speed. We investigate which visual concurrent feedback modalities allows to reach and maintain a target speed (maximum or intermediate). Our first results on children with motor disabilities (n=10) show that some modalities improved walking performance and helped patients to better control their walking speed. In particular, a combination of targets anchored in the real world with a time indication seems to be effective in maintaining maximum walking speed, while simple moving objects could be used to control speed.

**Keywords:** Cerebral palsy, walking rehabilitation, augmented reality, serious game, feedback

Index Terms: Human-centered computing—Human computer interaction (HCI)—HCI design and evaluation methods—Interaction techniques Interaction paradigms—Mixed / augmented reality— Applied computing—Life and medical sciences

#### **1** INTRODUCTION

Cerebral Palsy (CP) is the most common cause of childhood disability, affecting 17 million people worldwide [1]. Gait in children with CP is characterized by a slower walking speed, a shorterstride length, a lower cadence, and more time spent in double-limb support [2-4]. Studies clearly show a deterioration of gait temporal/stride parameters and kinematics over time when there is no therapeutic intervention [5]. Augmented Reality (AR) is a promising therapeutic means for gait rehabilitation games [6,7]. It allows the patient to walk in the real world, which includes real surfaces, obstacles, and people. It also potentiates therapeutic gains by increasing patient motivation and engagement in therapy and reducing boredom [8,9]. Finally, AR allows to provide real-time visual and auditory feedback to the patient, which is an essential aspect of motor rehabilitation. Motor relearning requires extrinsic (or "augmented") feedback to convert, supplement and enhance internal sensory information, providing knowledge of results (the outcome

of the movement, KR) and *knowledge of performance* (i.e. the kinematic quality of the movement, KP) [10–12]. This feedback can be delivered in a wide variety of presentations called "modalities".

Our research targets the creation of a helmet-based AR game for walking rehabilitation. The core feature is to detect gait parameters and provide augmented feedback to guide the children in performing exercises. This article focuses on two preliminary questions: which feedback modalities could allow to reach a maximum speed? and which feedback modalities could allow to better control the walking speed?

In the remainder of this article, we present the principles of walking rehabilitation, and the benefits of serious games and feedback. Then we explain the technical elements of our system, our feedback model adapted to AR and the current mini-games proposed in our application. Finally, we detail our user evaluation and discuss the first results.

#### 2 CURRENT STATE OF RESEARCH

#### 2.1 Principles of gait rehabilitation

In order to increase learning a new skill or to optimize motor recovery, current motor learning theories recommend task-specific practice, variable practice, high intensity of practice and the use of augmented feedback during therapy sessions [13-15]. In a metaanalysis of functional gait training in children with CP, Booth et al. reported a beneficial impact on walking speed, endurance, gross motor function and other gait-related outcomes, with persisting positive effects after cessation of intervention [16]. Regarding the improvement in walking speed, the largest effect size resulted from a long rehabilitation period (more than 40 hours spread over 10 weeks on average). Recently, other teams have tested the intensification of walking rehabilitation by increasing the intensity of the effort (maximal effort walking). The authors showed that increased intensity and repeated sessions reduce the duration of the protocol and produce good results in children with disabilities [17, 18]. Another key element to optimize motor learning is the motivation of the patient [19]. Recently, a systematic review by Novak et al. highlighted the importance of context-focused therapy and goal-directed training to improve the clinical picture of the patients with CP [20].

#### 2.2 Serious game for children with CP

Initially, games in rehabilitation for CP consisted of reusing commercial games and devices in a therapeutic context [21,22]. In their review, Lopes et al. [9] reported high levels of compliance, motivation, and engagement with game-based interventions, both at home and in the clinic. But regarding the effectiveness of the use of games, the results of the studies show both positive and negative results. One hypothesis with these mixed results is that the game was not specifically developed for therapeutic use and did not respect the principles of walking rehabilitation mentioned above. Serious games should be relevant, progressive, consistent with therapeutic goals and must deliver an appropriate practice intensity.

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#### 2.3 Feedback for rehabilitation

In rehabilitation context, the content of the feedback may vary depending on the aim of the therapy, e.g. the kinematics of movement or the kinetics of the gait motor performance. Augmented feedback provided to the patient could refer to the pattern of the movement, or the outcome of the movement with respect to a goal, or amplifies the environmental dynamics [23,24]. Feedback is used to modify specific motor behaviors, to make the learning process more explicit, to enhance the practice environment, to facilitate the learning of complex task or to speed up the learning process [11,25-27]. The effectiveness of this augmented feedback depends on the teacher's expertise, the display parameters, the frequency and timing of presentation [28]. When using an AR headset, there are many ways and modalities to provide the augmented feedback. In their meta-review of biofeedback interventions for individuals with cerebral palsy, MacIntosh et al. [29] list the feedback characteristics to consider: method of presentation, movement variable, focus of attention (KP or KR), timing, frequency and autonomy.

