ANNEMIEKE REIMER

# Assessment of Fine Motor Skills in Children with Visual Impairment

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The work presented in this thesis was carried out at Bartiméus, Institute for the Visually Impaired, Zeist, the Netherlands. The research was done in close collaboration with the Scientific Institute for Quality of Healthcare (IQ healthcare). This institute is part of the Radboud Institute for Health Sciences (RIHS), one of the approved research institutes of the Radboud University Nijmegen Medical Center.

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|---|---|
| Promotor | Prof. dr. M.W.G. Nijhuis-van der Sanden |
| Copromotoren | Dr. F.N. Boonstra (Bartiméus, Zeist) |
| | Dr. R.F.A. Cox (RUG) |
| Manuscriptcommissie | Prof. dr. R.G.J. Meulenbroek |
| | Prof. dr. B.F. van der Meulen (RUG) |
| | Prof. dr. G.H.M.B. van Rens (VUmc) |

A visually impaired child
will always indicate,
if you ask what he sees,
he does not know what you see.
*(statement of many children with visual impairment)*

# Contents

# Chapter 1

# General introduction and outline of the thesis

Vision is one of the most important sensory channels for staying in contact with the world around us[1]. Vision provides information about our environment without the need for proximity, as is relevant in taste, touch and smell [2]. "Sensory experience from the external world can influence the way the brain wires itself up after birth"[2]. In infancy, visual experiences are not only crucial for normal development of the visual system[2], but in particular the interaction with the social and physical environment is crucial for the development of perception and perceptuo-motor control based on neural networks in the brain.

Vision provides important feedback to the vestibular, proprioceptive and sensory systems and consequently motor development is impeded in cases of early visual impairment (VI)[3]. Directly after birth, visual information is crucial for balance control during lifting and holding your head, sitting up, rolling, standing, walking and reaching[3]. Moreover, vision is the most important facilitator for exploring the environment in relation to oneself[4] through functional and symbolic play[5]. This thesis focuses on motor control, measurement of the development of fine motor skills and possible interventions in children with visual impairment.

Brambring[6] defined primary and secondary functions of vision in the acquisition of motor skills. The primary functions are important for adequate reactions to changes in the environment, for keeping a person in touch with the environment and for anticipating in advance dangerous situations. As primary functions he mentioned: (1) an incentive function to get engaged in movements[7,8]; (2) a spatial function to permit the simultaneous and precise perception of the position, shape and size of (moving) objects and the spatial position of objects and subjects[9]; (3) a protective function to recognize and anticipate dangerous situations sufficiently in advance; (4) a controlling function to track the performance of a movement, which is particularly decisive for new or complex movements; and (5) a feedback function to monitor the quality of executed movements; that is, to fine-tune and automatize movement sequences.

The secondary functions[6] are defined as (1) a social feedback function to encourage children to try certain motor actions or to desist from others, mainly expressed non-verbally and conveyed through glances, facial expressions and gestures from others in the neighbourhood, and (2) an observation function to imitate motor acts that are performed by other children or adults. Learning is reinforced by rewards and sanctions, which are given mostly non-verbally. Children with low vision are less stimulated to move and to practise activities because they are less encouraged by the physical and social environment.

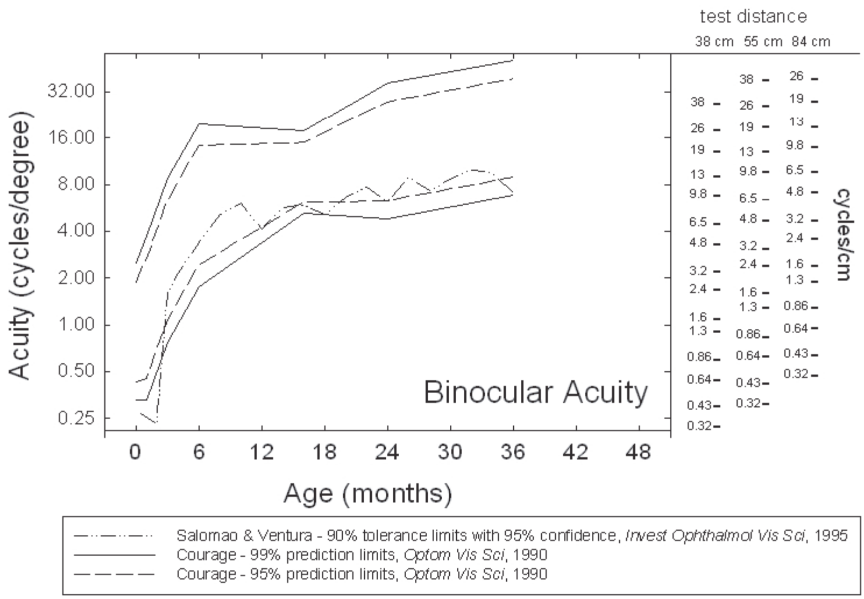
### *Postnatal development of the visual system*

From birth to complete maturity, the eye increases in size. The axial length increases rapidly in the first 18 months[10,11], and most of the growth of axial length is completed by the age of 13; most of the postnatal growth of the corneal diameter occurs in the first 7 years of life[11]. The following information gives an indication of normal visual development in young children from birth to 3 years and the related functional brain development[2].

Although at birth the visual system is fully present, it develops after birth based on visual stimulation from the environment. The pupils are not yet able to dilate fully. The newborn often has not yet developed adult eye movements e.g. fixation. The newborn has a very limited ability to discriminate colour, has limited visual fields and an estimated visual acuity of between 20/1200 and 20/400[2,12,13].

Measurements with forced preferential looking suggest an ability of 20/100 by the age of one year and 20/20 by the age of 3 years[13,14]. There is preference for high-contrast black and white designs. Significant improvement occurs during the first few months of life. Vision develops when it is used, so providing visual stimuli at the right moment is essential. The visual system does not reach full maturity before adulthood. Myelination of the visual system has its peak in the first year after birth but continues into the third decade of life[15-17]. When the brain and the fovea mature the visual acuity improves rapidly after birth. The fixation is intermittently present.

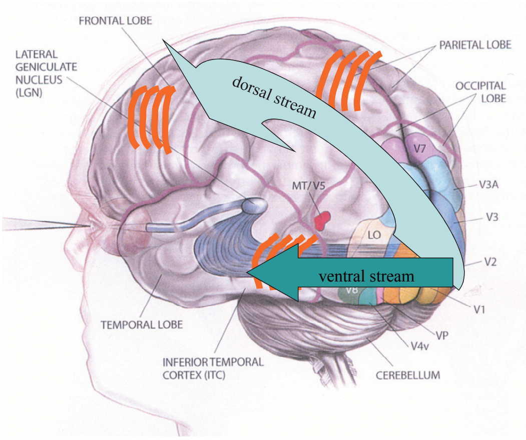
**Figure 1. Development of visual acuity (VA): Development of VA measured binocularly by Teller acuity cards (grating acuity at 55 cm), and Snellen equivalent (optotype acuity at 6 metres). (Figure derived and adapted from Teller acuity cards™ II handbook, 2005[18], permission pending).**




The visual fields expand after birth, ranging from approximately 30% of adult values at 2 months to 75%–80% at 8 months and 100% at 2 years[19,20].

The functional development of brain substrates for the perception of complex visual scenes needs continuous interaction with the environment leading to myelination of neurons, growth of dendrites and changes in the density of synapses. Especially in early years there is a spurt of synapse growth followed by a period of pruning around the time of puberty[2].

**Figure 2. The ventral stream and dorsal stream[21]. M.M. van Genderen, Bartiméus, 2003, with permission.**




Two main streams of visual information are important in relation to motor development: the ventral stream runs to the temporal cortex areas, it has been proposed that this stream is related especially to visual recognition and the interpretation of the visual world ('What/Who'). The dorsal stream runs to posterior parietal cortex areas, it has been suggested that it plays a role in the estimation of distances and positions of (moving) objects ('Where')[22,23]. In this model, both networks play a role in unconscious processing of visual-spatial information. Information from both streams is important for action planning: to reach and grasp or to walk over obstacles one needs to know *what* the obstacle is and *where* it is. Recently, however, a different functional interpretation of the ventral and dorsal stream has been proposed, connecting them more directly to the planning and control of action, respectively[24]. When motor activities are practiced the visuospatial information and related motor actions become functionally coupled. This means that learning fine motor skills will be connected to the specific abilities of the child to process: visuospatial information: disturbed acuity or a smaller visual field will have different implications for motor control and motor learning. This needs to be taken into account carefully in designing learning environments and interventions.

**VISUAL IMPAIRMENT**

In this thesis the guidelines of the World Health Organization (WHO)[25] were used to classify visual impairment (VI)[26,27]. In the Netherlands, in children between 0 and 15 years, VI is present in 8/100.000 per year (www.vision2020.nl), based on the extrapolation of data from Scandinavian blindness registers to the corresponding age groups in the Dutch population[26]. A minority of children with VI need special education (752 children in 2012), whereas 2373 children in 2012 were taught within regular schools[28]. In the Netherlands, child development is longitudinally monitored at different calendar ages by the Dutch Centre for Youth Health. Early identification of VI is part of this monitoring programme[29-31].

The first WHO criterion for classifying *low vision* is based on the visual acuity of the best eye. Low vision is present if, despite optimal glasses or lens correction, visual acuity is less than 0.3 (20/67 Snellen Equivalent), but is better than or equal to 0.05 (≥20/400), and/or there is corresponding field loss to less than 20°(20 degrees)[25]. Visual acuity is expressed as a fraction: a visual acuity of 1/10 means that one can recognize a standardized symbol at a distance of one metre while the normal eye can see see the same symbol at a distance of 10 metres.

As a consequence of low visual acuity problems arise with the recognition of shape and size and the spatial position of (moving) objects and subjects[9] with, as a consequence, reduced anticipation and monitoring of precise movements, and possibly diminished feedback on the quality of executed movements, for instance in reading and handwriting. Moreover, recognition of facial expressions, gestures and motor acts of other people is diminished.

The second WHO criterion for *low vision* is the size of the field of view. This is indicated in degrees. When a visual field is smaller than 30 degrees, which is accompanied by permanently impairment of vision, this will be classified as a VI. Blindness is defined as: vision of ≤ 3/60 and a field of view of ≤ 10 degrees[25].

Examples of possible effects of visual field defects on motor skills are problems with recognition and action planning related to objects in the unseen part of the visual field and problems with spatial orientation and mobility[32]. Visual field defects are subdivided into[32] central visual field defects and peripheral field defects[32]. In central field defects there are abnormalities (scotoma) in the area of central fixation. If there is a central field defect the older child will try to fixate eccentrically. The consequences of this are that although a child can move easily across a room and pick up a grain of rice or see a spot, they will have problems with reading and writing[32,33].

Abnormalities with peripheral visual field defects can cause partial field loss or hemianopia (loss of the right or left side of the field) or loss of the lower field. As a consequence of hemianopia the child may have a lack of detection or awareness of objects and events in the unseen visual field leading to less exploration and no anticipation of environmental changes (for instance grasping a rolling ball). Objects can be presented in the child's remaining visual field and children need to learn to adapt their head and posture position to be able to anticipate earlier. In case of visual field effects in the lower half of the visual field (e.g. in cerebral visual impairment) there is an increased risk of falling over objects on the floor. When performing fine motor skills the head is more in a prone position and there will be a need to set the table diagonally position[32].

Near visual acuity is not mentioned as a criterion for visual impairment. Consequently a reduction of *near vision* is not always diagnosed adequately in young children. A reduction of near vision can be measured with a method for near visual acuity[34] on a distance of 40 cm from the age of four years. The consequences of low near vision can be (for instance) shortening the distance between eyes and symbols during reading or writing, and during the manipulation of small objects. This shortening of distance has an effect on posture when performing fine motor skills. The visual field will be smaller, leading to a lower perception of details in social interaction and the environment. So, *low near vision* will have an impact on both the primary and secondary functions of vision.

All children with VI in the studies in this dissertation were include on the following criteria: a visual acuity of at least 0.05 (20/400) and ≤ 0.3 (20/67), that is, severe to mild impairment; with and without visual field defects and birth at term (i.e. ≥ 36 weeks of gestation) with normal birth weight. Co-morbidity and/or cognitive impairments were exclusion criteria.

A large number of children with VI in the studies presented in this thesis have nystagmus. Nystagmus is a condition in which the eyes make repetitive, involuntary oscillations[35,36] often resulting in reduced vision. These involuntary eye movements can occur from side to side, up and down or in a circular pattern. As a result, both eyes are unable to fixate steadily on objects that need (or are desired) to be viewed. A visually impaired child may hold the head in a position (head turn) in order to minimize the effect of the nystagmus and to improve visual acuity[37]. This means that the (asymmetric) preferred position of the head (head turn) is applied when a high degree of accuracy is needed, for instance in writing or reading tasks, catching a ball or finding visual stability during balance control.

## THE INFLUENCE OF VISION ON (FINE) MOTOR DEVELOPMENT

As previously mentioned, sensory systems are used for staying in contact with the world around us. The subsystems are the visual and auditory system but also the smell, taste, balance, vestibular, proprioceptive (position of the body) and touch sensations[38-40]. Motor development of children and sensory functions are inextricably linked. Children with VI are partly or completely deficient in the input of one of the vital sensory systems. Therefore they need to learn to use other sensory functions more intensively. If visual information is incomplete or impoverished, the information necessary for action becomes more dependent on other sensory information: tactile, auditory, vestibular and proprioceptive information and the sense of smell.

Research indicates that the scores of kinaesthetic perception of children with VI are lower[41]. In general, children with VI need to be extra motivated and stimulated to actively explore their environment and to become engaged in fine motor activities (for instance writing). Children with severe VI appear to have an increased prevalence and seerity of regulation disorders[42].

There are several theories about perception and the role of perception in motor control. In this thesis perception is seen as an active process of detecting and attuning to the relevant information in the environment via all available sensory channels together. Action is necessary for gathering and integrating the perceptual information, which, in turn, guides action. Perception–action couplings can loosely be defined as temporary stable, softly assembled synergies between perception and action subsystems that are functional in specific action contexts[7,43]. The visual system, in particular, is part of perception–action couplings that allow us to meet the complex demands coming from a dynamic surrounding in a task-specific way. The presence of a visual impairment affects such perception–action couplings[44]. In goal-directed movements, visual information is relevant at the start of the movement to detect information about the configuration, distance and direction of (moving) objects[45,46] and during the movements to control the actions in such a way that goals are reached[6].

Eye-hand coordination begins to develop as the infant starts tracking moving objects with his or her eyes and reaching for them. From eight weeks, babies begin to follow adults, and around three months of age objects, with the eyes and try to reach and grasp[12,47,48]. By six months infants reach out for small objects and start to perceive stereopsis, which means that they develop the ability to judge whether objects are nearer or further away. Infants become increasingly aware of the environment by means of vision and are able to judge whether they can reach objects or not. At around nine months of age, most babies begin to pull themselves up to a standing position. Vision motivates and monitors movement toward a desired object. By ten months of age, it is expected that a baby should be able to grasp objects with the thumb and forefinger. By 12 months of age, most babies will be crawling and trying to walk[49]. Gibson and Walk concluded that the ability to discriminate depth and perceive the possibilities for locomotion that a surface affords is related to a child's abilities to crawl and walk[50]. At that age, babies are able to judge whether, and how, they might go down a ramp. They recognize familiar objects and pictures in books and can scribble with a crayon or pencil[48]. By three years the child can complete a simple board correctly (based on visual memory), do simple puzzles, draw a crude circle and put pegs into holes. By 5–7 years, it is known that the basic functions of early sensory areas of the cortex have completed their development.

In the development of eye-hand coordination, visual acuity and the size of the visual field are critical components. For manual activity it is important that the gaze is stabilized, so adequate postural control of the head and trunk is crucial[51]. This means that children with VI have specific challenges and need to adapt while learning motor skills in daily life. Children with VI experience barriers in developing adequate postural control, noticing objects, imitating movements[52], initiating social interaction and monitoring the consequences of their own actions. This will influence their playing skills and social contacts. For children with VI the social and physical environment needs to be adapted in such a way that the environment will facilitate learning experiences. Children with VI without behavioural difficulties can develop sufficient abilities in a playful manner. However, they need adapted play materials and external feedback to strengthen the intrinsic motivation to move and explore[5,53].

**INTERVENTIONS TO INCREASE MOTOR SKILL LEARNING IN CHILDREN WITH VI**

Children with VI are frequently referred for physiotherapy assessment and intervention when there is an indication of problems with motor control. Intervention and treatment of *sensory* impairments should begin as early as possible in a positive emotional setting that enhances the child's motivation and relationship with carers[54].

Most research is focused on gross motor skill learning in children with VI[55-63]. As can be expected in children with VI, gross motor skill performance is generally poorer than in children without VI. Motor milestones such as walking independently, walking up and down stairs, running and jumping are reached at a later age and velocity and accuracy are lower. Although fewer studies focus on fine motor skills, the literature indicates that fine motor skills acquirement is also slower in children with VI[58-61], observed as less exploration, imitation, practice and refinement of manipulating skills. Children with VI need more time to learn an adequate grip when using a spoon, blocks and crayons, and 'school skills' such as block building, pasting, colouring, handwriting and using scissors also appear to be delayed[68]. There are only a few studies related to evidence of the negative influence of vision on fine motor skills in the case of amblyopia[69-71].

Generally, preschool occupational or paediatric physical therapy interventions focus on fine and gross motor activities, sensorimotor integration and perceptual training[72] and orientation and mobility tasks[4]. A review of the effects of *motor* interventions to improve motor, cognitive and/or social functioning was given by Houwen et al.[73] in 2014. In this review it is concluded that motor interventions have beneficial effects on the motor skill performance of children with VI in both gross and fine motor skills. Moreover, it was concluded that in particular manual dexterity tasks requiring speed and accuracy were difficult for children with VI[73] .

All in all it can be concluded that visual impairment has substantial consequences for motor learning and motor performance. However, there is evidence that these consequences are partly the result of diminished learning experiences. To detect the children at risk it is crucial to get an insight into the motor developmental trajectories of children with VI using reliable and valid motor performance tests. These tests are available for developing normally sighted children but not yet for children with VI. This thesis focuses on the development and measurement of fine motor skills in children with VI.

## STRUCTURE OF THIS THESIS

In **Chapter 2** we carry out a study to explore the influence of visual impairment on the performance of goal-directed movements. Little is known about potential differences in motor control, and whether it is fully developed or accurate in children with VI. In this study we have focused on differences in motor control between children with visual impairment and children with normal vision when performing repetitive goal-directed aiming movements.

We expected that children with visual impairment would be less accurate in the execution of aiming movements, and we expected that the lack of visual information would influence the fluency of discrete movements more than the fluency in cyclical repetitive movements.