#### 3 TOWARDS AN AR GAME FOR WALKING REHABILITATION

Our objective is the design and development of the first helmet-based AR serious game for walking rehabilitation of children with CP, taking into account the latest advances in terms of motor learning, feedback and walking rehabilitation programs. Our work follows a serious game development framework proposed by Amenguai Alcover [30]. Our multidisciplinary team is composed by therapists (3 physiotherapists), researchers (2 in computer science, 1 in rehabilitation science, 1 in movement science) and a software engineer.

#### 3.1 System characteristics

We use the Microsoft Hololens as AR headset [31]. Our application is developed with Unity 2019.2.8f1 (64-bit) and Mixed Reality Toolkit (MRTK version 2). Hololens tracking is accurate enough to measure the position of the user without time drift [32]. Using the AR headset tracking data, we have developed an algorithm called HoloSD for measuring the real-time gait parameters of children with CP: walking speed, cadence, step length and global distance travelled. Our algorithm is reliable in comparison to a reference motion analysis tracking system (submitted paper).

#### 3.2 Feedback model

On the basis of the classification by MacIntosh et al. [29], we have established a model of feedback suitable for AR rehabilitation applications (Figure 1). A feedback is characterized by:

- Its timing of presentation, defined by:
  - the frequency: continuously, at regular intervals, by bandwidth or threshold, with fading, autonomously, by event;
  - the time: concurrent or terminal, with or without delay;
- Its spatial behavior: 3D position, moving speed, referential anchor;
- The sensory channel (visual, auditory or haptic) and characteristics of the presentation (shape, color, level, etc.)
- The eventual gaming aspects: feedback linked to a score, challenge, encouragement, or reward.

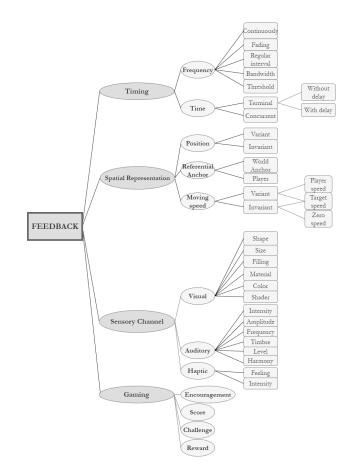


Figure 1: A model of feedback suitable for AR applications.

#### 3.3 Feedback modalities for walking speed

In a rehabilitation application, feedback will be used to communicate information to patients about their motor performance. In the case of walking, the therapeutic objective is to re-learn a walking pattern, and to improve the quality and quantity of walking. The final measured variables will be: speed, distance, step length and cadence. This study focuses on practicing walking speed.

We have designed 6 simple mini-games in order to study the impact of feedback on the patient's ability to reach and maintain maximum speed, as well as on his or her ability to respect a target speed. The task consists of walking 30m in a straight line at a given speed. The 6 mini-games and modalities of feedback that we designed are:

- Mini-game 1: A blue round shape moves at the target speed;
- *Mini-game 2*: A blue round shape moves at the target speed, color changes if the participant succeeds (green) or fails (red) to reach the target speed;
- Mini-game 3: A blue round shape moves at the target speed, color changes with a gradient (green-yellow-red) depending on the speed of the participant;
- *Mini-game 4*: A blue round shape stays in front of the participant (3m), color changes if the participant succeeds (green) or fails (red) to reach the target speed;
- *Mini-game 5*: A blue round shape stays at the other end of the hall, color changes if the participant succeeds (green) or fails (red) to reach the target speed;

• *Mini-game 6*: Five green round shapes are positioned every 5m, color changes in red if the participant fails to reach them on time. A time gauge around the shape decreases before the color change to red.

We deliberately used simple and identical round shapes for the evaluation. Feedback modalities differ mainly by their mechanism of color variation and their spatial anchoring, therefore their apparent moving speed. Figure 2 illustrates some of the holograms and feedback that the patient sees during the experimentation.



Figure 2: Application overview. The menu scene welcomes the patient. The aim of the study is given with vocal instructions. Colored spheres are four examples of feedback modalities on walking speed.

#### 4 USER STUDY

The two research questions are:

- Q1 Which feedback modalities allow to reach a maximum speed?
- Q2 Which feedback modalities allow to control the walking speed?

#### 4.1 Experimental procedure

The application starts with a welcome message showing the holographic environment and introducing the patient to the aim of the clinical study. Then, the calibration scenes consist of 2x15m walking at maximal speed and 1x30m walking at natural speed, with no feedback except startup vocal instruction. The calibration gives 2 values that will serve as targets for the patients:  $WS_{MAX}$  the maximal walking speed (the best of the 2 trials) and  $WS_{INTER}$  the intermediary walking speed with the equation (1).

$$WS_{\rm INTER} = WS_{\rm NATURAL} + \frac{WS_{\rm MAX} - WS_{\rm NATURAL}}{2}$$
(1)

Three mini-games are then played for warm-up and familiarization with the system and the holograms. When ready, patients start explicitly the next phase. Each mini-game consists of walking 30m in a straight line at a target speed ( $WS_{MAX}$  or  $WS_{INTER}$ ), with one of 6 feedback modalities or without feedback (control). Each child will therefore complete 14 mini-games, in random order, punctuated by rest periods.

#### 4.2 Participants and data collection

At this date, 10 naive participants participated in this ongoing study (age 14.3 +/- 1.7. 6 boys and 4 girls). They were all patient with cerebral palsy, undergoing rehabilitation at our center, who functioned at Gross Motor Function Classification System (GMFCS) I

 II [33], i.e youths walking in most settings environment with or without technical aids. Six children walked without technical aids, four walked using crutches.

Experimental sessions occurred in June and July 2020 in the Ellen Poidatz Foundation Rehabilitation Center. Each participant wore the AR headset and followed the instructions given by the application. At the end of the procedure, they completed a questionnaire on their user experience. Data (real-time position, walking speed, step length, cadence of the patient) were recorded with the AR headset and retrieved with the Windows Device Portal. The outcomes extracted for each trial were:

- When the target is the maximum speed (Q1): average speed, time spent above the target, time to reach the target.
- When the target is the intermediate speed (Q2): average speed, smallest difference between average speed and target speed, time spent around the target (+/- 1 standard deviation).

Legal tutors of each participant have given their written consent to collect and use their clinical data. This study respected the Ethic consideration from Helsinki's convention and received the agreement of our National Ethic committee.

#### 4.3 Results

All participants completed the entire game session. Table 1 shows how many patients reached their maximum and intermediate speed targets in each mini-game. Each mini-game resulted in more than 80% of patients having an average speed above their calibrated maximum speed. This score even reached 100% for mini-games 1 and 6.

Figure 3 presents for each mini-game based on  $WS_{MAX}$  the percentages of patients who achieved their best result in that mini-game. The best average speed is reached during mini-game 6 for 40% of patients and during mini-game 4 for 30%. Mini-game 4 also allows 30% of the patients to remain continuously above their maximum speed. Control and mini-games 2 and 3 do not support maximum average speed performance.

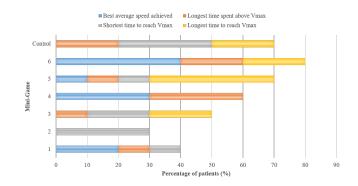


Figure 3: For each mini-game based on  $WS_{MAX}$ , percentages of patients who achieved their best result in that mini-game: best average speed, shorted time to reach target, longuest time above target.

Concerning the second objective of this study (to control the speed of patients), during the mini-game 3, 90% of patients walked with an average speed very close to their target speed (less than 1 SD). In mini-games 1 to 3, which aim is to follow the round shape, scores range from 60 to 90% (Table 1). Moreover, 70% of patients reach their maximal time spent around  $WS_{INTER}$  with these mini-games (Figure 4).

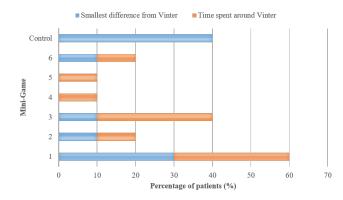


Figure 4: For each mini-game based on *WS*<sub>*INTER*</sub>, percentages of patients who achieved their best result in that mini-game: smallest difference with target, time spent around target.

Table 1: Percentage of patients achieving maximum and intermediate speed targets based on the mini-game.

Goal	Mini-game 1	Mini-game 2	Mini-game 3	Mini-game 4	Mini-game 5	Mini-game 6	Control
Target WS <sub>MAX</sub>	100	90	80	90	90	100	100
Target WS <sub>INTER</sub>	70	60	90	40	50	40	70

#### 4.4 Discussion

This clinical study on children with CP is currently in progress. Statistical tests have not been presented here due to lack of power but the qualitative results provide some initial insights.