In an earlier study, the ManuVis was developed as a test for fine motor skills in children with VI at the age of 6–10 years to determine whether children need extra support. In **Chapters 3 and 4**, two studies are described regarding the applicability of the ManuVis[74] for children with VI at the age of 3–11 years. The test–retest reliability and inter-rater reliability of the ManuVis were investigated, reference scores were collected and we compared the fine motor skill development of children with VI with typically developing children.

**Chapters 5** and **6** focus on magnifier training for children with VI and its additive value for fine motor skills. Children were trained to use a magnifier in a trail-following task. A stand magnifier enlarges the symbols of the trail and provide a stable image. In the experiment in this thesis, children with VI had to follow trails visually, from a start to an end location, with or without a stand magnifier. In **Chapter 5** we test the effect of training on task performance comparing trail finding with and without a magnifier. In **Chapter 6** we measure the potential spin-off effect of magnifier training on other fine motor skills of children with VI. We expected that training with a magnifier would improve their attentional focus when performing fine motor skills and that this is beneficial to fine motor skill performance.

**Chapter 7** discusses the influence of visual impairment on motor development and the implications for clinical practice. Suggestions are formulated for further research.

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# Chapter 2

## Effect of visual impairment on goal-directed aiming movements in children

Annemieke Reimer
Ralf Cox
Nienke Boonstra
Bouwien Smits-Engelsman

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**ABSTRACT**
This study investigated potential differences in motor control between children with a visual impairment (diagnosed albinism; *n*=11, mean age 8y 4mo [SD 7mo]; seven males, four females,) and children with normal vision (*n*=11, mean age 8y 4mo [SD 7mo]; six males, five females). Mean near visual acuity in the albinism group was 0.19 (SD 0.07, Snellen: 20 ⁄ 104). Children performed two types of movements (discrete and cyclic) in two orientations (azimuthal and radial, i.e. along the viewing and lateral direction), and with two amplitudes (10 and 20cm). All movements were performed in two subsequent target conditions: with and without visual information on the target location. Overall, children with visual impairment displayed larger endpoint variability. Discrete movements and movements over large distances were less fluent in both groups, but especially in the children with visual impairment. Children with visual impairment seemed to have more difficulties with calibrating the sensory information. Specifically, they made larger errors along the lateral direction, when the target was not visible. Results suggest that children with visual impairment have specific differences in motor control compared with children with normal vision, which are not all directly related to their poorer vision.

## INTRODUCTION

Several studies have demonstrated the importance of visual guidance for achieving speed and accuracy in goal-directed movements. Although brief visual samples of the movement environment are sufficient for reasonably precise, closed- loop control[1], vision remains of significant importance for optimal accuracy, even after extended practice[2]. Visual information for controlling goal-directed movements is used in different ways depending on the movement phase[3,4]. In the initial (ballistic) phase of a rapid aiming movement, vision makes a significant contribution to determining the direction of the movement. One or more saccades direct the eyes to the target location. Because this usually precedes limb movement, the eyes focus on the target before the finger[5,6]. In the final (homing-in) phase of the movement, vision is particularly important for movement accuracy. Visual feedback enables corrections in a later part of the trajectory so that the hand can reach the goal precisely. Therefore, fine eye hand coordination requires setting up a temporary, task-specific synergy between the eyes and hand[7].



Movement control in children aged 6 to 8 years becomes more dependent on visual information because the ability to use alternative perceptual strategies becomes limited[8]. A straightforward hypothesis would be that visual impairment has a negative influence on children's eye–hand coordination, as less visual information is available to guide the movement towards its target. Although differences in motor development between children with and without visual impairment have been observed[9-11], scant experimental research has been carried out to investigate potential differences in the kinematics of goal-directed movements between these groups under different movement conditions.

In the present study, children with visual impairment diagnosed with albinism, and children with normal vision performed repeated goal-directed aiming movements[12], first several times with and subsequently several times without visual information of the target location. As mentioned above, speed and accuracy of such movements depend on real-time information from the visual system.

In addition, information coming from the proprioceptive system also plays an important role[13,14]. Between the two phases of the task (i.e. with and without view of the target location), the influence of the visual and proprioceptive subsystems will be different. Accuracy is expected to be lower in the second phase and it is likely that the movements will be guided more on the basis of proprioception, even though there is feed- back from the hand and visual memory of the experiment is set-up[15,16]. However, in the first phase of the task the visual feedback might be used to calibrate the proprioceptive system.

In addition to the two target conditions, several different movement conditions were presented. By varying the orientation, mode, and amplitude of the movements it was possible to examine the interaction of visual and proprioceptive information over a wide range of naturally occurring movement conditions. The possible influences these variations might have on movement control in the two groups can be described in two ways.

First, children had to carry out the movements along the viewing direction (azimuthal) as well as the lateral direction (radial). The literature concerning asymmetries in goal-directed movements report that each hand is faster when moving in the ipsilateral hemispace[17], and with respect to accuracy, there are also reports of fewer errors towards the ipsilateral side of the body[18,19]. The level of integration between visual and proprioceptive feedback also varies with movement orientation. Hand-position estimates rely more on proprioception along the lateral direction than along the viewing direction, and are also more accurate along the lateral direction[20]. Based on these findings, the impact of visual impairment on movement speed and accuracy is expected to be different between these two movement conditions, as well as between the two vision conditions.

Second, children had to perform the (repeated) movements in a discrete and a cyclic mode, i.e. with a distinct start signal at each individual movement and in a continuous fashion respectively. Both kinds of movements are frequently performed in daily life, but are considered to rely on different control mechanisms[21]. Discrete movements have a clear beginning and end and, therefore, are usually controlled more by feedback than cyclic movements. Visual guidance is the most important control input for these corrective movements. During the main part of the movement, as well as during the homing-in phase of a discrete movement, many corrective changes can be made based on visual feedback; however, this will lead to longer movement times.

Contrary to discrete movements, cyclic (or rhythmical) movements are more of a ballistic nature. These movements rely more on an open-loop control strategy in which less-corrective sub-movements are made and fewer changes in speed occur[21]. It might be that children with a visual impairment, generally, rely more on open-loop control. However, it is unclear whether this leads to better performance. Moreover, using an open-loop control strategy, especially in the second phase of the task (without view of the target location) requires good calibration of the proprioceptive subsystem based on experience and fine-tuning in the first phase (with view of the target location). Little is known about such sensory calibration, and whether it is fully developed or accurate in children with visual impairment. The aim of this study was to investigate potential motor control differences between children with a visual impairment and children with normal vision in repetitive aiming goal-directed movements.

## METHOD

### *Participants*

A group of 11 children with a visual impairment diagnosed with albinism (seven males, four females), mean age 8 years 4 months (SD 7mo), and a group of 12 children with normal vision (seven males, five females), mean age of 8 years 6 months (SD 7mo) participated in this experiment. Albinism is a hereditary genetically determined disorder of the melanin synthesis within pigment cells that has widespread and variable effects on the eyes, visual system, and the skin, often accompanied by a misdirection of the optic nerve fibres[22]. This study only included children who had no demonstrable neurological disorder, intellectual impairment, nor other pathology.

2

Nine children with a visual impairment attended regular primary education and two attended classes for special education at the Bartiméus, Zeist, the Netherlands. All children in this group were recruited from Bartiméus' archives. Children with normal vision all attended regular primary education, and were contacted through the Ichthus and De Sluis School, Zeist. Permission for the study was obtained from the board of managing directors of the institute. After written invitation, informed consent was received from parents of the participants. The study was performed in accordance with the ethical standards of the Declaration of Helsinki (1964).

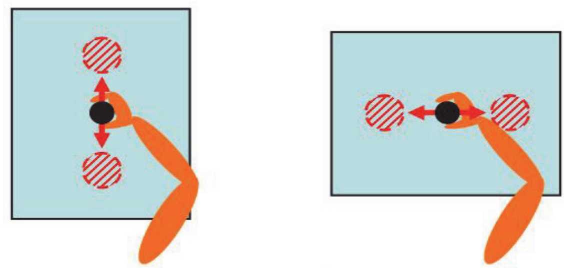
Mean visual acuity in children with visual impairment (measured in 10 children with the nystagmus) was 0.19 (SD 0.07) Snellen 20/104. In addition, they had varying degrees of nystagmus, which is typical for albinism[23]. People with nystagmus reduce the negative effect of the condition by holding their heads at a tilted and/or rotated way (torticollis). Thus each child had a different head orientation to ensure that it was least disturbing.

Fine-motor skills were tested with the Manuvis test[24], which provides specific norms for poor-sighted children. All children who scored higher than the 15th centile were classified as having normal motor development. One child with normal vision who scored lower than the 15th centile was not included in the analyses, leaving 11 children in the comparison group. On average, the children with visual impairment scored 13% lower on the Manuvis test and performed the fine-motor tasks slower than the children with normal vision. Hand preference was indicated by the hand that children used to write[25] In each group there were two left-handers.

### *Material and procedure*

Children had to move a small puppet over the surface of a digitizer (sample rate 206Hz; Wacom, Saitama, Japan; type Cintiq 18sx) which was positioned horizontally in front of the shoulder of their preferred hand (Figure 1). The digitizer incorporated a high-luminance LCD monitor (SXGA 24-bit full colour), which was used to display the targets that served as the beginning and end-points for each movement. Targets were circles of 2.5 cm in diameter.

**Figure 1.** **Top view of the experimental set-up for two orientation conditions: (a) azimuthal and (b) radial. In both conditions, movements were performed for target distances of 10cm and 20cm, and in a discrete and continuous fashion. In all conditions movements were performed 10 times with, followed by 10 times without, visual information of the target location.**




Before the start of each trial, the child was asked to position the puppet in the starting circle on the digitizer, after which the experimenter started the experiment. A random period of about 0.5 to 1.5 seconds later, an acoustic signal was given and the target circle appeared on the digitizer. This was the indication for the child to move the puppet as fast and as accurately as possible towards the target location. Children performed the experiment with their preferred hand, and they could observe the puppet's motion as well as that of their arm at all times. All children started with a practice session.

Several different movement and vision conditions were presented to each child in separate blocks. To determine whether amplitude has a differential effect, participants had to move the puppet over two distances: 10cm and 20cm between starting circle and target circle (Index of Difficulty is 3 and 4, respectively; with A the amplitude of the movement and W the target width[12]). Moreover, movements were per- formed in two orientations relative to the body: azimuthal and radial, i.e. along the viewing direction and along the lateral direction respectively. Furthermore, the mode of the movements was varied as discrete and cyclic. In the discrete mode, the procedure described above was repeated 10 times. In the cyclic mode, after the starting signal, children moved the puppet between the two targets for a period of 6 seconds. The order of presentation of the eight resulting movements condition blocks (2 amplitudes 2 orientations 2 modes) was counterbalanced across participants.

Finally, within each of the movement conditions, visual information about the location of the target circle was varied (target condition). Movements were performed first with visual information of the target's location, then as part of a series without visual information of the target's location. In every block the without-target condition was always presented after the with-target condition.

**Data analysis and dependent measures**

Movements were recorded and analyzed using OASIS software[26]. First, data -handed participants was switched between left and right to pool it with that of the right-handed participants. Second, the first movement of each condition was not used in the analyses, and the median value was used to filter out possible outliers in every condition. After this, a set of dependent variables was calculated for each child and each condition by averaging individual movements. Finally, each of these variables was entered in a repeated measures analysis of variance with three within participant factors (amplitude, orientation, and mode) and one between participants factor (vision group). Analyses of variance (ANOVAs) were performed separately for each of the two vision conditions. The assumptions underlying ANOVA were checked by spread-versus-level plots and residual plots, and by homogeneity of variance tests (Levene test). Significance was set at $p<0.05$ (two-tailed).

Four dependent variables were derived from the movement data: endpoint variability, reaction time, movement time, and peak-over-mean velocity. As a measure of movement accuracy, the endpoint variability was calculated. Endpoint variability was defined as the distance (centimeters) between the centre of the puppet and the centre of the target circle. Reaction time was defined as the temporal delay (seconds) between the acoustic signal and the start of the movement. Reaction time was only determined for the discrete conditions. Movement time was defined as the single movement duration (seconds), i.e. the time it took the child to move the puppet once from the starting circle to the target circle. Measurement of movement time started at the first displacement of the puppet and ended when its speed dropped below 0.2 cm/s in the target circle. Finally, as a measure of the ballistic nature of the movement, and to quantify the extent to which movements were produced by open-loop or closed-loop control, the ratio of peak-over-mean velocity was calculated.

## RESULTS

The experimental design required participants to perform two types of movements: movements towards the body or midline (i.e. retraction or adduction) and movements away from the body or midline (i.e. protraction or abduction). For three of the dependent variables (movement time, reaction time, and peak-over-mean velocity), preliminary analysis revealed no difference between these two types of aiming movements, and data were pooled within each condition; however, differences were found for endpoint variability in both target conditions.

Tables 1 and 2 display group averages (pooled) of the four dependent variables as a function of the amplitude, orientation, and mode of the movements, in the conditions where visual information on the target's location was present (with-target condition) or absent (without-target condition) respectively. There were no missing data.

**Table 1. Dependent variables for each vision group as a function of amplitude, orientation, and in the with-target condition [a]**

| | Visually-impaired group (n=11) | | | | | | Normal vision group (n=11) | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | Amplitude (ID) | | Orientation | | Mode | | Amplitude (ID) | | Orientation | | Mode | |
| | 10cm (3) | 20cm (4) | Azimuthal | Radial | Discrete | Cyclic | 10 cm (3) | 20 cm (4) | Azimuthal | Radial | Discrete | Cyclic |
| EV (cm) | 0.478 | 0.552 | 0.495 | 0.534 | 0.393 | 0.636 | 0.396 | 0.452 | 0.419 | 0.429 | 0.328 | 0.520 |
| MT (s) | 0.755 | 0.988 | 0.884 | 0.860 | 0.978 | 0.765 | 0.744 | 0.982 | 0.844 | 0.883 | 0.909 | 0.817 |
| RT[b] (s) | 0.577 | 0.503 | 0.498 | 0.582 | 0.540 | - | 0.417 | 0.432 | 0.411 | 0.438 | 0.425 | - |
| PoM | 1.952 | 2.026 | 1.995 | 1.983 | 2.133 | 1.845 | 1.863 | 1.929 | 1.911 | 1.881 | 1.970 | 1.823 |

[a]Values in table are means. [b]This variable is only relevant to discrete mode. ID, Index of Difficulty; EV, endpoint variability in centimetres; MT, movement time in seconds; RT, reaction time in seconds; PoM, peak- over-mean velocity.

**Table 2. Dependent variables for each vision group as a function of amplitude, orientation, and mode in the without-target condition[a]**

| | Visually-impaired group (n=11) | | | | | | Normal vision group (n=11) | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | Amplitude (ID) | | Orientation | | Mode | | Amplitude (ID) | | Orientation | | Mode | |
| | 10cm (3) | 20cm (4) | Azimuthal | Radial | Discrete | Cyclic | 10 cm (3) | 20 cm (4) | Azimuthal | Radial | Discrete | Cyclic |
| EV (cm) | 1.169 | 1.325 | 1.127 | 1.367 | 1.267 | 1.226 | 0.959 | 0.925 | 0.970 | 0.915 | 0.993 | 0.892 |
| MT (s) | 0.749 | 1.021 | 0.893 | 0.877 | 1.044 | 0.725 | 0.763 | 1.018 | 0.860 | 0.921 | 1.010 | 0.770 |
| RT[b] (s) | 0.511 | 0.486 | 0.494 | 0.502 | 0.498 | - | 0.425 | 0.417 | 0.422 | 0.0421 | 0.422 | - |
| PoM | 1.872 | 1.930 | 1.921 | 1.881 | 2.073 | 1.728 | 1.822 | 1.860 | 1.842 | 1.840 | 1.961 | 1.720 |

[a]Values in table are means. [b]This variable is only relevant to discrete mode. ID, Index of Difficulty; EV, endpoint variability in centimetres; MT, movement time in seconds; RT, reaction time in seconds; PoM, peak- over-mean velocity.

**Endpoint variability**

In the without-target condition (see Table 2) a significant overall difference was found between the two vision groups with respect to endpoint variability ($F(1,21)=10.71$, $p=0.004$). In the with-target condition (see Table 1) the overall difference in endpoint variability was not significant ($F(1,21)=3.99$, $p=0.059$), which might be due to insufficient power. Children with visual impairment were less accurate than children with normal vision: mean error sizes were 1.25cm versus 0.94cm in the without-target condition, and 0.51cm versus 0.42cm in the with-target condition respectively. An interaction effect of orientation and vision group in the without-target condition ($F(1,21)=5.20$, $p=0.033$) revealed that movement accuracy in the visually-impaired group was poorer in the radial orientation than in the azimuthal orientation (1.37cm vs 1.13cm), while it was about the same in the normal-vision group (0.92cm vs 0.97cm).



When the target was visible, differences in endpoint variability were not equal in size for different amplitudes ($F(1,21)=10.13$, $p=0.004$) and for different modes ($F(1,21)= 56.92$, $p<0.001$).Wider movements (20cm/ID, Index of Difficulty=4) and cyclic movements resulted in more error in both vision groups. These differences between movement conditions were not present when the target was no longer visible, and performance was generally poorer.

As mentioned above, there were differences for endpoint variability with respect to the direction of the movements (i.e. between movements to proximal targets and distal targets) in both target conditions. In Table 3, endpoint variability in both target conditions is separated for proximal and distal movements. Differences in accuracy are quite large in the without-target condition and oppositely directed to those in the with-target condition. Without visual feedback of the target, movements away from the body are significantly more accurate than movements towards the body ($F(1,21)=10.66$, $p=0.004$). With visual feedback of the target, the (opposite) effect proved not to be significant ($F(1,21)=4.15$, $p=0.055$).

**Movement time**

Overall, movement time was not significantly different between children with visual impairment and children with normal vision. In both target conditions movement time varied as a function of amplitude, in such a way that movements over a larger distance took more time to perform ($F(1,21)=194.48$, $p<0.001$, in the with-target condition; and $F(1,21)=120.73$, $p<0.001$, in the without-target condition). A similar difference was present for mode, such that discrete movements took more time than cyclic movements ($F(1,21)=13.84$, $p=0.001$, in the with-target condition; and $F(1,21)=35.85$, $p<0.001$, in the without-target condition).