All children reported a high level of satisfaction. They noted that the experience was a real challenge. The calibration scenes allow to adapt the difficulty of the mini-games to the patient's walking abilities, so the children found the application suitable, stimulating and that it would be a good therapeutic tool to improve their walking skills.

First results show that feedback modalities displayed in an AR game influence the real-time walking speed performance in children with motor disabilities. Concerning our first research question, the vast majority of patients achieved their maximum speed in the minigames. The worst score is 80% for mini-game 3, which could be due to the lack of comprehensibility of the modality (color gradient as a function of speed). A visually fixed modality in the user referential, therefore always visible, and with a simple behavior (binary color change) shows good results (mini-game 4). Mini-game 6 gives the best results for maintaining maximum speed: 100% of patients have their average speed above the maximum target speed, with 40% having their best average speed, and 20% having their longest time above the maximum target speed. Feedback modalities are richer than the other ones because they are combined: multiple stationary and world-fixed visual objects with colored information about the time remaining to catch them. The majority of children said that this was their favourite mini-game, because it motivated them to catch the round shape in time. It may suggest that providing intermediate targets is useful for maintaining attention.

Although mini-games 1, 2 and 3, with an object moving at the target speed in the world frame of reference and a colour change, are not the most effective for average speed, they allow children to reach their maximum speed target more quickly. There may be an extra motivation at the start of the exercise (catching up with the

target) to study further.

Concerning our second research question, the results are rather the opposite. Mini-games 1, 2 and 3 allow for better control of speed since taken together 5 out of 10 children reach their best score (i.e. they have an average speed closest to their intermediate goal). And with these modalities 7 out of 10 patients reach their best score for time spent around the goal. In terms of feedback modalities, these mini-games share the same characteristics: referential anchor is the world, position and size are variable and speed is equal to target speed. Only the color characteristic differs. Here the simplest seems the most effective: an object that moves without changing color.

Finally, having the final application in mind, children suggest improvements on the game aspect. More game objectives and the ability to customize the environment are currently being explored.

Recently, Borim et al. explored the impact of motor task and environmental constraints on gait patterns during treadmill walking in a fully immersive virtual environment. They reported that gait patterns were impacted by solving motor tasks, and by environmental constraints, in healthy young adults, suggesting increased need for balance control. But they concluded that virtual environmenttraining holds potential for improving gait and balance control. [34]

#### 5 CONCLUSION

We have presented a preliminary study for the development of an AR game for walking rehabilitation. On the basis of a sensory feedback model compatible with AR, we designed mini-games to practice walking in a straight line, assisted by a speed feedback. We are currently conducting a study with children with cerebral palsy to compare the impact of different feedback modalities on reaching and maintaining a maximum or intermediate speed. Initial results indicate that some modalities improved walking performance and helped patients to better control their walking speed. In particular, we will further investigate the influence of spatial anchoring and speed of simple visual representations on performance. Moreover, since rehabilitation must be practiced intensively and repeatedly, our next clinical study will also evaluate the motivational aspect of the modalities.

#### ACKNOWLEDGMENTS

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# Questionnaire PACES







Nom :				Prénom :		ID :
Groupe :	0	Test	/0	Témoin	Date de l'éva	luation ://20
Nom de l'éva	luate	ur :			Fonction de l'év	aluateur :

Ce questionnaire nous permet de savoir si tu as aimé l'activité que tu as pratiqué ces quatre dernières semaines. Cette activité était soit les séances sur *tapis roulant*, soit le *jeu sérieux ARRoW CP*.

Après chaque phrase, coche le petit rond avec l'option qui convient le mieux pour toi. Si tu as besoin d'aide ou que tu ne comprends pas bien une phrase n'hésite pas à demander à la personne qui t'accompagne.

1. J'ai apprécié ça	Pas du tout d'accord	Pas d'accord	Sans opinion	D'accord	Tout à fait d'accord
2. Je me suis ennuyé	Pas du tout d'accord	Pas d'accord	Sans opinion	D'accord	Tout à fait d'accord
3. Je n'ai pas aimé ça	Pas du tout d'accord	Pas d'accord	Sans opinion	D'accord	Tout à fait d'accord
4. J'ai trouvé ça agréable	Pas du tout d'accord	Pas d'accord	Sans opinion	D'accord	Tout à fait d'accord
5. Ce n'était pas amusant	Pas du tout d'accord	Pas d'accord	Sans opinion	D'accord	Tout à fait d'accord
6. Ça m'a donné de l'énergie	Pas du tout d'accord	Pas d'accord	Sans opinion	D'accord	Tout à fait d'accord
7. Ça m'a rendu triste	Pas du tout d'accord	Pas d'accord	Sans opinion	D'accord	Tout à fait d'accord
8. J'ai pris du plaisir	Pas du tout d'accord	Pas d'accord	Sans opinion	D'accord	Tout à fait d'accord
9. J'étais bien dans mon corps	Pas du tout d'accord	Pas d'accord	Sans opinion	D'accord	Tout à fait d'accord
10. Ça m'a apporté quelque chose	Pas du tout d'accord	Pas d'accord	Sans opinion	D'accord	Tout à fait d'accord
11. C'était très excitant	Pas du tout d'accord	Pas d'accord	Sans opinion	D'accord	Tout à fait d'accord
12. Ça m'a frustré	Pas du tout d'accord	Pas d'accord	Sans opinion	D'accord	Tout à fait d'accord
13. Ça n'était pas du tout intéressant	Pas du tout d'accord	Pas d'accord	Sans opinion	D'accord	Tout à fait d'accord
14. Ça m'a donné un fort sentiment de réussi	te Pas du tout d'accord	Pas d'accord	Sans opinion	D'accord	Tout à fait d'accord
15. Ça m'a fait du bien	Pas du tout d'accord	Pas d'accord	Sans opinion	D'accord	Tout à fait d'accord
16. J'aurais préféré faire autre chose	Pas du tout d'accord	Pas d'accord	Sans opinion	D'accord	Tout à fait d'accord

## Merci de ta participation

# Serious Video Game Booklet



MYODASHY

ARRoW-CP

NIRVANA

ALL CALL

Plateforme NIRVANA

- Les jeux sérieux engagent les jeunes à pratiquer **une** activité physique.<sup>13</sup>
- Les jeux sérieux améliorent la **vitesse de marche** ainsi que certaines **activités motrices fonctionnelles** (tels que les transferts)<sup>14,15</sup>
- Les jeux sérieux entraînent un haut niveau de satisfaction et d'adhésion aux séances de rééducation, accroissent la motivation des jeunes, augmentent la participation et l'engagement.<sup>16,17</sup>
- Les jeux sérieux favorisent les **apprentissages**, entre autres la rétention d'informations, la capacité à gérer le multitâche et la rapidité de réaction face un nouvel évènement.<sup>18</sup>
- Les jeux sérieux sont utilisés comme traitement ou palliatif dans de nombreux domaines de santé : gestion de la douleur, obésité, troubles du spectre autistique, rééducation post-AVC, pédiatrie, cancer...<sup>19</sup>



#### Foire Aux Questions

Pourquoi développer des jeux sérieux alors que de plus en plus de messages mettent en garde contre l'excès d'utilisation des écrans ? Les chercheurs sont unanimes pour affirmer que les jeux sérieux de rééducation ne sont pas « du temps d'écran en plus ». L'objectif est thérapeutique, il n'est pas occupationnel ou de divertissement. Les recherches actuelles n'ont montré qu'un faible lien entre le temps total passé devant les écrans et l'obésité<sup>20</sup> Par ailleurs, il convient de différencier la visualisation passive de vidéo projetée à la télévision, et l'engagement physique dans un jeu sérieux de rééducation. Les enfants sont en mouvement pendant toute la durée du jeu. Le risque d'obésité est donc écarté dans ce contexte. Enfin, les études révèlent une faible relation entre le temps total passé devant les écrans et les syndromes de dépressions, des troubles du comportement, de l'anxiété et du mal-être. Cela dépend du contenu regardé par l'enfant mais aussi du contexte d'utilisation et de l'accompagnement.<sup>20</sup> Dans l'utilisation des jeux sérieux de rééducation, les contenus sont adaptés à l'enfant et un thérapeute encadre en permanence la séance.

#### Les jeux sérieux peuvent-ils déclencher des crises d'épilepsie ?

Les personnes sensibles à la lumière ou aux variations brutales d'image peuvent subir une crise d'épilepsie avec ce type de dispositif, mais il existe souvent des facteurs associés au déclenchement de crises d'épilepsie comme la fatigue, le stress ou l'usage excessif. Par mesure de précaution, tous les enfants ayant déjà présenté une crise d'épilepsie liée à l'utilisation d'un écran ne pourront pas bénéficier de cette prise en charge.