**Table 3. Endpoint variability in both target conditions separated for movements to proximal (left/down) and distal (right/up) targets[a]**

| | | | Visually-impaired group (n=11) | | Normal vision group (n=11) | |
|---|---|---|---|---|---|---|
| | | | **Proximal movement** | **Distal movement** | **Proximal movement** | **Distal movement** |
| With target | Amplitude (ID) | 10cm (3) | 0.448 | 0.507 | 0.392 | 0.400 |
| | | 20cm (4) | 0.511 | 0.592 | 0.422 | 0.481 |
| | Orientation | Azimuthal | 0.487 | 0.502 | 0.417 | 0.420 |
| | | Radial | 0.472 | 0.596 | 0.397 | 0.461 |
| | Mode | Discrete | 0.341 | 0.445 | 0.336 | 0.319 |
| | | Cyclic | 0.618 | 0.654 | 0.478 | 0.562 |
| Without target | Amplitude (ID) | 10cm (3) | 1.374 | 0.963 | 1.072 | 0.846 |
| | | 20cm (4) | 1.519 | 1.130 | 1.039 | 0.811 |
| | Orientation | Azimuthal | 1.366 | 0.887 | 1.123 | 0.816 |
| | | Radial | 1.527 | 1.206 | 0.989 | 0.841 |
| | Mode | Discrete | 1.465 | 1.069 | 1.186 | 0.799 |
| | | Cyclic | 1.428 | 1.024 | 0.925 | 0.858 |

[a]Values in the table are means.
[b] The two orientation conditions perfectly separate movements into orthogonal directions: In the azimuthal orientation, movements were in the down-up direction. In the radial orientation, movements were in the left-right direction. ID, Index of Difficulty

### Reaction time

No effect of the different movement and target conditions or between the vision groups was found for reaction time, although a trend can be detected from the results. Averaged over all conditions, reaction times for the children with visual impairment were 0.54 seconds in the with-target condition and 0.50 seconds in the without-target condition. For the children with normal vision this was 0.42 seconds in both target conditions.

### Peak-over-mean velocity

Peak-over-mean velocity varied as a function of the amplitude and the mode of the movement in both target conditions. Peak-over-mean velocity was higher for movements over a larger distance ($F(1,21)=14.97$, $p=0.001$, in the with-target condition; and $F(1,21)=9.66$, $p=0.005$, in the without-target condition). With respect to mode, peak-over-mean velocity was higher for discrete movements than for cyclic movements ($F(1,21)=55.67$, $p<0.001$, in the with-target condition; and $F(1,21)=88.24$, $p<0.001$, in the without-target condition).

A significant interaction between mode and vision group in the with-target condition ($F(1,21)=5.93$, $p=0.024$) revealed that for the discrete movements, children with a visual impairment had a higher peak-over-mean velocity than children with normal vision (2.13 vs 1.97). For the cyclic movements this difference did not appear to be present (1.84 vs 1.82). No further group effect was found.

## DISCUSSION

The purpose of this study was to investigate potential motor- control differences between children with a visual impairment and children with normal vision in repetitive aiming movements. It is the first study concerning children with visual impairment in this respect, especially those diagnosed with albinism who have varying degrees of nystagmus[23]. Results demonstrated that children with visual impairment were less proficient in performing the movements under the various conditions presented. The study compared children's performance under various movement conditions (amplitude, orientation, and mode) and target conditions (i.e. visibility of the target location). Arguably, these conditions give rise to different types of motor control[21].



### Accuracy of movement

The hypothesis that children with visual impairment are less accurate in the execution of aiming movements was confirmed in this experiment. In particular, and quite surprisingly, movement accuracy suffered more in this group in the phase with no visual information on the location of the target. A possible explanation for this might be found in calibration processes between sensory subsystems. This argument of calibration has two aspects: developmental and task specific.

It is known from studies of early motor development that the vestibular and proprioceptive subsystems are calibrated using visual feedback[11]. This calibration is necessary for postural control, but also for fine-tuning of prehension movements. Prechtl et al.[11] reported that in infants with profound visual impairment a delay is present in the development of proprioception caused by the lack of visual integration, and that it is still unclear if this delay is fully recovered later in life. It is reasonable to suspect that proprioception is also less adept in children with visual impairment, albeit less severe. Less optimally calibrated sensory subsystems might have led to less accurate movement control when the target was not visible.

Calibration plays a crucial role in adaptive action control, under changing task constraints or when sensory information about these constraints changes[27,28]. Even in the absence of a visible target, children still had some global visual information of the task setting (e.g. edges of the digitizer) and of the movements of their own arm. This information might have been used to continue to guide the movements visually (or at least partially), and if so it is likely that the children with poorer vision benefited less from this information. As a result, under these less than optimal feed- back conditions, their 'drift' away from the optimal movement trajectory would be larger, as was observed.

For both groups, when the target was no longer visible, accuracy deteriorated more for movements towards the body or midline. This suggests that the calibration process is different for proximally and distally directed movements. Perhaps related to this, for the children with normal vision, endpoint variability was about the same for the two orientations. However, the children with a visual impairment made larger errors in directing the puppet when moving along the lateral direction. All the children with visual impairment who participated in this study had albinism with nystagmus to some degree. The natural coping strategy for this is to hold the head at a certain angle, called ocular torticollis, to reduce the effects of this nystagmus. The result of this is a limited gaze, which had a negative influence on accuracy in the radial orientation. Another factor could be that radial movements rely more on visual information than on proprioceptive information[13]. Both factors may be responsible for the larger impact of poor vision in these tasks.

***Movement onset***

Visually-guided arm movements, such as reaching or pointing, are accompanied by saccadic eye movements that typically begin prior to movement initiation of the arm[6]. The group of children with visual impairment all had nystagmus. The type of head position, to ensure that the nystagmus was the least disturbing, varied for each child. It is possible that this influenced movement onset time because focusing on the target is more difficult, explaining the trend in reaction times between the two groups.

**Fluency of movement**

In the cyclic condition, vision is less crucial for guiding the movements. It is known that cyclic movements are performed under open-loop control, which relies less on visual control[19]. Results showed that peak-over-mean velocity, as a measure of the fluency of the movements, was lower during cyclic tasks. This suggests that fewer corrective movements were made in this condition, which makes the overall movement more fluent. When accuracy demands are higher, more changes in velocity peaks are expected[6,29]. Results of the current study concur with these previous findings and suggest an explanation for the differences in fluency between the two groups in the discrete tasks. Having less visual information seems to influence the fluency of discrete movements more than the cyclic movements.

The main limitations of the present study are concerning methodological issues and utilization. Methodologically, a more thorough research set-up is needed, for instance, using motion-capture and eye-tracking devices and/or electromyogram, to scrutinize the control systems involved in the various movement conditions and the calibration processes in the two target conditions. With respect to utilization, the research group is quite small, and although aetiological variations were limited, the group is still heterogenic in many respects. This makes an easy translation of the results to a medical or therapeutic setting quite difficult, because many more factors are involved, the influence of which we know very little about.

## CONCLUSION

This study shows that there are several differences between children with visual impairment and children with normal vision with respect to the control of aiming movements. Results emphasize that more research is needed regarding the influence of degraded visual information on movement in natural viewing and movement conditions. Follow-up studies might focus on the influence of nystagmus and eye movements. There is also scope for study into the differential role of developmental and task-related changes in the calibration and integration of vision and proprioception. A careful analysis could provide valuable insight into the development of motor control in children with visual impairment.

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# Chapter 3

## Measurement of Fine-Motor Skills in Young Children with Visual Impairment

Annemieke Reimer
Ralf Cox
Nienke Boonstra
Ria Nijhuis-van der Sanden


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**ABSTRACT**
Insight into the typical motor development of children with visual impairment (VI) is necessary in order to recognise whether children with VI are at risk of motor developmental problems, and to evaluate the effectiveness of exercise interventions. In 2003 the ManuVis was published with reference values for children with VI of ages from 6-11 years. This paper reports on a follow-up study of the ManuVis focused on: a) comparison of fine motor skills between children with VI and normal sighted (NS) children; b) sampling norm-references for children with VI in the 4-11 years age range to increase validity; and c) test-retest and inter-rater reliability. In total 256 children with VI and 162 NS children were included in the study. The results demonstrated that children with VI needed significantly more time than NS children to perform all test items, especially at younger ages. Performance time decreased in both children with VI and NS children from the younger to the older age groups, but NS children reached their minimum at a younger age. Test-retest reliability on the items varied from moderate to excellent and inter-rater reliability was excellent. The results suggest that children with VI have slower and more prolonged motor learning than NS children. The ManuVis differentiates between typical and atypical fine-motor performance of children with VI between 4 and 9 years of age, and is useful for monitoring fine-motor skills in children with VI from 4 years to (at least) 11 years.

## BACKGROUND AND RELEVANCE OF THIS STUDY

Brambring defined primary and secondary functions of vision in the acquisition of motor skills[1]. Primary functions are important for adequate reactions to changes in the environment, for instance vision plays a role in the detection of environmental cues to engage in movement, and visual-spatial perception plays a role in the detection of the position, shape and size of (moving) objects. Visual information plays an important role in providing feedback during the execution of movement, and on the results (for instance, whether a goal has been achieved or not). Secondary functions of vision are related to motor learning: for instance observation and the imitation of movement is one of the first motor learning strategies of young infants. Learning is strengthened by rewards and sanctions that are mostly expressed non-verbally, so children with low vision are less motivated to move because they are less stimulated by the physical and social environment[1]. Delays in motor development in children with visual impairment (VI) can be interpreted as related to visual constraints in movement execution, or to the conditions for skill learning. Motor skill learning processes are influenced by the expectations of the social environment, which are lower in children with VI than in normal sighted (NS) children[2]. People in the social environment need to be aware of the extra stimulation and of the support these children need to enable them to use alternative sensory information and to explore different strategies to reach their goal[1]. In clinical practice, it therefore seems important to gain insight into the typical motor development of children with VI and to develop instruments that enable clinicians to recognise those children at risk of motor developmental problems compared to their peers with VI. Currently, the use of valid and reliable instruments in daily rehabilitation practice for children with VI is scarce. In this article we describe a study involving the psychometric characteristics of the ManuVis[3], a measurement instrument to test fine motor skills in children with VI.



### Prevalence of visual impairment in children

Blindness is defined as a visual acuity (VA) of less than 0.05. In the Netherlands the prevalence of blindness was estimated in 2005 at 0.3/1000 in children between 0 and 14 years. The prevalence of severe and moderate visual impairment (0.05<VA<0.3) is about twice as common and estimated at 0.6/1000. The prevalence of all visual impairments (VA <0.3) in the age group between 0 and 15 years was estimated to be 0.9/1000 in the Netherlands, based on the extrapolation of data from Scandinavian blindness registers to the corresponding age groups in the Dutch population[4]. The incidence of VI in children is low, so the development of measurement methods such as a test of fine motor skills that is adapted for this population has not been a priority. The consequences of VI on motor development and daily participation are obvious, however, so reliable and valid instruments for this group of children at risk of stagnation in their fine motor skill learning seem to be highly important.

**Sensory-motor development and visual impairment**

Motor development and learning are geared by the interactions of a child with the material and social environment. Perceptual systems and motor systems become coupled into task-specific functional units called action systems[5,6]. It is well known that motor and perceptual development are strongly intertwined, working in unity rather than being separate processes[7,8]. Since the visual system provides information on shape, size, colour, distance, location and movement velocity, and the direction of objects and people in the environment, all in one glance, it is an essential part of action systems throughout life and for the development of young children in particular. In children with VI, sensory-motor experiences are, to a certain extent, limited or impoverished, and it is clear that this leads to differences in motor development and learning, compared to their NS peers[9-11]. Children with VI will have more problems focusing attention to relevant cues, which is especially important when anticipating changing and dangerous situations and when children learn by imitating movements made by others. In goal directed movement, visual information is relevant at the start of the movement to detect information about distance and the direction of movement and objects[12,13], and during the movements to control the actions in such a way that goals are reached[1]. Even if children with VI have adequate acuity for certain activities, they may not be able to deploy and interpret visual input in the same functional way as NS children, based on fewer experiences and learning opportunities compared to NS children[1]. Based on visual impairment and decreased learning experience children with VI generally have slower motor learning, and it has even been reported that certain motor milestones are progressed in a different order. Quantitative and qualitative differences in motor performance between children with VI and NS children are described in detail in many publications[11,14-23]. Taken together, it can be concluded from these studies that children with VI generally perform more poorly than their sighted peers in both gross and fine motor skills. Variability between individuals with VI is great, however. Only weak evidence is found for a relationship between the degree of VI and gross motor development and manual dexterity, and between amblyopia/strabismus and fine motor skills. A weak relationship is found between movement interventions and the level of motor skill performance. All other possible influencing variables tested on motor skill performance were inconclusive[10]. In a recent study Haibach et al.,[16] used the Test of Gross Motor Development (TMGD-2; [24]) to test the influence of the severity of VI, age, and sex on gross motor skills performance in children with VI at 6-12 years old. Although blind children scored significantly lower, no group difference was found between the children with severe and moderate VI and no influence of age and sex on test outcomes was found. These results confirm the findings in the review study[19]. This is not what might be expected: during typical child development, motor coordination gradually improves with increasing age[24-28] which would also be expected in children with VI. This means that either the tests used are not sensitive or adequate enough to detect the increasing performance of children with VI or that motor learning stagnates in children with VI based on lack of experience in sports and motor learning. This would mean that early detection and intervention, including instructions to persons in the social environment, are needed. It is found that early adaptation to visual impairment and increased orientation leads to higher mobility and motor performance important for social integration[29-31]. This social participation and acceptance is an important issue for children of 10-15 years with VI: for them this was related to independence and autonomy, and to psychological and emotional well-being[32].

**Fine-motor skills and visual impairment**

The major part of literature focused on the general motor development of children with VI. Especially fine-motor skills may be acquired more slowly by children with VI since vision enables an attunement to information for exploration, imitation, practice, and the refinement of manipulating skills. Development in grasp patterns take more time (complicating the use of spoons, crayons, etc.), and 'school skills' such as block building, pasting, colouring, handwriting and using scissors also appear to be delayed [33,34]. For the majority of children with VI, it is imperative to maximise the use of vision at an early age to promote optimal development across all domains of functioning[35]. A typical posture during uni-manual and writing tasks is characterised by greater flexion in the neck and a smaller distance between the eye and desk, while during bimanual tasks they often lift material closer to their face in order to shorten the working distance.

Children with VI have greater difficulties with the calibration of sensory information during the execution of fine motor tasks[36] such as handwriting[37]. Impoverished visual access to play materials and, often, little intrinsic motivation to explore small objects in children with VI may also limit their ability to train fine-motor skills[38,39]. Deficits in the motor performance of children with amblyopia but normal sight were greatest in manual dexterity tasks requiring both speed and accuracy, which was particularly seen in children with strabismus[40]. Caputo and colleagues (2007) used the MABC-I in a study with 4-6 year old children with congenital strabismus and normal visual acuity before surgery and found that they needed significantly more time, especially in uni- and bi-manual dexterity tasks, compared to children without strabismus[41]. Houwen and colleagues used three items of the MABC-I to compare 48 children with VI to 48 NS children of 7-10 years of age. Children with VI needed more time than their NS peers in uni-manual tasks and writing skills. Just as in gross motor skill development in general, no significant difference was found between children with moderate and severe VI, although children with moderate VI had better bi-manual coordination in the 7- 8 year age range, and better writing skills in the 7-10 year age range[18]. To summarise, the above mentioned studies confirm that children with VI generally have slower motor learning compared to NS peers, and this differs between tasks: they need more time, perform less accurately and use variable strategies to compensate for the lack of visual guidance.

**Measuring motor skill development in children with VI**

As mentioned before it seems to be important that motor development in children with VI is monitored in order to signal deviant development due to inaccurate stimulation. A suitable assessment instrument for children with VI, and reference scores specific to this group of children are therefore necessary. A recent systematic review[19] showed that many studies only used questionnaires to measure motor skills, or used parts of tests developed for NS children without adaptation for children with VI, or with only minor adaptation in the material or test procedures, such as more contrast in pictures or extra instructions. None of the tests used were investigated for validity and reliability in the specific population, as advised in test development[42]. Most of the studies included only a small number of children with VI (as a result of the low prevalence), and meta-analysis was impossible as a result of the large variety of instruments used. In a recent review[43] it appeared that most of the studies used existing norm-referenced tests, such as the Bruininks-Ozeretsky Test of Motor Proficiency (BOTMP), the Movement Assessment Battery for Children (MABC) or the Test of Gross Motor Development-2 (TMGD-2), questionnaires such as the Children's Physical Activity Form (CPAF) or specific tasks without norm references, to test motor development in children with VI. Most of these tests were used without adaptation. If adaptations were made, they focused on material or instruction to enlarge visibility, and sometimes only the items in instruments which were considered independent of visual input were used. Most studies adapted to missing reference values by using a group of NS children as a control group. The validity of conclusions was threatened in most studies by substantial inter-individual variation. We found only a few studies which aimed to test the psychometric qualities of existing motor tests adapted to children with VI. Recently the updated Bayley-III[25,26,44] has been adapted for children with low motor performance and low vision[45,46]. Comparable research had been done with the Dutch second edition of the Bayley-II[47,48]. In both tests the accommodations were focused on minimising impairment bias, without altering what the test measures. Pilot findings demonstrated that a subgroup of children benefits from the adapted version of the BSID-III[46], and a larger study found that the adaptations resulted in a higher score on the cognition scale, but not on the motor scale[45]. This test focuses on infants from the age of 0 to 48 months and the reference data was sampled in a mixed population including special needs children, which means that for children with VI between 4-11 years another instrument is needed. Recently the psychometric properties of the Test of Gross Motor Development-2 (TGMD-2) was tested in 75 children aged 6-12 years with VI[49]. Only adaptations in the colour and contrast of materials were made. The internal consistency of the TGMD-2 was high (alpha = 0.71-0.72) and the inter-rater, intra-rater, and test-retest reliability was good to excellent (ICCs ranging from 0.82 to 0.95). The influence of age and sex was confirmed by factor analysis. The authors concluded that the TGMD-2 is appropriate to assess gross motor skills of primary-school-age children with VI, but no reference data was presented as necessary for using such a test in clinical practice.

Taken altogether, it can be concluded that only a few instruments are available for the measurement of gross and fine motor skills in children with VI, and within them age ranges vary and for clinical practice norm references specific for children with VI are necessary.

**The present study**

In previous publications we have presented an instrument to test fine motor skills in children with VI, the ManuVis[3,50-52]. It appears that testing gross motor skills is more common in children with VI than testing fine motor skills in a standardised way, however, and therefore we decided to expand the research on the ManuVis to make it applicable in clinical practice. Such a test can be used to detect children with VI who are at risk of motor performance problems, and to monitor fine-motor skills over time, and test possible intervention effects in both clinical practice and research. The ManuVis (see Table 1 and Figure 1) is based on items adapted from existing valid test assessments: the Movement Assessment Battery for Children version I (MABC I;[27], and the General Movement Coordination Test (GMCT;[28]).The inter-rater reliability of the Movement ABC (Dutch version ranged from 0.95 to 1.00[53]. The test-retest reliability was examined in young children (4-5 years) and this varied from 0.88 to 0.98[54,55]. Test-retest reliability from the General Movement Coordination Test was studied in a group of 45 children and was 0.79 for the manual dexterity (GMCT;[28]).

Adaptations of the ManuVis are not focused on increased visibility but allow a child to adapt to their impaired vision using other sensory information. The earlier version of the ManuVis only included children of 6-11 years. We decided to focus on younger age groups (4-6 years) as well, because children need fine motor skills in their younger years before they go to school. Early detection of possible deviations in the typical development of fine motor skills in children with VI seems to be necessary especially at school age.