#### La rééducation numérique va-t-elle remplacer les thérapeutes ?

Les jeux vidéo sérieux de rééducation sont des moyens supplémentaires mis à disposition des thérapeutes pour rééduquer les patients. Les thérapeutes planifient et accompagnent l'enfant lors des séances de manière à optimiser les effets positifs de la rééducation.

Comment bénéficier de cette prise en charge efficace et innovante ? Votre enfant se verra proposer par son thérapeute un programme de rééducation incluant un jeu sérieux en fonction de ses capacités et de l'objectif thérapeutique visé.

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# **ARRoW-CP** Case Report



# COMPTE-RENDU

# Protocole de rééducation de la marche









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# Protocole de rééducation ARRoW CP

## Présentation du protocole

Le but de l'étude clinique est d'évaluer un protocole de rééducation de la marche de 4 semaines réalisé avec une solution numérique ou un tapis roulant pour améliorer la prise en charge kinésithérapique post-opératoire. Votre patient a réalisé 12 séances de rééducation de la marche assistée par le jeu ARRoW-CP réparties sur 4 semaines. Chaque session comprenait **un nombre croissant de sprints de marche de 30 secondes chacun, avec un temps de repos décroissant** :



## Description du Hololens

Le Hololens est un casque de réalité augmentée permettant de projeter des hologrammes virtuels dans le monde réel. Pendant les séances de rééducation de la marche, votre enfant pouvait voir et entendre des retours sur ses performances, en temps réel ou sous forme de bilan de fin de session. Le but étant de faciliter et de rendre plus amusante la rééducation de la marche.



Enfant portant le casque Hololens



Capture d'image issue du jeu ARRoW CP

# Identification du participant

Nom			
Prénom			
Sexe F 🖂	M		
Date de naissance	2008		
Pathologie Paralysie cérébrale			

#### Données préopératoires

GMFCS I 🛛	II 🗆	III 🗆
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Aide technique de marche en intérieur Aucune

Aide technique de marche en extérieur Aucune

#### Opération

Date //06/2021

Libération des rotateurs internes. Ténotomie des adducteurs à droite. Allongement aponévrotique des IJ. Transfert du DF. Ostéotomie fémorale distale de dérotation ext. Aponévrotomie jumeaux et soléaire. Transfert Demi jambier ant sur cuboïde. Allongement aponévrotique du jambier post. Ostéotomie de fermeture externe du pied droit type Lichtblau. Ostéotomie de relèvement du premier métatarsien.

Prise en charge au CRF Ellen Poidatz			
Médecin référent			
Kinésithérapeute référent Mme			

Etude clinique
Numéro ID 🌑
Calendrier
Bilan pré-étude : 04/10/2021
Début des séances ARRoW CP : 04/10/2021
Fin des séances ARRoW CP : 28/10/2021
Bilan post-étude : 29/10/2021
Bilan follow-up (date prévisionnelle) : 27/04/2022

# Résultats globaux du patient lors de l'étude ARRoW CP

### Commentaire général

a suivi l'intégralité des séances du protocole ARRoW CP, ce qui équivaut à 12 sessions et 84 sprints de marche. Elle s'est montrée très enthousiaste. Certaines séances ont été réalisées en deçà de son intensité maximale alors que d'autres séances au contraire ont été plus dynamiques (auto-évaluation de l'intensité perçue en moyenne égale à 6/10).

## Evolution du bilan clinique pré et post-étude

	PRE-ETUDE	POST-ETUDE	EVOLUTION
Test de marche de 6	269 m	375 m	+106 m
minutes	0.747 m/s	1.042 m/s	+ 0.295 m/s
Muscle Power Sprint	0.818 m/s	1.131 m/s	+ 0.313 m/s
Test (max, mean)	0.771 m/s	1.076 m/s	+ 0.305 m/s

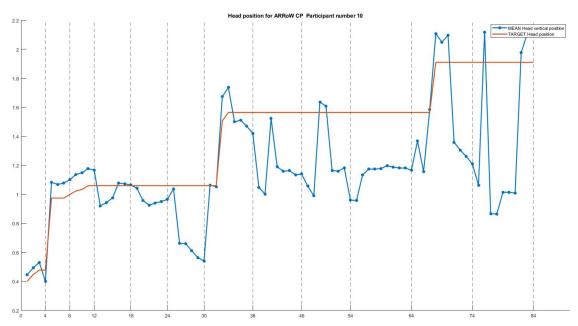
Pour information, un changement de +0.1 m/s est considéré comme cliniquement pertinent pour attester que l'enfant s'est amélioré.

# Résultats détaillés du patient lors de l'étude ARRoW CP

#### Hauteur de tête

La *hauteur de tête de référence* correspond à la meilleure hauteur de tête moyenne calculée au cours des précédents sprints. Ainsi pour un sprint donné, si la moyenne de hauteur de tête de l'enfant pendant la durée totale de son sprint était supérieure à 90% de cette valeur référence, le feedback terminal pendant le repos lui indiquait qu'il marchait en étant bien redressé.

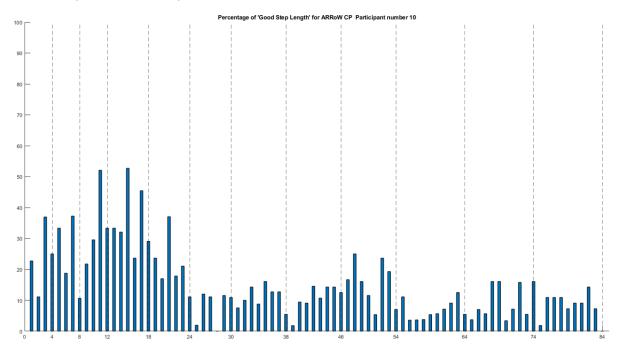
Pour son objectif de *hauteur de tête* (courbe orange) a augmenté régulièrement au cours des 4 semaines, ce qui signifie qu'elle a marché de plus en plus redressée au fil des semaines. Sa hauteur de tête moyenne est restée stable avec des fluctuations entre les différents essais (marqueurs bleus) et les différentes semaines (séparées par des pointillés). Ainsi, nous pouvons voir que les trois premières semaines, marqueurs bleur rempli ses objectifs.



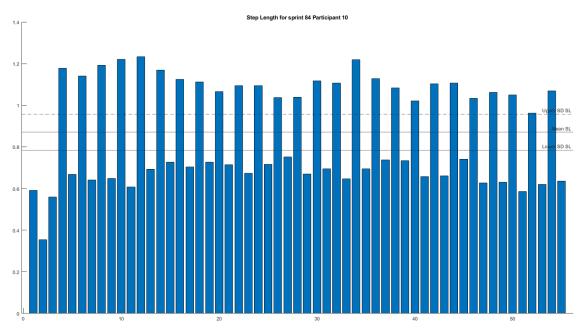
#### Longueur de pas

La longueur de pas moyenne au cours de chaque sprint a été calculée, ainsi que l'écart type. Un pas était considéré comme bon si sa valeur était comprise dans cet intervalle. Si le pourcentage de bon pas était supérieur à 70%, le feedback terminal pendant le repos lui indiquait qu'il avait une marche symétrique.

Pour elle n'a au cours des sprints jamais atteint l'objectif de + de 70% de « bons pas », ce qui signifie qu'elle présente des pas asymétriques. Son meilleur score est de 52.7% de bons pas, mais en moyenne il a 15.1% de bons pas sur l'ensemble des sprints. La fatigue liée au nombre de sprints réalisés ne semble pas avoir eu d'impact.



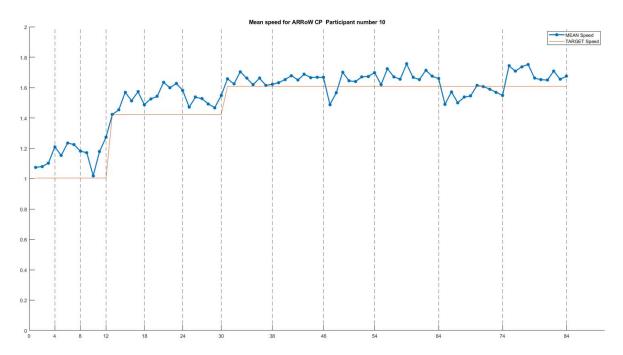
Si nous nous intéressons à la longueur des pas enregistrée au cours d'un sprint particulier, la figure suivante montre bien l'asymétrie de longueur des pas, un pas est systématiquement plus court que l'autre :



#### <u>Vitesse</u>

La vitesse de référence est la vitesse de calibration enregistrée à chaque début de semaine (meilleure vitesse moyenne lors du MPST). Si la vitesse moyenne du sprint était supérieure ou égale à cette vitesse, le feedback terminal pendant le repos lui indiquait qu'il avait atteint sa vitesse cible.