To summarise: the aim of this study is to obtain insight into differences in fine motor skills between children with VI and NS children, over different age ranges, to provide norm reference scores for the ManuVis for children between 4-11 years, and to investigate the test–retest reliability and inter-rater reliability of the ManuVis.

**Figure 1.** **Picture of all materials used in the ManuVis for all six items: (1) money box, (2) wooden board with twelve rings, (3) nuts with bolt, (4) beads with cord, (5) board with lace, (6) paper for the writing task and (7) open container.**

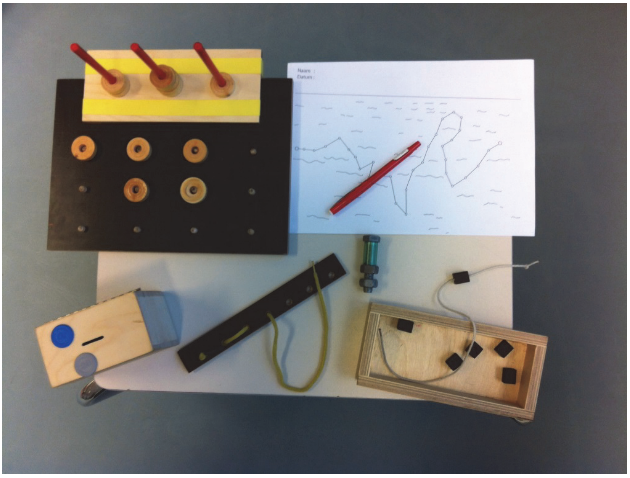


**Table 1. Description of all six items and corresponding procedures of the ManuVis[3] compared to the original Movement ABC (MABC) 27 and General Movement Coordination Test (GMCT)[28]**

| |
|---|
| *One-handed skills* |
| **1. Putting coins in a money box** |
| With the "*Putting coins in a money box*" item, the target position for the coins is known because the other hand feels the position and can remain there to provide a reference point. The 10 plastic coins are in an open container, so that their starting position is relatively fixed. The box is with the long side to the child. The task is carried out with both left and right hand, starting with the child's preferred hand. Start timing when the hand is lifted to move. The scores in seconds from both hands are totalized. *In the MABC: there are 12 coins and they are arranged in four horizontal rows of three coins. The box is with the short side to the child. This is only used in age band 4 - 6 years.* |
| **2. Putting rings on rods** |
| This item introduces an additional spatial factor: changing starting and target positions. Twelve wooden rings must be placed on three vertical round rods. The 12 rings are initially in a fixed position on a wooden board in three straight rows of four rings in front of the rods. The task is to place the first ring on the first rod, the next ring on the next rod and so on. The rings may be picked up in any order. The task is performed with both the left and right hand, starting with the child's preferred hand. Start timing when the hand is lifted to move. The scores in seconds from both hands are totalized. If there is a sequence error during the performance then there is made a correction in seconds. Per ring a mean score in seconds is calculated by dividing the total score in seconds by 24 (12 rings per hand) and multiplied by 24 + 2 per error. *In the GMCT 24 rings are used for each hand, which are located on a wooden board in four rows of six, they are not fixed.* |
| *Two-handed skills* |
| **3. Screwing nuts onto a bolt** |
| This item includes an additional manual dexterity factor. Two nuts must be placed on the bolt using a sort of counter-movement and then screw on using fingers and thumb. In terms of positioning objects in space, this task is easier because the child starts with a nut in one hand and the bolt in the other and these must then be brought together. The nuts are placed in open container and the bolt is in front of the child behind the container. Start timing when the hands are lifted to move. |

*In the MABC three nuts are used for age band 9-10 years. The nuts are placed in a horizontal row at right angle to the bolt.*

**4. Threading beads**

Six octagonal beads in an open container must be threaded onto a piece of cord. One hand manipulates the bead and feels for the hole and the other hand threads the cord through. The length and the width have a different size. The holes are always in the short side of the beads. Start timing when the hands are lifted to move.

*In the MABC there are six beads for 4-years old children and 12 beads for 5-6 years old children. The length and width are the same. The beads, with the holes facing up, are in a row on the table-top mat.*

**5. Threading lace**

This item requires greater understanding of the action to be performed. The children must be able to form a mental image of what they are doing. The lace comes out of the back of the board and must be threaded in again from the same (back) side. When the lace comes through to the front, it must be threaded back in again from the front (i.e. there must be no loops in the cord over the edge of the board). There are six holes. Start timing when the hands are lifted to move.

If the children make a mistake, movement time is corrected. For example the child makes one noose, then there is an addition in seconds. Per hole a mean score is calculated by dividing the total scores in seconds by 6 and multiplied by 6 + 2 per error.

*In the GMCT the board is with 12 holes, 6 holes at one side and two laces.*

*Pre-writing-task*

**6. Drawing dots**

The last manual dexterity item adds an extra component for visually impaired children. The item uses dots placed in circles to measure eye-hand coordination. Only one pre-writing task is executed (the simple one with omission of the background drawings on the paper). The task shows the relative degree of success and difficulty of eye-hand coordination and which effect vision has on accuracy and posture. The tester can also observe how the pen is held and used. The child is asked to work as quickly and accurate as possible. Start timing when the first dot is put in. There is an addition in seconds if dots are placed inaccurately: the score is a combination of time and accuracy. Penalty seconds are calculated as follows:

1 error = dot over the edge of a circle; 2 errors = a circle missed or a dot next to a circle.

| n errors | n penalty sec. | n errors | n penalty sec. |
|---|---|---|---|
| 0 | 0 | 15-20 | 5 |
| 1-2 | 1 | 21-27 | 10 |
| 3-5 | 2 | 28-35 | 15 |
| 6-9 | 3 | 36-44 | 22 |
| 10-14 | 4 | > 44 | 30 |

*In the GMCT there were two writing tasks: placing dots with different accuracy. Scoring: total time and an addition in seconds if dots are placed inaccurate for correction*

Procedure: After a first training attempt, all items are performed once, in order to keep the test time in twenty minutes. The tasks were repeated if there was an execution error.



## METHODS

### Study design

To expand the age range of the existing ManuVis and to enlarge the reference population this prospective cohort study was set up which was embedded in a larger study focusing on intervention strategies for children with VI. Data from baseline measurements was used for this study. The entire research study was approved by the Medical Ethics Committee (CMO 2010/037 Arnhem Nijmegen/NTR=2494) and was conducted in accordance with the Declaration of Helsinki. After written invitation, all parents of the participants gave written informed consent.

**Participants**

Children with VI were selected from the databases of institutions for visual rehabilitation in the Netherlands. WHO guidelines were used to classify visual impairment[4,56]. The inclusion criteria for children with VI in this study were adopted from the rehabilitation protocol of Bartiméus: visual acuity of at least 0.05 and ≤ 0.3, no comorbidity and/or cognitive impairments; and birth at term (i.e. ≥36 weeks of gestation) with normal birth weight. Blind children were not included in this study. The eye disorders underlying the visual impairment in the participants are described in detail in Table 2.

**Table 2. Eye disorder categories for all children with visual impairment**

| | | |
|---|---|---|
| 1 | Albinism | 70 |
| 2 | Congenital nystagmus | 41 |
| 3 | Retinal dystrophy | 29 |
| 4 | Cataract | 27 |
| 5 | CNSB | 13 |
| 6 | Aniridia | 11 |
| 7 | Cone- rod dystrophy | 9 |
| 8 | Optic nerve defects: optic nerve atrophy | 7 |
| 9 | Hyperopia (> 4D) | 7 |
| 10 | Congenital glaucoom | 6 |
| 11 | Myopia (>S -7) | 6 |
| 12 | Achromatopsia | 5 |
| 13 | Coloboma | 4 |
| 14 | Congenital optic nerve defects / atrophy | 4 |
| 15 | Microphthalmia | 3 |
| 16 | Stargardt disease | 1 |
| 17 | Macular hypoplasia | 1 |
| 18 | Peters anomaly | 1 |
| 19 | Remaining group / diagnosis unknown | 11 |
| | TOTAL | 256 |

The NS children in the control group attended regular schools in the neighbourhood of the Dutch city of Utrecht. None of the children had known cognitive or physical impairments. In total 164 children with VI and 91 NS children in the age groups of 4-11 years participated and were assessed for this study. To increase the sample, and because preliminary analysis (see Section 3.1) revealed minimal to no differences between new participants and participants in the initial ManuVis study in 2003[3] participants in the initial study, 92 children with VI and 71 NS children, were also included for analysis in the present study. The total sample consisted of 256 children with VI and 162 NS children. The characteristics of all children are presented in Table 3.

**Table 3. Age and sex of all children with normal sight (NS) and visual impairment (VI), between brackets in cursive the number of children already included in the initial ManuVis study 3**

| | Age in years | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|
| | **4** | **5** | **6** | **7** | **8** | **9** | **10** | **11** | **new/already included** | **Total** |
| | **Normal Sight** | | | | | | | | | |
| Boys (n) | 5 | 7 | 11 (*10*) | 17 (*3*) | 19 (*6*) | 11 (*11*) | 6 *(6)* | 2*(2)* | 40 /38 | 78 |
| Girls (n) | 5 | 9 | 20 (*4*) | 17 (*6*) | 13 (*5*) | 9 (*7*) | 5 *(5)* | 6*(6)* | 51/ 33 | 84 |
| Total (n) | 10 | 16 | 31 (*14*) | 34 (*9*) | 32 (*11*) | 20 (*18*) | 11 *(11)* | 8*(8)* | 91/ 71 | 162 |
| | **Visual Impairment** | | | | | | | | | |
| Boys (n) | 27 | 32 | 37 (*11*) | 20 (*10*) | 23 (*9*) | 21 (*13*) | 9 *(9)* | 11*(11)* | 117/ 63 | 180 |
| Girls (n) | 7 | 19 | 18 (*4*) | 7 (*4*) | 9 (*6*) | 8 (*7*) | 6 *(6)* | 2*(2)* | 47/ 29 | 76 |
| Total (n) | 34 | 51 | 55 (*15*) | 27 (*14*) | 32 (*15*) | 29 (*20*) | 15 *(15)* | 13*(13)* | 164/ 92 | 256 |

In order to determine the test-retest reliability of the ManuVis, 19 children with VI (20 were invited, but one child was only tested once) between 4 and 7 years old, were tested twice, at a one-week interval by the same tester (BH). The scores from administration of the first test were added to the dataset. For inter-rater reliability, the performance of 20 children between 4 and 8 years old (VI: *n*=15; NS: *n*=5) was directly scored by two testers (JL / RC) during the test, and the other (AR) was scored later from video recordings.

3

**Material & Procedure**

***Development and content of the ManuVis***

For NS children, fine-motor skills are generally tested with the Movement Assessment Battery for Children (MABC). The first version of MABC-I was introduced in 1992,[27] and in 2007 a second version was presented (MABC II;[57]. The MABC is divided into four age bands, each containing eight test items classified into three sections testing manual dexterity (3 items), ball skills (2 items), and balance control in static and dynamic conditions (3 items). The items are comparable over age bands but differ in task difficulty (e.g. putting coins in a box increases in difficulty to putting pins into a small hole). Fine motor skills are tested using a uni-manual, a bimanual and a prewriting task. To increase validity and to enable comparison over age ranges we decided to use the test items of the ManuVis[3,51] with the same instructions and procedures in all age groups, to avoid an increasing load on visual perception which would interfere with testing motor capacity in children with VI. We were aware of possible floor effects, but we judged the advantage of the comparability of task performance in different age groups in this specific group with VI more important. Test items and materials were selected from the MABC I and we added three items from the GMCT[28]. This test of ten items was developed to test both gross and fine motor performance in the age range of 6-10 years. The test items were adapted for children with VI in such a way that they could use sensory information to increase their performance. The materials are shown in Figure 1, and in Table 1 the six ManuVis items are described, including the procedures and adaptations compared to the original items.

### *Test items of the ManuVis*

The ManuVis contains six fine-motor items; two items for testing uni-manual tasks, three items for testing bi-manual tasks, and one item to test visual-motor integration using a pre-writing task. For all items performance time was scored in seconds, whereas performance time was defined as the time elapsed between the moment the hand is lifted and the end of the hand's final movement. In three items the item score was based on performance time and the addition of compensation seconds conform the manual (see Table 1): for the items *putting rings on rods* and *threading lace* in case of procedural errors, for the 'pre-writing task' in case of accuracy errors.

### *Test procedure*

The children were instructed to work as quickly as possible. To reduce the total time of testing for the children, we choose to use each item only once, if it was sufficiently performed. If a procedural mistake or breakdown situation was observed, the tester interrupted the attempt as quickly as possible and gave corrective instructions and/or demonstrations, after which the child started again. Total testing of the new group took between 15 and 20 minutes, which was comparable to the initial study.

## Data Analysis

For each individual child the total score for each of the five items was noted in seconds, all item scores are summarised as a *total score 1-5* (in seconds). There was a missing item for one child, for which we imputed the adequate item mean score (based on group and age), in order to be able to compute a *total score 1-5* for this child. Scores for the pre-writing task were analysed separately because the item score was based on performance time and errors.

To test possible differences between the new sampled data and data from the initial ManuVis study, (see Table 3), we performed Univariate ANOVAs on all six item scores and *total score 1-5* separately for both groups (children with VI and NS children) and all age groups.

To answer the research questions univariate ANOVAs were conducted to test the differences between VI and NS groups and age groups (4 -11 years) for each item and the *total score 1-5*. In cases of a significant age effect, Bonferroni post-hoc analyses were performed, to investigate the differences between age groups, in children with VI and NS children separately.

Test-retest and inter-rater reliability were calculated using intra-class correlation coefficients (ICCs) and standard error of measurement (SEMs) for each item and the total score. ICCs were interpreted using the following criteria: 0.00– 0.49 poor; 0.50–

0.74 moderate and 0.75–1.00 excellent[58]. For each ICC obtained, a 95% confidence interval (CI) was calculated to provide a range of values that was likely to cover the true population value. The SEM describes the error in interpreting an individual's test score. The SEM allows for estimation of the 'true' test performance using a reliability coefficient, and is computed by the standard deviation of the *scores* multiplied by the square root of one minus its reliability coefficient [$SEM = SD \times \surd 1 - ICC$][59]. As a criterion for acceptable precision of the SEM, a value SD/2 was used[60]. All data was analysed in the statistical programme IBM SPSS (Version 21.0). A significance level (alpha) of .05 was applied throughout.

## RESULTS

### Preliminary analysis

Preliminary analysis of the similarity of the datasets of the initial ManuVis study and the new data for the children between 6 and 9 years revealed no significant differences in scores for nearly all items. Only for the children with VI, at the age of 7 years, did the item *Drawing dots* differ significantly ($t(31) = 7.84$, $p = .01$; Initial group ($n$=14): $M$ = 52.5s, $SD$ = 23s; new group ($n$=13): $M$ = 43.7s, $SD$ = 7.4s. At the age of 9 years a significant difference was found in the item *Putting rings on rods* ($t(27) = 5.9$, $p = .01$; initial group ($n$ = 20): $M$ = 38.6s, $SD$ = 7.5s; new group ($n$ = 9): $M$ = 36s, $SD$ = 3.5s. No significant differences were found for the *total score 1-5* (data not shown). Given these results, all further analyses were based on the combined dataset.

3

### Descriptives

For the purpose of clinical applications the mean scores and the scores belonging to both the 15th percentile and the 5th percentile are presented per test item and the age band for the VI and NS group separately in Table 4. No value for the 5th percentile is reported when either the group size was too low or the difference between the 5th and 15th percentiles was too small.

In both two items *putting rings on rods* and *threading lace* we found procedural errors leading to the addition of compensation seconds conform the manual (see Table 1). For *putting rings on rods,* this only occurred in one NS child of 5 years old, and in five children with VI (four children of 4 years and one of 5 years old). For *threading lace,* this occurred in three children with VI: one who was four years and two who were five years old.

**Table 4. Mean scores (M) in seconds, the 15th percentile (p15) and 5th percentile (p5) per test item per age group for normally sighted (NS) and visually impaired (VI) children**

| Age (years) | | 4 | | | 5 | | | 6 | | | 7 | | | 8 | | | 9 | | | 10 | | 11 | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Number of children | NS | 10 | | | 16 | | | 31 | | | 34 | | | 32 | | | 20 | | | 11 | | 8 | |
| | VI | 34 | | | 51 | | | 55 | | | 27 | | | 32 | | | 29 | | | 15 | | 13 | |
| | | *M* | *P15* | *P5* | *M* | *P15* | *p5* | *M* | *P15* | *P5* | *M* | *P15* | *P5* | *M* | p15 | *P5* | *M* | *P15* | *P5* | *M* | *P15* | *M;* | *P15* |
| 1. coins | NS | 47 | 54 | na | 43 | 53 | na | 34 | 38 | 41 | 34 | 3 | 42 | 32 | 37 | 40 | 29 | 36 | 36 | 28 | 32 | 26 | 29 |
| | VI | 54 | 67 | 77 | 47 | 54 | 71 | 44 | 52 | 63 | 39 | 49 | 56 | 36 | 41 | 46 | 37 | 43 | 67 | 33 | 38 | 32 | 39 |
| 2. rings | NS | 60 | 80 | na | 52 | 66 | na | 42 | 47 | 53 | 38 | 45 | 50 | 35 | 40 | 52 | 33 | 39 | 41 | 29 | 32 | 28 | 32 |
| | VI | 77 | 97 | 149 | 57 | 72 | 85 | 52 | 66 | 76 | 44 | 52 | 60 | 41 | 50 | 54 | 38 | 45 | 53 | 36 | 43 | 32 | 36 |
| 3. nuts | NS | 50 | 70 | na | 45 | 68 | na | 33 | 43 | 43 | 30 | 40 | 48 | 23 | 29 | 38 | 22 | 27 | 35 | 16 | 23 | 17 | 23 |
| | VI | 64 | 82 | 120 | 51 | 75 | 89 | 43 | 61 | 66 | 35 | 54 | 69 | 29 | 38 | 48 | 29 | 41 | 47 | 32 | 39 | 24 | 29 |
| 4. beads | NS | 48 | 63 | na | 41 | 50 | na | 32 | 38 | 43 | 31 | 37 | 50 | 27 | 35 | 42 | 26 | 32 | 37 | 21 | 24 | 20 | 25 |
| | VI | 82 | 137 | 166 | 57 | 71 | 102 | 48 | 59 | 83 | 39 | 51 | 74 | 35 | 50 | 55 | 30 | 36 | 56 | 28 | 34 | 28 | 32 |
| 5. lace | NS | 51 | 72 | na | 39 | 52 | na | 30 | 36 | 59 | 29 | 36 | 50 | 24 | 33 | 50 | 21 | 28 | 30 | 15 | 20 | 17 | 24 |
| | VI | 76 | 113 | 216 | 54 | 67 | 132 | 43 | 55 | 70 | 35 | 51 | 69 | 30 | 37 | 59 | 24 | 30 | 37 | 27 | 31 | 22 | 33 |
| 6. dots | NS | 58 | 76 | na | 54 | 64 | na | 41 | 50 | 58 | 35 | 44 | 48 | 30 | 35 | 39 | 29 | 32 | 40 | 23 | 26 | 23 | 28 |
| | VI | 96 | 131 | 201 | 70 | 91 | 114 | 60 | 74 | 112 | 48 | 61 | 102 | 43 | 55 | 74 | 36 | 41 | 68 | 35 | 39 | 38 | 46 |
| 7. Total score | NS | 257 | 296 | na | 218 | 263 | na | 172 | 202 | 225 | 162 | 186 | 227 | 142 | 157 | 212 | 131 | 150 | 154 | 109 | 126 | 108 | 129 |
| 1-5 | VI | 353 | 508 | 575 | 264 | 318 | 371 | 231 | 287 | 310 | 192 | 251 | 297 | 171 | 198 | 241 | 158 | 183 | 237 | 155 | 163 | 137 | 169 |

na: not applicable

### Effects of visual impairment and age

Results of the univariate ANOVAs on each of the six item scores, and the *total score 1-5* are presented per age band for the children with VI and NS in Table 5 and Figure 2. For all items we found significant main effects for between group differences (children with VI needed consistently more time) and for age (time needed to perform the tasks decreased with increasing age). Post-hoc analysis (Bonferroni) revealed significant differences between VI and NS children in each age group for *threading beads*, *drawing dots* and for the *total score 1-5.*