Pour elle a réalisé 86.9% des sprints au-dessus de sa vitesse cible, ce qui est une très bonne performance. Elle est restée très régulière sur sa vitesse moyenne par sprint. Ses progrès sont notables. Malgré l'augmentation du nombre de sprints à réaliser par séance, elle est parvenue à conserver sa vitesse de marche régulière et au-dessus de sa vitesse objectif.



Fait à Saint-Fargeau Ponthierry, le 29/10/2021

#### Signature

Anne-Laure Guinet, kinésithérapeute et doctorante



Pour toute question relative au jeu ARRoW CP, merci de contacter :

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Flashez ce QR code pour accéder à une vidéo du jeu

## Partenaires du projet ARRoW CP



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### ÉCOLE DOCTORALE



Sciences et technologies de l'information et de la communication (STIC)

Titre: Retours sensoriels multimodaux en réalité augmentée pour la rééducation de la marche chez des enfants atteints de paralysie cérébrale

Mots clés: Réalité Mixte, Rééducation de la marche, Apprentissage moteur, Retours sensoriels, Paralysie cérébrale, Chirurgie multisite

Résumé: La paralysie cérébrale est la pathologie neurologique pédiatrique la plus fréquente dans le monde entraînant notamment des troubles de la locomotion. La chirurgie multisite des membres inférieurs, qui réaligne le système musculo-squelettique en un seul temps opératoire, a prouvé son efficacité pour améliorer la qualité de la marche, en particulier les paramètres cinématiques et cinétiques. Cependant, les capacités de marche, comme le périmètre de marche, la vitesse de marche et la longueur de pas, demeurent trop souvent inchangées. L'analyse de la littérature scientifique a conduit à explorer l'hypothèse qu'une amélioration des protocoles de rééducation postopératoire conduirait à une amélioration de l'activité de marche. Pour celà, ce travail de thèse a permis le développement et l'évaluation clinique du premier jeu sérieux de rééducation de la marche en réalité augmentée adapté à l'enfant, et utilisable par un centre de rééducation en complément de la prise en charge traditionnelle. Ce jeu sérieux s'appuie sur les théories ex-

istantes de planification de l'entraînement de la marche et sur les modèles de feedbacks pour l'apprentissage moteur. Cette recherche est le fruit d'un croisement entre plusieurs disciplines scientifiques partageant l'ambition de s'inscrire dans la médecine de demain, qui se veut participative, prédictive, préventive, personnalisable, et basée sur les preuves. Ainsi, nous avons contribué aux sciences de la rééducation en développant en collaboration avec les parties prenantes un jeu sérieux de rééducation de la marche, nous avons investigué des notions plus fondamentales en apprentissage moteur, notamment sur l'importance des éléments de jeux et nous avons exploré le champ des sciences et technologies de l'information et de la communication par le développement d'outils et de contenus utilisables en réalité augmentée. L'utilisation d'un jeu sérieux de rééducation autonome devrait permettre d'améliorer les résultats postopératoires, notamment en augmentant la motivation et l'adhésion des jeunes à la rééducation.

**Title:** Multimodal sensory feedback in augmented reality for gait rehabilitation in children with cerebral palsy **Keywords:** Mixed reality, Gait rehabilitation, Motor learning, feedback, cerebral palsy, single-event multi-level surgery

Abstract: Cerebral palsy is the most common cause of childhoodonset, lifelong physical disability in most countries. Abnormal gross and fine motor functioning reflecting abnormal motor control are the core attributes of CP. These motor problems can lead to difficulties with walking. Single-event multi-level surgery of the lower limbs, which realigns the musculoskeletal system in a single operation, has proven to improve the gait quality, particularly the kinematic and kinetic parameters. However, walking ability, walking distance, and walking speed remain unchanged. After analyzing the scientific literature, we hypothesize that improving the postoperative rehabilitation protocols could improve activities and participation domains. For this purpose, this thesis allowed the development and the clinical evaluation of the first active video game for gait rehabilitation in augmented reality adapted to the child and usable by a rehabilitation center in addition to conventional care. This active video game is based on theories of the gait training program and feedback models for motor learning. This research comes from the smooth intersection of several scientific disciplines sharing the ambition to be part of tomorrow's healthcare, which is participatory, predictive, preventive, customizable, and evidencebased. Thus, we have contributed to motor rehabilitation sciences by developing an active video game for gait rehabilitation in collaboration with stakeholders. We have investigated more fundamental motor learning notions, notably the importance of game elements and feedback. We have explored the field of information and communication sciences and technologies by developing tools and content usable in augmented reality. The use of an active video game for gait rehabilitation should improve postoperative results, particularly by increasing the motivation and adherence of young people to therapy.

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