**Table 5. Results of Uni-variate ANOVA's to test differences in item scores and Total score 1-5 between Groups (Visual Impaired and Normal Sighted) and between Age categories in years (4-11 years)**

| Item | Group df: (1, 417) | Age (years) df: (7,417) | Group * Age df: (7,417) |
|---|---|---|---|
| 1. Putting coins in moneybox | *F* = 49,64; *p* = .00 | *F* = 33.37; *p* = .00 | not significant |
| 2. Putting rings on rods | *F* = 30.07; *p* = .00 | *F* = 38.49; *p*= .00 | not significant |
| 3. Screwing nuts onto a bolt | *F* = 32.84; *p* = .00 | *F* = 31.34; *p* = .00 | not significant |
| 4. Threading beads | *F* = 50.15; *p* = .00 | *F* = 25.16; *p* = .00 | *F* = 3.74; *p* = .003 |
| 5. Threading lace | *F* = 21.14; *p* = .00 | *F* = 18.59; *p* = .00 | not significant |
| 6. Drawing dots | *F* = 59.91; *p* = .00 | *F* = 28.93; *p* = .00 | *F* = 2.13; *p* = .040 |
| 7. Total score 1-5 | *F* = 66.41; *p* = .00 | *F* = 54.65; *p* = .00 | *F* = 2.26; *p* = .029 |

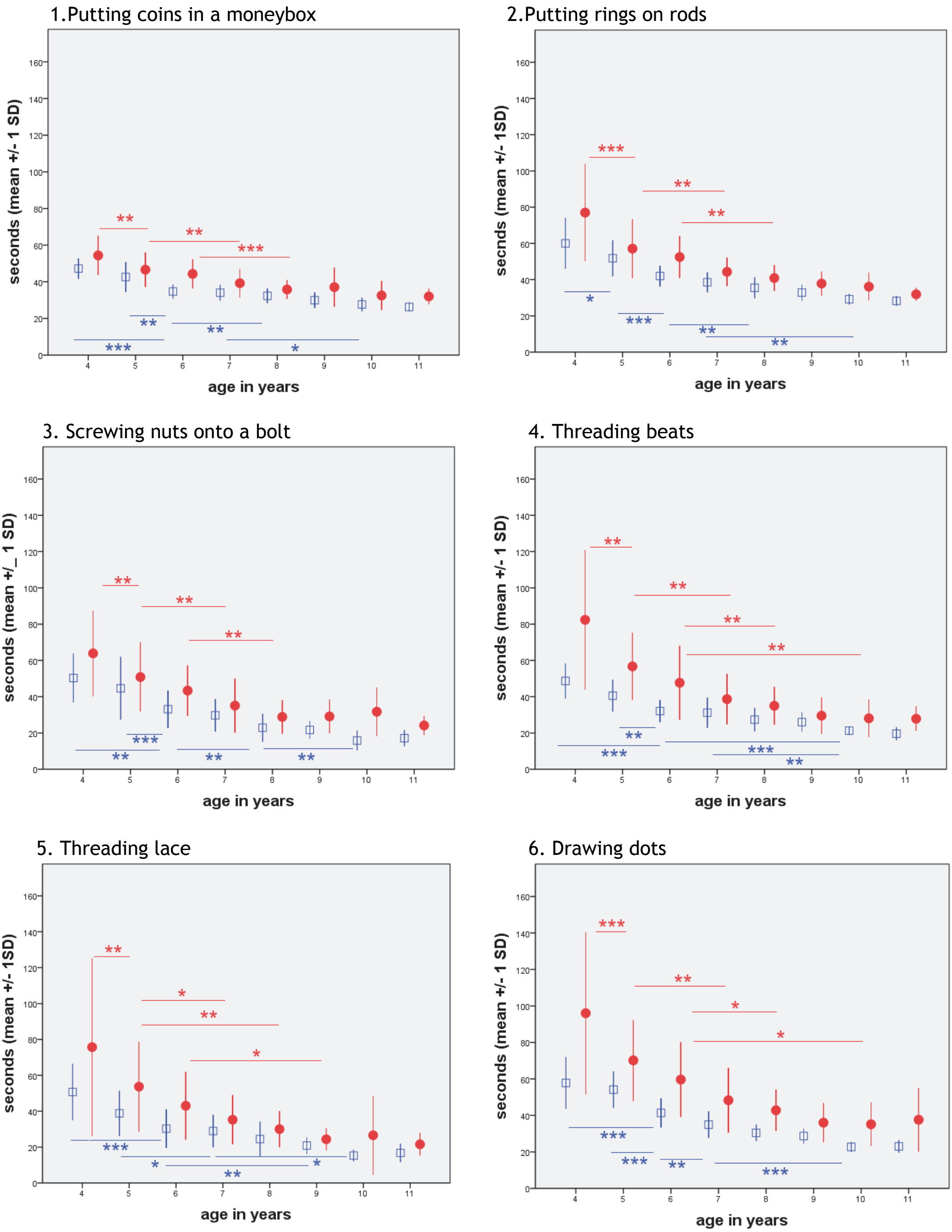
In both groups for all items, the time needed to perform the tasks decreased with increasing age up to 10 years. As can be seen in Figure 2, performance time does not decrease further (floor effect) in most items and in the *total score 1-5* for children of 10 and 11 years. Children with VI needed significantly more time for all tasks in each age group than did NS children. Figure 2 shows that there are also significant differences between age groups in all item scores and *total score 1-5*. In children with VI, especially between 4 and 5 years and between 5 and 7 years, performance time decreases, while in the NS group this is earlier between 4-6 years and 5 and 6 years. Differences between the VI group and NS group are largest in the younger age groups.

There were also significant Group*Age interaction effects for *threading beads*, *drawing dots* and for the *total score 1-5.* (see Table 5), showing that the decrease in performance time as a function of age was at a higher age in children with VI compared to NS children. Although the age-related progress in fine-motor skill performance in children with VI seems slower, it proceeds over a longer time but does not reach the normal level, at least not by the age of 11 years.

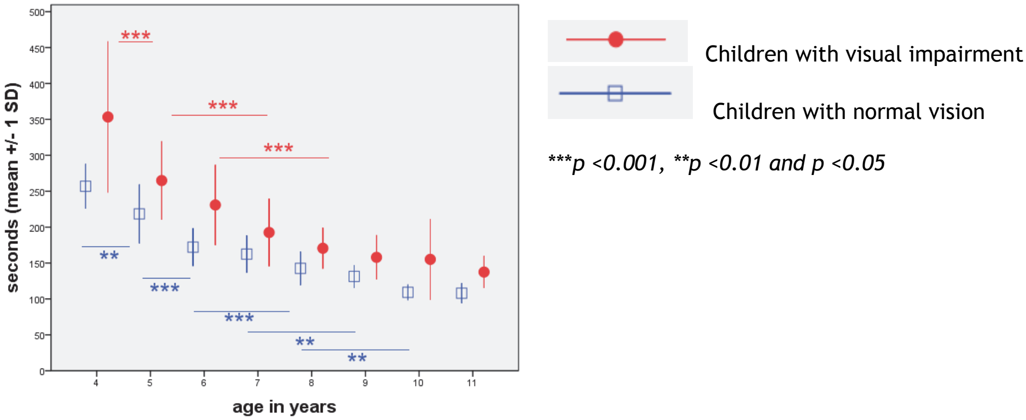
### Reliability of the ManuVis

The ICC scores of the test-retest are moderate to excellent (see Table 6). SEMs for the test-retest reliability were between 0.52 and 1.26 and all values are below the cut-off standard deviation of 2. For the inter-rater reliability ICC was excellent and the SEM below .007. It can be concluded that ManuVis reliability is sufficient.

**Figure 2. Mean (M) and standard deviation (SD) in seconds of the scores per group for each item and for Total score 1-5**



7. Total score 1-5




3

**Table 6. Results of the analyses for test - retest and inter-rater reliability with the Intra-class Correlation Coefficients (ICC) value per test item and for the Total score 1-5**

| | Test - retest reliability | | | Inter- rater reliability | | |
|---|---|---|---|---|---|---|
| | ICC | CI (95%) | SEM | ICC | CI (95%) | SEM |
| 1. Putting coins | 0.58 | 0.18 - 0.81 | 1.13 | 0.98 | 0.98 - 0.99 | 0.03 |
| 2. Placing rings | 0.74 | 0.44 - 0.89 | 0.91 | 1.00 | 0.99 - 1.00 | 0 |
| 3. Nuts onto bolt | 0.67 | 0.32 - 0.86 | 1.26 | 1.00 | 0.99 - 1.00 | 0 |
| 4. Threading beads | 0.79 | 0.53 - 0.91 | 0.83 | 1.00 | 0.99 - 1.00 | 0 |
| 5. Threading lace | 0.72 | 0.40 - 0.88 | 1.16 | 0.99 | 0.99 - 1.00 | 0 |
| 6. Drawing dots | 0.89 | 0.74 - 0.96 | 0.52 | 0.99 | 0.99 - 0.99 | 0.07 |
| 7. Total score 1-5 | 0.85 | 0.66 - 0.94 | 1.01 | 1.00 | 0.99 - 1.00 | 0 |

ICC= Intra-class Correlation Coefficients, CI = 95 % Confidence interval, SEM= Standard Error of Measurements

## DISCUSSION

This study aimed to obtain insight into fine motor skills typically for children with VI compared to those of their NS peers. We aimed to provide ManuVis reference norms for use in clinical practice for children between 4-11 years. We also wanted to investigate the test-retest and inter-rater-reliability of the ManuVis.

This study shows that children with VI need more time to perform fine-motor tasks and have a prolonged period of fine-motor skill learning compared to their NS peers. The differences between groups were largest in 4 and 5 year old children, demonstrating that children with VI apparently need more time to learn adaptation strategies in order to perform fine-motor skills in an adequate way. Inter-individual variability within the VI group was much higher than in the NS group, however, it was clear that in both groups of children (VI and NS), a learning-effect could be observed as performance time decreased with increasing age although the slope in the curves in Figure 2 differs for the different tasks. In Task 1 (*putting coins into a box*) the curve is relatively flat, and the difference between VI and NS children is relatively constant over age groups. Apparently in this task sensory adaptation is already adequate at a younger age and the decrease in seconds is related to motor learning in both groups. This is different for the more complex task in Item 2 (*placing rings*), although as in Task 1, an uni-manual task, the increase in accuracy load results in a steeper curve. The same is seen in the difference between the bi-manual tasks: the curve in Item 3 (*screwing nuts onto a bolt*) is less steep than in Items 4 and 5 (*threading beads or lace*): in these tasks Item 3 is also more guided by the material than in Items 4 and 5, in which the child needed to search for strategies and also use sensory strategies. The time needed for Item 6 (*drawing dots*) does not decrease consistently in children with VI between 9, 10 and 11 years in contrast to normal sighted peers. This task is most influenced by visual input. The inter-rater reliability of the ManuVis was excellent, indicating that test procedures are well described. The ICC score for test-retest reliability were sufficient for *putting coins* and *screwing nuts onto a bolt*, whereas the ICC scores for *threading beads*, *drawing dots* and the *total score 1-5* were excellent. The results of the reliability study demonstrate that the measurement procedures of the ManuVis are adequate. All children understood the task instructions, only in the youngest children were a small number of procedural problems reported, which can be pre-empted by an extra trial during testing (not one retry but two).

As mentioned before it seems to be important to monitor motor development in children with VI so as to be able to signal deviant development due to inaccurate stimulation. The results of this study demonstrate that the ManuVis is a suitable, reliable and valid measurement instrument to test the fine motor skills of children with VI at the age of 4-11 years. The presented norm and reference scores specified for this group of children with VI make the ManuVis useful in clinical practice and research. The only other comparable instrument focusing on fine motor skills is the recently updated Bayley-III[44,61] adapted for children with low motor performance and low vision[45] or the previous version of the Dutch second edition of the Bayley – II[47,48]. These instruments focus on younger children aged 0-48 months, however, and no norm references are yet available for use in individual testing. It is advisable to use the ManuVis in combination with a valid, reliable and norm-referenced test for gross motor performance, however based on a recent review[43] it appears that only a few studies aimed to test the psychometric qualities of existing motor tests adapted to children with VI. Concerning gross motor skills, the psychometric properties of the Test of Gross Motor Development-2 (TGMD-2) adapted for children aged 6-12 years with visual impairments (VI), showed high internal consistency and good to excellent reliability[49]. The influence of age was confirmed in a factor analysis. In a recent study[16] of the TMGD-2[24], the influence of age on test outcomes was not confirmed, which means that the test is not sensitive to changes in coordination which gradually improves with increasing age based on development and learning[24-28,57,61],which would be expected in children with VI. More research in larger groups is necessary to test whether the TGMD-2 is appropriate to assess the gross motor skills of primary school age children with VI and to sample the norm-referenced data necessary for use in decision-making in clinical practice.

As can be seen in the results, inter-individual variation in performance is much greater in the VI group compared to the NS group, especially in the younger age groups and in the more complex tasks. This is in agreement with the literature[10]. This variation can be related to differences in visual input or differences in motor learning. Taking into account that perceptual systems and motor systems become coupled into task-specific functional units[5,6], and motor and perceptual development are strongly intertwined[7,62], the most important point for a child with VI is learning to adapt to the inborn visual constraints by increasing sensory-motor experiences as much as possible. Research has already pointed out that even when children with VI may have adequate acuity for certain activities, they may not be able to deploy and interpret visual input in the same functional way as NS children, based on fewer experiences, stimulation and learning opportunities compared to NS children[63]. Only weak evidence is found for a relationship between the degree of VI and gross motor development and manual dexterity[10] and this means that, at least partly, variability between children is caused by differences in experiences and motor learning. Especially in learning fine-motor skills, exploration, imitation, practice, and the refinement of skills play an important role. The results of the study demonstrate that variability between children is high, even in older children, which is possibly merely determined by differences in learning experiences rather than being due to differences in visual acuity. It is assumed that variability is related to difficulties with the calibration of sensory information during execution of a fine motor task[21,50] especially seen in the results of more complex tasks such as handwriting[37]. We expected that variability in motor performance would decrease in the older age groups as result of motor learning, which is observed in NS children, but to a lesser extent in children with VI. Although we cannot rule out that the relatively small group size, especially in the older age groups, was one of the reasons, we conclude that differences in experience also play a role. Because of the low prevalence of VI, we advise sampling data during clinical practice and updating reference data regularly.

The younger group children with VI (4-6 years) were proportionally slower in performing all items. Typically, in younger children the visual system (or gaze) provides information on shape, size, colour, distance, location and movement velocity, and the direction of objects and people in the environment, all in one glance. Therefore, particular in this age is that they need to learn to focus attention on relevant cues, to detect information about distance and direction of movements and objects[12,13] in order to control movement in an adequate way. They need to learn to use different sensory information systems and need to repeat skills often enough to automate the movements. That such possibilities to use sensory information increase motor performance can be seen in Figure 2: the differences between VI and NS groups are smaller in tasks with a fixed start and end-position in which children can rely on sensory information to reiterate the skill. For example, there is a difference between the task *screwing nuts onto a bolt,* where visual attention is only needed at the start, and the task *threading beads and lace* where visual information is needed for cord insertion (see Figure 2). Visual information is needed for the pre-writing task (*drawing dots*) from the start to the end. In more complex tasks with fewer predefined start and target positions, children with VI learn less from imitation and need to find strategies themselves to use adequate sensory information that can help them to be faster and more accurate in task performance. The pre-writing task was the most difficult for all children (VI and NS) and both accuracy and speed increased until the age of 11 years. It is obvious that in this task the primary role of vision, namely the visual-spatial perception of the target position and the feedback during the execution of movements inclusive of feedback on the results, is greatest, with less possibilities to use other sensory strategies.

We made choices in the development of the ManuVis. As in other studies we started with a selection of items in already existing valid, reliable and norm-referenced tests, however, concerning adaptations to children with VI, we made choices that were comparable to other authors, like increasing contrast, using adequate coloured material or choosing items which were considered independent of visual input. But we also used adaptations that enabled the child to use sensory strategies in task performance, because we hypothesised that this is what you expect a child to do in motor activities in daily life. Another important choice was to use the same items for all age groups, and for both the VI and NS groups. This can be seen as a strength of this study (and the ManuVis) but also a limitation. In most instruments, such as the MABC, task difficulty increases in higher age bands. Mostly task difficulty is manipulated by increasing accuracy load. NS children use visual input for goal detection, and feedback during motor action to fulfil the task as adequately as possible. Because the measured performance is determined by both the visual information processing and the motor action, we wanted to stabilise the visual input to be able to focus on the motor performance measurement. Based on our results we can conclude that using the same task was adequate for detecting differences between children with VI and NS children, and was sensitive enough to find differences in age groups, at least for the children with VI. Using such a standardised measurement procedure for all age groups allowed insight in the age effect on changes in motor performance.

On the other hand, a weakness of this choice in the ManuVis is the presence of some floor effects, more common in the results of the NS children and especially seen in the item *putting coins into a moneybox*, but also observable in other tasks starting at different ages. As can be seen in Table 4 however, only in a few items in the older age groups did the mean values and the values of the p15 and p5 show a small difference.

Accordingly we judge that the ManuVis as suitable for children with VI up to 9 years old to determine possible motor skill problems, while for intra-individual evaluation the test can be used until 11 years. For future use of the ManuVis we advise an increase of task difficulty in at least some of the test items for children of 10 and 11 years, and possibly older.
The extended norm scores for children with VI are appropriate for monitoring the development of children with VI, however, in most tests of fine motor skills the quality of movement is also tested. Because the visual system (or gaze) plays such an important role in fine-motor skill performance, children with VI search for alternative solutions to perform the task at hand, but not always in the most effective or efficient way. For example, by changing their posture, and reducing the viewing distance, sometimes with a distance of only a few centimetres between their squinting eyes and the objects in their hands, action control and movement fluency are hindered significantly. This also means that qualitative information about the way movements are performed needs to be described in the scores in seconds as important information for coaching and intervention. As can be seen in the results, the difference when compared with NS children increases when the role of visual control in the end state of the movement increases, for instance in the pre-writing task *drawing dots*. This phenomena is especially present in younger children who need to learn how to control their movements but lack the opportunity to learn by imitation. In children with VI who need to learn to make use of compensatory strategies, it is therefore important to teach them adequate strategies such as optimising viewing distance, relying more on proprioceptive and sensory information, and minimising the influence of nystagmus by holding the head in a tilted and/or rotated way, often called torticollis[51]. In addition attention must be given to adequate lighting and contrast of material.

For future research a longitudinal research design monitoring development of fine motor skills in individual children with VI is advisable, in order to gain more insight into individual developmental trajectories and the influencing factors. Also in such a set-up the research on fine motor skills can be combined with that of gross motor skills. It is clear that working together in international networks, where research groups work together, helps to obtain more reliable data on patient groups where there is low prevalence of visual impairment in children.

## CONCLUSIONS

It can be concluded that the ManuVis is a reliable and valid instrument with which to measure fine motor skills in children with VI. It can be used in clinical practice to compare the individual performance of a child with VI to their age-related peers with VI without co-morbidities. This instrument is not reliable for blind children. The youngest children with VI in particular need more time to perform fine motor skills compared to their NS peers, while the differences decrease with age but not completely diminish. It is clear that children with VI learn to make use of compensatory strategies, however these strategies take time. Differences are greater in complex uni-manual and bi-manual tasks, especially if visual information on the end state of the movement plays an important role. The results of this study, and those of other studies, suggest that inter-individual variability in performance is at least partly due to differences in experience and motor learning. This would mean that early detection and intervention, including instructions to people in the environment, are needed. The availability of this specific instrument can help in the planning and evaluation of preventive intervention strategies for children with VI. The prevention of fine motor problems can facilitate a normal start in school and can enhance the social participation of children with VI.

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# Chapter 4

## Development of an Age Band on the ManuVis for 3-Year-Old Children with Visual Impairments

Annemieke Reimer
Annemiek Barsingerhorn
Anneloes Overvelde
Ria Nijhuis-van der Sanden
Nienke Boonstra
Ralf Cox


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**ABSTRACT**

*Aim*: To compare fine motor performance of 3-year-old children with visual impairment with peers having normal vision, to provide reference scores for 3-year-old children with visual impairment on the ManuVis, and to assess inter-rater reliability.

*Method*: 26 children with visual impairment (mean age: 3 years 7 months (SD 3 months); 17 boys) and 28 children with normal vision (mean age: 3 years 7 months (SD 4 months); 14 boys) participated in the study. The ManuVis age band for 3-year-old children comprised two one-handed tasks, two two-handed tasks, and a pre-writing task. *Results*: Children with visual impairment needed more time on all tasks ($p < .01$) and performed the pre-writing task less accurately than children with normal vision ($p < .001$). Children aged 42–47 months performed significantly faster on two tasks and had better total scores than children aged 36–41 months ($p < .05$). Inter-rater reliability was excellent (Intra-class Correlation Coefficient = 0.96–0.99).

*Conclusions*: The ManuVis age band for 3-year-old children is appropriate to assess fine motor skills, and is sensitive to differences between children with visual impairment and normal vision and between half-year age groups. Reference scores are provided for 3-year-old children with visual impairment to identify delayed fine motor development.

**INTRODUCTION**

Fine motor skills allow us to successfully interact with our physical and social 30environment by providing the possibility to manipulate objects. Young children develop these skills, among other ways, by playing with toys. These experiences prepare them to engage in daily life, which contribute to their independence and increase their self-esteem as they grow older. Vision is essential for the development and performance of fine motor tasks, so differences in fine motor skills between children with visual impairment and children with normal vision are to be expected. However, inter-individual differences in motor skills between children with visual impairment are large[1] and not related to the degree of visual impairment[2]. Apparently, these inter-individual differences are at least partly due to differences in motor skill learning in daily life. To detect delays in motor skill learning as early as possible, insight in typical development in children with visual impairment is necessary.

Brambring[3] proposed that vision is essential for increasingly efficient ways of attuning to (i.e., detecting and using) relevant information for action. Vision facilitates motor control, as do other senses, in the context of a specific task as well as in response to an ever-changing environment. Specific sub-functions entail responding to and anticipating upcoming events, monitoring task performance, and providing feedback on the execution of goal-directed movements. Secondary functions of vision related to motor learning include the observation and imitation of the movements of others (e.g., peers or parents) as well as picking up on and responding to the encouragements of caregivers, which are mostly expressed nonverbally through gestures and facial expressions.



Motor skill learning is also influenced by the expectations of social environment, which are often lower for children with visual impairment than for children with normal vision[4]. In 2005, the prevalence of low vision in children aged between 0 and 14 years in the Netherlands was estimated at 0.6/1,000[5]. Research has demonstrated that children with visual impairment have poorer motor skills[2,6,7], delays in functional play[8], and reach motor milestones later than their peers with normal vision[9]. It has been suggested that developmental delays related to motor skills observed in children with visual impairment could be caused by less opportunities and poorer experience, rather than an inability to acquire these motor skills, see e.g.[10,11].

The developmental trajectory of children with visual impairment often differs from that of children with normal vision. For this reason, the motor capabilities of a child with visual impairment should not be expected to match with those of a child of the same age with normal vision. To determine whether a child with visual impairment exhibits developmental delays, motor skills need to be assessed relative to those of peers with visual impairment. Since children with visual impairment are at greater risk of less-than-optimal development of manual dexterity[12], an objective and reliable measure is needed to monitor fine motor development of young children with visual impairment.

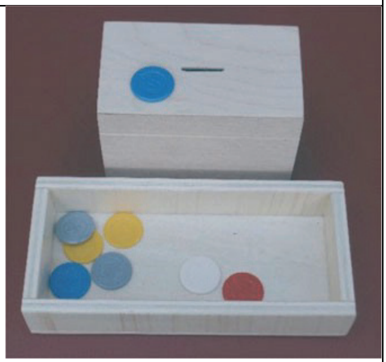
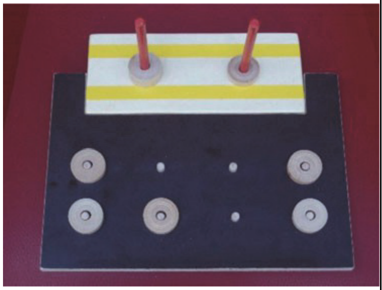
In a previous study, we developed the ManuVis, a test specifically developed to measure fine motor skills of children aged 4–11 years with visual impairment[1,13,14]. We observed that there were major differences between children with visual impairment and children with normal vision at the age of 4 years than between groups of older children. This was reported earlier by Brambing[15], who emphasized the relevance of these findings in terms of readiness to start in school. Assessment of fine motor skills at the age of 3 years would allow for earlier intervention and advice, even before children start going to school (in the Netherlands). This prompted the decision to extend the ManuVis with a ManuVis age band for 3-year-old children.

Several tests are available to measure fine motor skills in 3-year-old children with normal vision. The Bayley Scales of Infant and Toddler Development (BSID) assess a broad development range for children from 1 to 42 months of age[16]; the Mullen Scales of Early Learning[17] are tailored for children aged up to 68 months; and the Manual Dexterity Scale of the Movement Assessment Battery for Children-2 (MABC-2)[18]) is available to test fine motor skills in children aged 3 to 16 years. Although all these tests are appropriate to assess children with normal vision, none of the tests fulfills the requirements of being suitable for 3-year-old children with visual impairment, nor do they allow for monitoring development over time. Both the Mullen Scales and the BSID are specifically designed for young children. For BSID-III, an adapted version for children with motor and/or visual impairments exists[19]; however, it is not suitable for children aged more than 42 months. The advantage of MAB-2 is the wider age range; however, even for 3-year-old children with normal vision, certain man-ual dexterity items are difficult to execute[20].

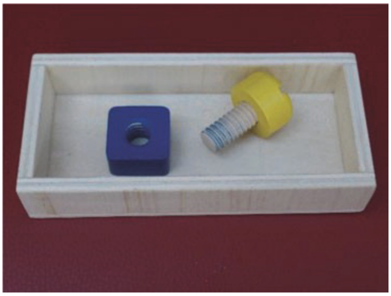
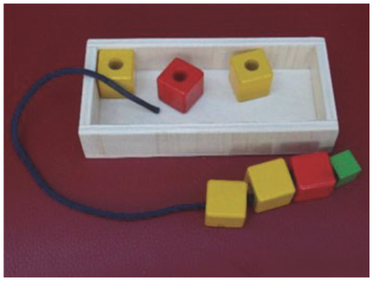
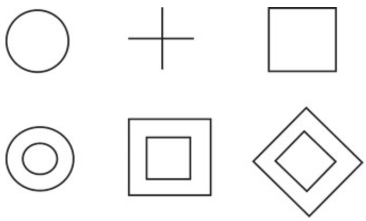
Therefore, we decided to carry out this study to provide a suitable instrument for clinical practice to measure fine motor skills in 3-year-old children with visual impairment and decided that a 3-year age band of the ManuVis[1,13,14] would allow for early intervention and longitudinal follow-up of the development in individual children. The ManuVis (4–11 years) consists of six different fine motor tasks (two one-handed tasks, three two-handed tasks, and a *pre-writing task*). See the Method section and Table 1 for an overview of the Manu-Vis items used in the age band for 3-year-old children and the adaptations made for these children in more detail. Test-retest and inter-rater reliability of the ManuVis is moderate to excellent, with Intra-class Correlation Coefficient (ICC) score ranging from 0.58 to 0.89 and from 0.98 to 1.00, respectively. Moreover, the test items were valid to discriminate between age bands. A more extensive description of the chosen items of the ManuVis (for 4–11-year-old children) and the validation are described by Reimer[1].

The aims of this study were to develop and evaluate an age band on the Manu-Vis for 3-year-old children with visual impairment by (1) comparing fine motor skill development of 3-year-old children with visual impairment and normal vision; (2) providing norm reference scores for 3-year-old children with visual impairment; and (3) investigating inter-rater reliability. We hypothesized that children with visual impairment would be slower in performing the fine motor tasks and would have more problems with accuracy in the pre-writing task. We also hypothesized that children aged 42–47 months would perform better than children aged 36–41 months in both visual impairment and normal vision groups.

**Table 1. Description of all tasks and corresponding procedures of the ManuVis for 3-year-old children compared with the ManuVis for 4–11 years**

| One-handed tasks | |
|---|---|
| 1. *Putting coins in a money box*<br>In the 'putting coins in a money box' task, the target position for the coins is known because the other hand feels the position and can remain there to provide a reference point. The 10 plastic coins are in an open container so that their starting position is relatively fixed. The box is placed with the long side facing the child. The task is carried out with both left and right hand, starting with the child's preferred hand. Start timing is when the hand is lifted to move, and stop timing is when the last coin strikes the bottom. Each hand is scored separately, and the scores in seconds from both hands are summarized to an item score.<br>*In the ManuVis (4–11 years), the same material and procedure are used.* |  |
| 2. *Putting rings on rods*<br>This task introduces an additional spatial factor: changing starting positions and target positions. Eight wooden rings must be placed on two vertical rods. The eight rings are initially in a fixed position in front of the rods on a wooden board in two rows of four rings. The task is to place the first ring on the first rod, the next ring on the second rod, and so on. The rings may be picked up in any order. If a child makes a procedural error (i.e., places two consecutive rings on the same rod), the child is reminded of the instructions once; subsequent errors are scored. To categorize procedural errors, four categories were described: (1) Correct execution, (2) Equal number of rings on the two rods, but there were procedural errors, (3) Unequal number of rings on the two rods, (4) All rings were placed on one rod.<br>The task is performed with both left and right hand, starting with the child's preferred hand. Start timing is when the hand is lifted to move, and stop timing is when the last ring is on the rod. Each hand is scored separately, and the scores in seconds from both hands are summarized to an item score.<br>*In the ManuVis (4–11 years), there are three rods and 12 rings for each hand.* |  |

4

| Two-handed tasks | |
|---|---|
| 3. *Screwing a nut onto a bolt*<br>This task includes an additional manual dexterity factor. A nut (35 mm) must be placed on a bolt (M 13 mm) using a counter-movement and then screwed on using fingers and thumb. In terms of positioning of objects in space, this task is easier because the child starts with a nut in one hand and the bolt in the other and these must then be brought together. The nut and bolt are placed in an open container. Start timing when the hands are lifted to move and stop when the nut is screwed until the end.<br>*In the ManuVis (4-11 years), there are two nuts (19 mm), and the size of the bolt is M 10 mm.* |  |
| 4. *Threading beads*<br>Six cubic beads (each side 27 mm) are placed in an open container with the holes facing up. These must be threaded onto a piece of cord. One hand manipulates the bead and feels for the hole, and the other hand threads the cord through. Start timing is when the hands are lifted to move, and stop timing is when the last bead has slit past the stiff part of the lace.<br>*In the ManuVis (4–11 years), there are six smaller beads (h = 16 mm/w = 12 mm/d = 4 mm) and the lace is thinner and the stiff end is shorter.* |  |
| 5. *Threading lace*<br>This task is not part of the ManuVis for 3-year-old children because of the degree of difficulty. | |
| 6. *Pre-writing task*<br>The pre-writing items are comparable with those in BSID-II. The task requires children to copy three geometrical objects (circle, plus sign, and square) and they have to draw a trace between two boundary lines of a circle, square, and diamond. The score is the amount of correct executions.<br>*The ManuVis (4–11 years) has a different writing task.* |  |

**METHOD**

***Design***

This prospective cohort study is embedded in a larger study focusing on interventions for children with visual impairment. Baseline data were used for this study.

The entire research study was approved by the Medical Ethics Committee (CMO 2010/037 Arnhem Nijmegen/NTR = 2494), and was conducted in accordance with the Declaration of Helsinki. All parents of the participants gave written informed consent.

***Participants***

Fifty-four 3-year-old children participated in this study, 26 children with visual impairment (mean age 3 years 7 months (SD 3 months); 17 boys and 9 girls) and 28 children with normal vision (mean age: 3 years 7 months (SD 4 months); 14 boys and 14 girls). The children were divided into two groups based on age. One group consisted of children aged 36–41 months (nine with visual impairment and 11 with normal vision).

The other group consisted of children aged 42–47 months (17 with visual impairment and 17 with normal vision).

Visually impaired children were selected from the databases of the institutions for visual rehabilitation in the Netherlands. The inclusion criteria for children with visual impairment in this study were as follows: visual acuity of at least 0.05 and ≤0.3, no known comorbidity and/or cognitive impairment, birth at term (i.e., ≥36 weeks of gestation), and normal birth weight. Children born prematurely were excluded because they are known to have a higher risk of motor performance deficits and other comorbidities[21,22]. The ManuVis[1] had the same exclusion criteria. The children had to be able to cooperate and understand the tasks of the ManuVis age band for 3-year-old children. The parents of the children selected for inclusion received an invitation letter, and were also contacted by telephone. If they were willing to participate and written informed consent was received, an appointment was made for the measurements. The clinical data of the children with visual impairment are described in Table 2.

**Table 2. Descriptive Data for Children with Visual Impairment (*n* = 26)**

| | Sex M/F | Age (months) | Diagnosis | Va Snellen equivalent | Strabismus | Refraction |
|---|---|---|---|---|---|---|
| 1 | M | 37 | Congenital stationary night-blindness (CSNB 2) | 20/130 | + | OD S-2/OS S-2 |
| 2 | M | 38 | Albinism | 20/125 | + | - |
| 3 | F | 39 | Congenital nystagmus | 20/100 | - | - |
| 4 | M | 39 | Congenital nystagmus | 20/160 | - | OD S –2/OS S –2 |
| 5 | M | 39 | Albinism | 20/200 | - | OD S +2/OS S +2 |
| 6 | F | 41 | Hyperopia[a] | 20/80 | - | OD S +3/OS S +2.75 |
| 7 | F | 41 | Albinism | 20/200 | +/- | ODS –4/OSS –4 |
| 8 | M | 41 | Cone dystrophy | 20/130 | +/- | ODS –4/OSS –4 |
| 9 | M | 41 | Albinism | 20/67 | + | - |
| 10 | F | 42 | Macular hypoplasia | 20/500 | + | - |
| 11 | M | 42 | Congenital stationary night-blindness (CSNB 2) | 20/100 | - | OD S –8 / OS S –7.75 |
| 12 | F | 43 | Retinal coloboma[a] | 20/130 | - | OD S –7/OS S –5 |
| 13 | F | 44 | Aniridia | 20/80 | - | OD S +2.5/OS S +2.5 |
| 14 | M | 44 | Congenital nystagmus | 20/125 | - | - |
| 15 | M | 44 | Albinism | 20/100 | - | - |
| 16 | M | 45 | Glaucoma (aphakia)[a] | 20/67 | + | OD S +18.75/OD S +18 |
| 17 | M | 45 | Aniridia | 20/160 | - | OD S +2.75/OS S +5 |
| 18 | M | 45 | Albinism | 20/200 | - | OD S +5/OS S +.5 |
| 19 | M | 45 | Albinism | 20/160 | + | OD S –1.25/OS S –1.25 |
| 20 | F | 46 | Albinism | 20/67 | - | - |
| 21 | F | 47 | Unknown | 20/67 | + | - |
| 22 | F | 47 | Albinism[b] | 20/80 | + | OD S +4.75/OS S +7.25 |
| 23 | M | 47 | Congenital nystagmus | 20/160 | - | OD S +2/OS S +3.5 |
| 24 | M | 47 | Albinism | 20/67 | + | OD S +4/OS S +4.75 |
| 25 | M | 47 | Albinism | 20/160 | - | - |
| 26 | M | | Albinism | 20/125 | - | - |

[a] The visual field is not completely intact. [b] Nystagmus is absent

Children with normal vision were living in the neighborhood of the Dutch city of Utrecht and were recruited from three local kindergartens (*n* = 15) and through a social network of other children with normal vision (*n* = 13) who had participated. Parents of 3-year-old children with normal vision received an invitation letter, a were also contacted by telephone. If they were willing to participate and written informed consent was received, an appointment was made for the measurements.

### *Adaptation of ManuVis Materials*

The materials of the original ManuVis were adapted to make the items suitable and more attractive for 3-year-old children. All components of the version for 3-year-old children and corresponding procedures are described in Table 1, including the differences with the original ManuVis items in the age band for 4–11 years. The extension of the ManuVis comprised two one-handed tasks (*putting coins in a moneybox*, and *putting rings on rods)*, two two-handed tasks (*screwing a nut onto a bolt*, and *threading beads*), and a pre-writing task with age-related forms and figures.

Test-retest reliability (ICC score between 0.58 and 0.89) and inter-rater reliability (ICC score between 0.98 and 1.00) for the test items and total scores of the ManuVis are moderate to excellent[1].

### *Procedure*

Testing was always done in a quiet and well-lighted room without interruptions.

All testers had experience in testing children with visual impairment. The performance of each child was recorded on videotape. The video camera was placed opposite the child.

Children were instructed to work as quickly as possible during one- and two-handed tasks and as accurately as possible during pre-writing task. The time necessary to complete the one- and two-handed tasks was measured in seconds, and the score of the pre-writing task was the amount of correct executions. If a procedural mistake occurred, the tester interrupted the attempt immediately, after which corrective instructions and/or demonstration of the task were given before the child started again. A procedural mistake in putting coins in a moneybox occurred when a child picked up two coins at the same time, and in putting rings on rods when a child placed two consecutive rings on the same rod. Furthermore, for both one-handed items, a procedural error occurred when a child changed hands or used both hands, and for the two-handed tasks, when a child picked up more than one bead at a time. Two new attempts were allowed. For the item putting rings on rods, the children were allowed to continue during the third attempt even in the occurrence of errors; however, these procedural errors were counted. Total testing took between 15 and 20 min.

It is known that children with visual impairment use fewer fingers and exhibit less variation in grip than children with normal vision[12], which might influence their performance on pre-writing skills. Therefore, pencil grip was observed and categorized as (1) primitive grips, (2) transitional grips, or (3) mature grips, based on the classification system by Schneck & Henderson[23].

Two pediatric physical therapists (AM Reimer (AR)/A Overvelde (AO)), who were experienced with this type of grip categorization, independently scored the grip from video. The highest level of the observed pencil grip was scored. Inter-rater reliability was calculated between the two observers. For further analysis, consensus on the final category was reached when the two researchers had different initial scores.

One child with normal vision and another with impaired vision were unable to perform any of the tasks, and were excluded from the analysis. Three children with visual impairment, aged 36–41 months, failed to execute the items of screwing a nut onto a bolt and threading beads, and two children with visual impairment, aged 36–41 months, failed to execute the item of threading beads. There was no video recording available for administration of pencil grip of one girl with normal vision.

### *Data Analysis*

Task scores for one- and two-handed tasks were based on the performance time in seconds, and total score was the sum of task scores 1–4. For the calculation of total score in case of a missing item, when a child could not complete the task, the lowest score in the same age category and group was used. Excluding these children from the analysis would have biased the results. In addition, for the item, putting rings on rods, procedural errors (e.g., placing two consecutive rings on the same rod) were reported. The score of pre-writing task was based on the number of correct executions (from 0 to 6). Scores belonging to the 5th and the 15th percentile were calculated for use as reference values in clinical settings.



The scores of children with visual impairment and normal vision were approximately normally distributed. Therefore, six univariate analyses of variance (ANOVA) with age (36–41 months and 42–47 months) and group (children with visual impairment and children with normal vision) as between-subject factors were conducted on each task score as well as the total score. Chi-square tests were performed to compare procedural errors on the item putting rings on rods between groups (visual impairment and normal vision) and age categories. To determine whether pencil grip and the total score of the pre-writing task were correlated, nonparametric Spearman's rho was calculated.

In order to determine inter-rater reliability, the performance of 12 children (five children with visual impairment and seven with normal vision) was scored twice. One tester (Karlijn Oude Wolbers/Janne van Essen) scored the items of five children with visual impairment during testing, whereas the second scorer (AR) scored later from video recordings. Seven children with normal vision who were scored during testing by Annemieke Gerrits and were later scored by AR from video recordings were also included. Inter-rater reliability was calculated using ICC and standard error of measurement (SEM) for each item separately as well as the total score. ICC score was interpreted using the following criteria: 0.00–0.49 is poor; 0.50–0.74 is moderate, and 0.75–1.00 is excellent [24]. For each ICC, the 95% Confidence Interval (CI) was calculated. The SEM describes the error in interpreting an individual's test score and is computed as follows: $SEM = SD \times \sqrt{(1 - ICC)}$, with SD as the standard deviation. The inter-rater reliability for the score of pencil grip was determined using the Kappa statistic.

All data were analyzed using IBM SPSS (version 21.0). A significance level (alpha) of 0.05 was applied throughout.

## *RESULTS*

Scores on the ManuVis are presented in Table 3. Univariate ANOVA score revealed significant differences between scores of children with visual impairment and normal vision for all tasks. Children with visual impairment needed on average 17 s more for putting coins in a moneybox ($F(1,50) = 26.44$; $p < .001$) and putting rings on rods ($F(1,50) = 19.44$; $p < .001$), 14 s more on screwing nut onto a bolt ($F(1,50) = 23.70$; $p < .001$), and 18 s more on threading beads ($F(1,50) = 10.80$; $p = .002$). The mean group difference in total score was 66 s, a difference that was significant ($F(1,50) = 32.34$; $p < .001$). Furthermore, children with visual impairment were less able to perform the pre-writing task ($F(1,50) = 16.95$; $p < .001$). In addition, ANOVA score showed a significant main effect between children aged 36– 41 months and 42–47 months. Older children were faster at putting coins in a moneybox ($F(1,50) = 6.46$; $p = .014$), screwing a nut onto a bolt ($F(1,50) = 6.38$, $p = .015$), and had a better total score ($F(1,50) = 4.92$; $p = .031$). No significant interactions were found.

A Chi-square test revealed no significant differences between children with visual impairment and normal vision, or between younger and older children on procedural errors in the item putting rings on rods. Six children with normal vision and three children with visual impairment were able to execute the task in correct order, 13 children with visual impairment and nine children with normal vision placed consecutive rings on the same rod, eight children with visual impairment and nine children with normal vision ended with unequal numbers of rings on each rod, and one child with normal vision placed all the rings on one rod.

Spearman's rho revealed no significant correlation between pencil grip and performance on the pre-writing task. All children used primitive or transitional grips during the pre-writing task. Half of the children with visual impairment used the primitive grasp with extended fingers. For the children with normal vision, the most frequently used grip was the transitional static quadripod grip: seven children applied this grip.

Table 3. ManuVis outcomes per task and per age group for children with visual impairment (VI) and children with normal vision (NV).

| Item | Group | Age category (months) | Mean (*SD*) | P15 | P5[a] | Range | ICC | CI (95%) | SEM |
|---|---|---|---|---|---|---|---|---|---|
| 1. Putting coins in a moneybox (s) | NV | 36-41 | **61** *(7)* | 66 | 77 | 46-77 | 0.96 | 0.86 - 0.99 | 2.78 |
| | | 42-48 | **51** *(11)* | 64 | | 36-77 | | | |
| | VI | 36-41 | **75** *(14)* | 91 | 92 | 53-93 | | | |
| | | 42-48 | **69** *(14)* | 87 | | 47-90 | | | |
| 2. Putting rings on rods (s) | NV | 36-41 | **46** *(12)* | 61 | 64 | 28-67 | 0.99 | 0.96 – 0.99 | 2.47 |
| | | 42-48 | **36** *(7)* | 46 | | 26-49 | | | |
| | VI | 36-41 | **59** *(20)* | 91 | 87 | 27-86 | | | |
| | | 42-48 | **57** *(16)* | 81 | | 41-87 | | | |
| 3. Screwing a nut onto a bolt (s) | NV | 36-41 | **20** *(10)* | 35 | 36 | 7-37 | 0.99 | 0.96 – 0.99 | 2.75 |
| | | 42-48 | **13** *(5)* | 19 | | 6-26 | | | |
| | VI | 36-41 | **35** *(10)* | 43 | 55 | 19-43 | | | |
| | | 42-48 | **26** *(15)* | 47 | | 6-57 | | | |
| 4. Threading beads (s) | NV | 36-41 | **48** *(11)* | 60 | 70 | 29-63 | 0.99 | 0.99 – 0.99 | 3.25 |
| | | 42-48 | **43** *(13)* | 57 | | 28-76 | | | |
| | VI | 36-41 | **66** *(20)* | 86 | 123 | 39-86 | | | |
| | | 42-48 | **62** *(30)* | 112 | | 33-127 | | | |
| 5. Pre-writing task (n) | NV | 36-41 | **3.2** *(1.8)* | 0 | 0.5 | 0-5 | 0.99 | 0.96 - 0.99 | 0.38 |
| | | 42-48 | **3.5** *(1.6)* | 1.6 | | 1-6 | | | |
| | VI | 36-41 | **1.1** *(1.9)* | 0 | 0 | 0-6 | | | |
| | | 42-48 | **1.7** *(1.6)* | 0 | | 0-5 | | | |
| Total item 1-4 (s) | NV | 36-41 | **174** *(26)* | 212 | 212 | 117-213 | 0.99 | 0.96 – 0.99 | 5.57 |
| | | 42-48 | **143** *(28)* | 174 | | 108-206 | | | |
| | VI | 36-41 | **235** *(56)* | 302 | 315 | 155-303 | | | |
| | | 42-48 | **214** *(54)* | 284 | | 148-321 | | | |

Mean scores, 15th and 5th percentiles as well as the range per group and age category are reported, along with the results on the interrater reliability per item. The scores for items 1 – 4 and *total item 1-4* are in seconds, the score for the pre-writing task is the number (between 0 and 6) of correct trials.

[a] For calculation of the 5th percentile data of the two age groups were combined because of the sample size.

### *Inter-Rater Reliability*

The intra-class correlation coefficient scores of each item were excellent, ranging from 0.96 to 0.99. SEM scores were below 5.57 (see Table 3). In addition, the interrater reliability for the assessment of pencil grip, expressed with the Kappa coefficient, was sufficient ($\kappa$ = 0.68, $n$ = 53).

## DISCUSSION

This study demonstrates that 3-year-old children with visual impairment on average have lower scores on the ManuVis age band for 3-year-old children compared with their peers with normal vision. Three-year-old children with visual impairment needed more time to complete all fine motor tasks and they had more difficulty with the accuracy of the pre-writing task. The inter-rater reliability of the ManuVis for 3-year-old children was excellent for all items. Inter-rater reliability for the assessment of the pencil grip was sufficient.

The item screwing a nut onto a bolt was difficult for the children with visual impairment; five of the 11 children with visual impairment aged 36–41 months could not complete this task. In addition, two of these children were also unable to perform the item threading beads. For the analysis, these missing values were replaced with the lowest value on that item for children within the same group and age category. This decision was based on the observation during testing that the motor skills of these children were insufficient to execute these tasks, which was confirmed by inspection of the data: they were also slow performers on the tasks that they were able to execute. Excluding them from the analysis would have led to the overestimation of results of the remaining children with visual impairment. We recommend using a similar approach in clinical practice to be able to compute a total score in the case of missing item. Letting children struggle with a task when they are clearly unable to perform it would presumably take longer, and therefore taking the lowest value is a conservative estimate.

For the task screwing a nut onto a bolt, minimal visual inspection is required; the task can be guided through haptic sensory information provided by the materials. Therefore, we expected that children with visual impairment would be able to perform this task. However, not all children with visual impairment executed this task adequately. In contrast, all children with normal vision were able to perform this task, which is consistent with the norms for a similar task in the Mullen scales for children aged 20–26 months[17]. Considerable difficulties children with visual impairment seemed to have with screwing a nut onto a bolt and threading beads might be caused by a lack of experience with such tasks, cf.[10,11].

Children with and without visual impairment had difficulties in executing the item putting rings on rods, specifically with the correct order of the rings. The majority of children placed consecutive rings on the same rod. This seems to reflect problems with understanding where to place the ring, rather than problems with performing the movements. It is unlikely that this affected the execution time on this task, since children still placed rings on one of the rods, although not necessarily on the correct one.

Pencil grip and scores on the pre-writing task were not correlated. This is in line with research of the effect of pencil grip on speed and legibility of handwriting[25]. The grip patterns were immature in both groups, but half of the children with visual impairment used a primitive grip with extended fingers whereas more children with normal vision were using the more mature transitional quadripod grips. Although we did not find a relationship between the ability to immature grip in young children to stimulate learning to write with a dynamic pen grip[26].

Previous research on the development of fine motor skills in children with visual impairment and with normal vision aged 4-11 years has shown an age-related decrease in execution time and an increase in the accuracy of writing tasks[1]. The item putting coins in a moneybox was also used in the original version of the ManuVis for 4 years, which allows comparison of the scores of the 3-year-old children with visual impairment with the scores of older children with visual impairment. The results of the present study with the ManuVis support the trend shown in the previous study with the ManuVis: Older children with visual impairment are faster at performing the task putting coins in a moneybox than younger children with visual impairment (mean scores at: 3 years = 72 s, 4 years = 54 s, 5 years = 46 s, 6 years = 44 s;[1]. Four-year-old children with visual impairment in our previous study performed the task putting coins in a moneybox at a similar level as done by 3-year-old children with normal vision in the present study, which demonstrates a possible delay in children with visual impairment.

4

The evaluation of fine motor skills in children with visual impairment is important. Identifying a child with motor deficits can prompt caregivers to employ motor tasks and play material that strengthen fine motor skills. For parents, it is difficult to estimate what a child can or cannot see, and this may lead to over- or underestimation of their capabilities. It is important for parents of children with visual impairment to enable them to engage in activities focusing on fine motor skills and to encourage practicing fine motor tasks. At this young age, there is an important role for positive parent–child interactions[27]. It is recommended to refer children to a developmental center as soon as severe visual impairment is suspected[28].

The main limitation of this study is the number of children included. This mostly affects the normative values for the ManuVis age band for 3-year-old children with visual impairment. Further research with more children is recommended to provide more valid reference scores. The sample was representative of 3-year-old children with visual impairment in the Netherlands. Although the number of children in each group was small, differences between the groups were significant. To reduce the possibility that delays in fine motor skills of children were caused by preterm birth rather than visual impairment, we excluded prematurely born children. We know from earlier studies that a preterm birth is a risk factor for brain damage and cerebral palsy. Even in the population of premature children without known deficits, the percentage of children with abnormal motor development is high. This ranges from 45% at the age of 2 years to about 37% at the age of 5 years[21,22,29-31].

Another limitation is that the design was cross-sectional. It would be advisable to monitor children with visual impairment and children with normal vision longitudinally to determine differences in individual developmental trajectories.

## CONCLUSIONS

This study found excellent inter-rater reliability for the ManuVis age band for 3-year-old children while measurement error was acceptable. Moreover, the test was sensitive to differences between children with and without visual impairment, and between age categories. The ManuVis can be used to assess the development of fine motor skills of child with visual impairment (in the age range of 3–11 years) by comparing performance with peers with and (without) visual impairment. Early identification of fine motor delay in children with visual impairment is encouraged to initiate coaching and preventive interventions as soon as possible[14]. Stimulating development of fine motor skills is recommended before children need these skills at school[32].

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# Chapter 5

# Young children's use of a visual aid: an experimental study of the effectiveness of training

Ralf Cox
Annemieke Reimer
Cornelis Verezen
AD Smitsman
Mathijs Vervloed
Nienke Boonstra

Dev Med Child Neurol 2009; 51(6): 460-7.

**ABSTRACT**

We report an experiment concerning the use of a stand magnifier by young children with visual impairments (21 males, 12 females; mean age 4y 8mo [SD 11mo]). Children had a normative developmental level and a visual acuity of 0.4 or less (≤20/50 in Snellen's notation). To measure magnifier use objectively, we developed a task that closely resembled the dynamics of its real-life (pre-reading) use. Children had to follow trails visually, from a start location to an unseen end location. This could only be done successfully and reliably by proper use of the magnifier. In addition to this, we analyzed the effect of specific training with the magnifier by using a repeated-measures (before and after training) matched- groups (with respect to age and near-visual acuity) design. Results established both the task's efficacy as an instrument for measuring magnifier use in young children and the effectiveness of the training. Improvement in task performance after training was found in both groups, except for the youngest children (<3y 6mo). On average, 1.8 times as many paths were followed in both groups after training ($p$=0.001). The without-magnifier training group became 2.5 times as good at finding the correct end location, where as the with-magnifier training group became 4.3 times as good ($p$=0.05).

## INTRODUCTION

Several studies recommend the use of low-vision aids for young children with a visual impairment[1-4]. Nonetheless, we still understand very little about the actual use of such aids in this group, from the scientific as well as from the clinical and applied point of view. Little is known, for example, about the relevant perceptual, motor, and cognitive factors that determine the successful and prolonged use of low-vision aids in a developmental context. Resulting from this is the lack of consensus among low-vision professionals and researchers on the minimum age at which it is sensible to introduce a low-vision aid to a child with a visual impairment. Moreover, we do not know exactly at what age and how children should be trained in order to use such an aid adequately. Nor do we know whether a training program at an early age will improve a child's willingness to use the aid. From a social and economical perspective this situation is unsettling. Obviously, the number of successful (early) prescriptions of low-vision aids to children with visual impairments would significantly be increased by a resolution of these issues.

### A measure of the effectiveness of a low-vision aid for young children

Related to the lack of knowledge, in our opinion, is the scarcity of clear-cut measures or evaluation procedures for the effectiveness of low-vision aids for children with visual impairments[5-7]. Clinical measures are often idiosyncratically tied to a specific low-vision professional and therefore not generally applicable or evidence-based. More objective measures almost always involve some ability that is directly related to reading, such as reading rate or comprehension rate. Although these are both functional and relevant, they are not appropriate for preschool children. For early prescription purposes, it would be important to have a measure that also applies to this group. A further point is that training described in detail in the literature, let alone evaluated (however, see Corn et al.[5] and Virgili and Acosta)[8].

5

As an exception, in an experiment by Ritchie et al.,[7] visually impaired children aged 18 months to 5 years had to name pictures and small objects with and without the help of a stand magnifier on two occasions. Results revealed an improvement in functional vision (i.e. ratio of correct responses) when using the magnifier, except for the children with the most severe visual disability. All children benefited from the magnifier after a trial period. The researchers reported that this improvement was independent of age, although their task required a developmental level of at least 2 years.

Two comments can be made with respect to that study[7]. First, the task was a static magnifier-aided naming task, which involved no manipulation of the magnifier. A task that would resemble more closely the actual movements involved in scanning or reading[9] would be more realistic and reliable. Second, the trial period with the magnifier is an uncontrolled factor in the experiment. In fact, specific training is likely to increase the size of the progress that might be achieved with the aid (e.g. see Corn et al.)[5]. The effect of such training on performance task remains an empirical matter.

**The present study**

The work presented in this paper is motivated by the fundamental and practical need to address the issues that have been raised in the preceding sections. We developed a task and a related training program that would encourage and help young children with visual impairments to develop their abilities in using a low-vision aid for near vision. At the same time, the task had to enable us to determine the effectiveness of the training program and make individual assessments. That is, it must provide a reliable and objective quantitative measurement of the individual child's abilities in using the magnifier, as well as the changes resulting from the training. The task was designed to be appropriate for preschool children but still entailed the kind of motor behavior (in particular magnifier movements) involved in reading and scanning. The stand magnifier was considered to be the most appropriate visual aid for our purposes because it offers a stable image and is easy to manipulate for children[3].

## METHOD

**Participants**

At the end of 2005, 57 children with a visual impairment were recruited from the patient files of the low-vision centres in the Netherlands. Of this group, 42 children agreed to participate. Children were selected on the basis of the following criteria: a visual acuity of 0.4 or less (≤20/50 in Snellen's notation) in the better eye after the best possible correction, no additional impairments, birth at term, and no previous experience with visual aids. Moreover, they had a normative developmental level, determined by the Reynell–Zinkin development scales for young visually impaired children[10], for which Dutch age norms[11] were applied.

Five children did not complete the study, because of withdrawal (three children) or organizational problems (two children). Four more children were excluded from the analyses because reassessment of their near-visual acuity revealed that they received inappropriate stimulus material. The remaining group of 33 children contained 21 males and 12 females. The average age in this group at the start of the study was 4 years 8 months (SD 11mo).

**Ophthalmological examination**

In the research group, 12 children had albinism (10 with nystagmus), five had cataract (three with nystagmus), four had nystagmus only, three had retinoschisis (one with nystagmus), two had aniridia (both with nystagmus), and there were seven children with either high hypermetropia, Mobius syndrome, retinitis pigmentosa, congenital glaucoma, achromatopsia (with nystagmus), optic nerve atrophy (with nystagmus), or retinoblastoma. This distribution is representative of Dutch children with visual impairments[12,13]. Visual acuity was measured for the right eye, left eye and both eyes on 3m and 5m charts (LH-test[14,15] and E-chart[16] respectively) under controlled lighting conditions in an ophthalmological setting. Near-visual acuity was determined with the LH- test (line and single) for children, also for the right eye, left eye and both eyes, at a distance chosen by the child and at 40cm[17].

A gross estimation of the visual field was obtained by confrontational techniques. Central scotomas could not be tested with perimetry in these young children. However, loss of function in the central area was observed when the child performed near-vision tasks[15]. Six children had visual-field defects; three of these had a normal central visual field and three probably had central scotomas. Indications for central scotomas were found in children with retinitis pigmentosa, retinoblastoma (after treatment), and congenital glaucoma.

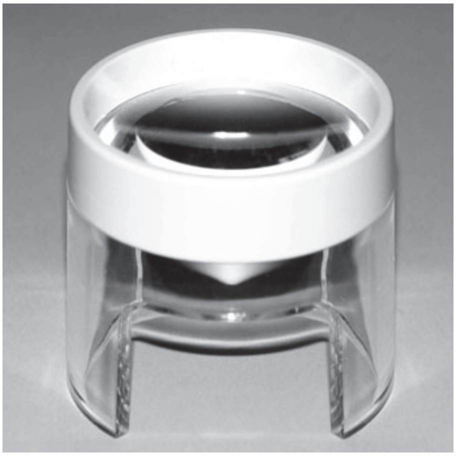
Objective refraction was obtained after cycloplegia and if necessary the spectacle correction was prescribed or changed before the experiment started. Four children with afakia wore glasses, and one child had intraocular lenses. All children with glasses had to wear them during the entire study (high hypermetropia n=8, hypermetropia with astigmatism n=12, myopia n=2, no correction n=6).

## MATERIAL

### Stand magnifier

The visual aid used in the experiment is shown in Figure 1. It is a 23.0 diopter (aspheric-lens) stand magnifier (Eschenbach, Nuremberg, Germany) with a 6·standard magnification. It has an equivalent viewing distance (EVD) of 4.3 cm[18]. The magnifier is 48mm high and its lens housing has a diameter of 52mm. These dimensions make it quite suitable for young children to manipulate. Because a stand magnifier rests on the surface of the underlying material, it provides a stable image and movements with the magnifier need to be made in only two dimensions.

**Figure 1. The magnifier used in this study: Eschenbach aspheric-lens stand magnifier (23.0 dpt, 6x magnification. Dimensions: 48mm high and52 mm diameter**



**Task**

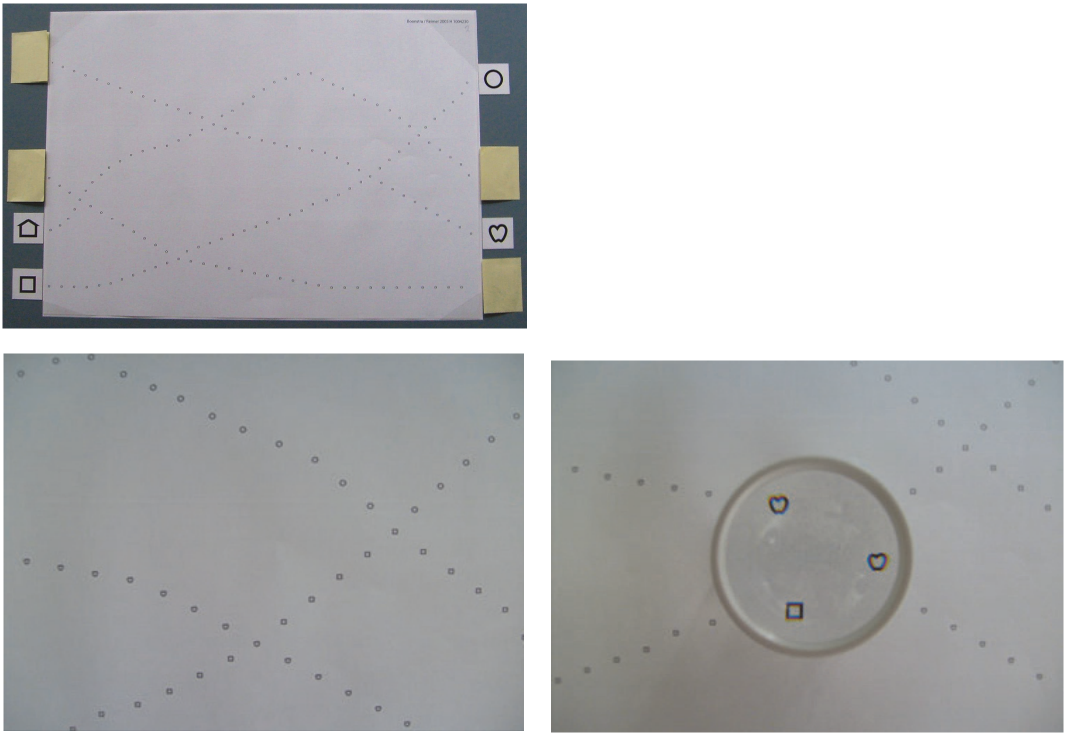
The stimulus materials used for the task were eight A3-sized (29.7 x 42cm) patterns, which were mounted on larger cardboards for stability (Figure 2). Each pattern included four different trails, each consisting of one type of small optotypes (Lea symbols: apple, circle, house and square),[14,15] somewhat like an 'ant trail'. The trails all connected a start picture to a hidden finish picture. The task comprised a 'trace-following game': starting at an initial location pointed out by the experimenter, the children had to find the corresponding finish picture, by following the trail with the magnifier.

Several sets of patterns were available with trails in optotype sizes that varied across a large range of M-values. The M-values reflect a logMAR (minimal angle of resolution) visual-acuity scale, in which the angular size of the optotypes changes by a factor of 0.1 log units at each step. To be absolutely certain that children were not able to identify the optotypes without the use of the magnifier, we selected a size that was three lines on the LH-chart[16] lower than the threshold M-value that a child had attained in the near-visual acuity test. As a result, children were still able to see the trails with their bare eyes, but for adequate task performance the magnifier was indispensable.

The distance between the optotypes was such that, when the magnifier was positioned on a trail, the image through the lens always contained at least two optotypes (of any kind) at once. Because of this, not only optotype information was available to the child, but the magnifier image also provided information on the movement orientation.

The patterns varied in orientation (horizontal, vertical, and circular) and type of crossings between the trails (interruption or continuation of the trail; see Figure 2). These variations were included to attract across-pattern (scanning-like) movements and to increase the necessity to use the magnifier respectively.

**Figure 2.** **The stimulus material:(a) one of the horizontally orientated patterns; (b) a detail of this pattern in the neighbourhood of two crossings;(c) a detail of the pattern as partially seen through the magnifier**





### Procedure

Before the start of the actual experiment, some simple example patterns in large optotypes (2.5M) were used to point out the correspondence between the start and finish picture, and to explain the ideas of trail following and crossings. These patterns were not yet done with the magnifier. A final introductory pattern with smaller optotypes was used to introduce the magnifier.

When the task was clear to the child, the first experimental pattern was introduced, which had to be performed with the magnifier. The experimenter placed the magnifier on the starting picture and asked the child to follow the trail. The patterns were presented in a random order. Only two trails had to be performed from each pattern before a new one was presented, resulting in a maximum number of 16 trails. When a child arrived at an incorrect end location, or somewhere during the trail indicated that a wrong turn was made, he or she was allowed to try one more time. If this second try also failed, a new trail was appointed. The experiment ended when all 16 trails were done or when the child refused to proceed. Finally, lighting conditions were controlled for by using task lighting that was directed onto the pattern by the experimenter (9000lux at close range to the patterns [10–15cm] and 1300lux at 40cm).

**Training and post-test**

To evaluate the effectiveness of the magnifier training in an objective way, two training groups were constructed. Half of the children were assigned to the group that trained with the magnifier (experimental group), the other half trained without the magnifier (control group). With this set-up it is possible to differentiate the effect of (mere) increased visual attention from the (additional) effect of the magnifier. The training groups were matched with respect to age and near-visual acuity. The without-magnifier training group (n=15, 9 males, 6 females) had an average age of 57 months (SD 12mo) and an average stimulus-material M-value of 0.35 (SD 0.13). The with-magnifier training group (n=18, 12 males, 6 females) had an average age of 55 months (SD 11mo) and an average stimulus-material M-value of 0.38 (SD0.15).

Training started within 1 week after the pretest. Children were visited at home or at their school by an early interventionist for 12 half-hour sessions over 6 weeks. The material and the magnifier were not made available to the children outside the training sessions. During the training sessions, presentation of the different patterns was varied as much as possible. In the with-magnifier training group, the size of the optotypes was again three lines on the LH-chart lower than the near-visual acuity, and much attention was paid to improvement of magnifier use. In the without-magnifier training group, the size of the optotypes was equal to the near- visual acuity, and children used their finger to follow the trails.

Within 1 week after the last training session children performed the post-test. The set-up was the same as during the pretest. In particular, both training groups worked with the magnifier again, and the patterns and trails were presented to each child in the same order as during the pretest. The experimenters were not the same ones as during the pretest and were not informed whether the child had trained with or without the magnifier.

**Data analysis**

Task performance was analysed in terms of both quantity and quality. Quantity of performance refers to the number of trails followed, irrespective of whether the correct end location was found. Quality of performance refers to the number of correct end locations that were found among these trails. To indicate changes in this quality (caused by the training) several measures were used, all based on the proportion correct, given by the ratio of correct end locations to the number of trails that were tried. A proportion correct of 0 means that no trail that was started resulted in the correct end location being found, whereas a proportion correct of 1 means that all trails ended successfully. The distinction between these quantitative and qualitative aspects of task performance is meant to reflect that, by using the magnifier incorrectly or not at all, it is still possible to (partially) follow a trail, but it is not possible to reliably arrive at the correct end location.

Analysis of variance was performed on the relevant data for both types of task performance to compare the differences between training groups and age groups. All proportion scores were arcsine transformed before being entered into the analysis of variance to account for the fact that they are not normally distributed.

## RESULTS

The characteristics and results of all 33 participants who completed the pre- and post-tests and all 12 training sessions are presented in Table 1.

### Task performance: quantitative

Generally, children in both training groups followed more trails in the post-test than in the pretest (Table 1; note that the maximum number of trails was 16). The average number of trails for the without-magnifier training group was 7.3 (SD 3.4) in the pretest and 13.5 (SD 4.0) in the post- test. For the with-magnifier training group these numbers were 6.9 (SD 4.7) and 12.1 (SD 5.9) respectively. Thus the quantitative task performance increased from pre- to post- test by a factor of 1.8 in both groups. A paired-samples t-test comparing the pooled number of trails in the pretest and post-test showed that this increase is significant: $t(32)=-7.46$, $p<0.001$ (two-tailed).

An analysis of covariance, with training group (with or without magnifier) as the intersubject factor and age and number of trails followed in the pretest as covariates, was performed on the number of trails followed in the post- test. This revealed that the number of trails followed before training was the only significant factor in determining post-test task performance: $F(1,33)=5.63$, $p=0.02$. Age and training group were not significant.

**Table 1. Raw scores in the pretest and post-test for the 33 participating children**

| Child | Age(mo) | Sex | NVA[a] | Pretest N of paths | Pretest N correct | Pretest Proportion correct | Post-test N of paths | Post-test N correct | Post-test Proportion correct |
|---|---|---|---|---|---|---|---|---|---|
| *Training group without magnifier* | | | | | | | | | |
| 1 | 37 | M | 0.13 | 3 | 0 | 0.00 | 12 | 4 | 0.33 |
| 2 | 40 | M | 0.10 | 1 | 0 | 0.00 | 6 | 0 | 0.00 |
| 3 | 43 | M | 0.20 | 4 | 0 | 0.00 | 16 | 10 | 0.63 |
| 4 | 47 | F | 0.15 | 10 | 1 | 0.10 | 16 | 8 | 0.50 |
| 5 | 51 | M | 0.20 | 10 | 5 | 0.50 | 12 | 10 | 0.83 |
| 6 | 51 | F | 0.06 | 6 | 0 | 0.00 | 16 | 14 | 0.88 |
| 7 | 55 | F | 0.13 | 3 | 0 | 0.00 | 16 | 15 | 0.94 |
| 8 | 55 | M | 0.30 | 10 | 5 | 0.50 | 8 | 6 | 0.75 |
| 9 | 56 | M | 0.16 | 8 | 4 | 0.50 | 16 | 13 | 0.81 |
| 10 | 58 | M | 0.19 | 9 | 0 | 0.00 | 16 | 6 | 0.38 |
| 11 | 67 | F | 0.10 | 6 | 1 | 0.17 | 5 | 1 | 0.20 |
| 12 | 70 | M | 0.25 | 12 | 6 | 0.50 | 16 | 12 | 0.75 |
| 13 | 71 | F | 0.08 | 12 | 12 | 1.00 | 16 | 16 | 1.00 |
| 14 | 72 | F | 0.14 | 8 | 7 | 0.88 | 16 | 16 | 1.00 |
| 15 | 77 | M | 0.24 | 8 | 4 | 0.50 | 16 | 16 | 1.00 |
| *Training group with magnifier* | | | | | | | | | |
| 1 | 37 | F | 0.05 | 2 | 0 | 0.00 | 0 | 0 | 0.00 |
| 2 | 39 | M | 0.08 | 3 | 0 | 0.00 | 1 | 0 | 0.00 |
| 3 | 39 | F | 0.13 | 1 | 0 | 0.00 | 1 | 0 | 0.00 |
| 4 | 49 | F | 0.08 | 6 | 1 | 0.17 | 16 | 16 | 1.00 |
| 5 | 50 | M | 0.25 | 4 | 0 | 0.00 | 9 | 8 | 0.89 |
| 6 | 52 | M | 0.11 | 6 | 0 | 0.00 | 16 | 6 | 0.38 |
| 7 | 53 | F | 0.06 | 10 | 2 | 0.20 | 16 | 15 | 0.94 |
| 8 | 53 | M | 0.16 | 1 | 0 | 0.00 | 8 | 2 | 0.25 |
| 9 | 54 | F | 0.1 | 16 | 9 | 0.56 | 16 | 15 | 0.94 |
| 10 | 54 | M | 0.2 | 2 | 0 | 0.00 | 16 | 16 | 1.00 |
| 11 | 57 | M | 0.3 | 3 | 0 | 0.00 | 10 | 10 | 1.00 |
| 12 | 58 | M | 0.25 | 8 | 0 | 0.00 | 16 | 16 | 1.00 |
| 13 | 61 | M | 0.13 | 12 | 1 | 0.08 | 16 | 16 | 1.00 |
| 14 | 63 | M | 0.16 | 16 | 7 | 0.44 | 16 | 13 | 0.81 |
| 15 | 66 | M | 0.08 | 10 | 0 | 0.00 | 16 | 10 | 0.63 |
| 16 | 67 | M | 0.16 | 6 | 5 | 0.83 | 12 | 12 | 1.00 |
| 17 | 71 | F | 0.13 | 8 | 3 | 0.38 | 16 | 16 | 1.00 |
| 18 | 75 | M | 0.08 | 10 | 3 | 0.30 | 16 | 16 | 1.00 |

[a]Values represent children's NVA in decimal notation, as given by the following (standard) formula: NVA = distance / M-value, where the threshold M-value is determined by the LH-chart[12] at the shortest distance chosen by each child. The size of the optotypes on the stimulus material was taken three steps (i.e. three lines on the LH-chart) lower than the threshold M-value for each child. Two children (no. 8 in the without-magnifier training group and no. 11 in the with-magnifier training group) worked with stimulus material that was only two steps lower, because the smallest available optotype size was 0.20M. NVA, near-visual acuity.

## Task performance: qualitative

Only five children did not improve their task performance qualitatively. One child in the without-magnifier training group already had a perfect score in the pretest and remained perfect in the post-test. Of the other four, one child was in the without-magnifier training group and three were in the group that trained with the magnifier. The number of correctly found end locations in the post-test was 0 for all four of these children. This means not only that there was no improvement, but also that their qualitative performance was the lowest possible. All of these children were younger than 3 years 6 months and, from the observations of their performance, it became clear that they had difficulties with the task. They either did not understand what was asked of them or were otherwise unable to meet the demands of the task. These four youngest children who did not progress were not included in the following analysis.

Table 2 presents several measures that reflect the quality of task performance for the two measurement times and the two training groups separately, all based on the proportion-correct scores for each individual child. Although the baseline scores appear to be different for the two training groups (0.20 with magnifier vs 0.28 without magnifier), a t-test on the arcsine-transformed proportion-correct data revealed that this difference was not significant: $t(26)=0.73$, $p=0.48$ (two-tailed). The difference between the training groups in the post-test (0.86 with magnifier vs 0.69 without magnifier) was significant, however: $t(26)=-2.06$, $p=0.05$ (two-tailed). This proved that both groups started out as equal, but that the with-magnifier training group performed better after the training period. The without-magnifier training group performed 2.5 times as well in the post-test as in the pretest (gain index 0.61), whereas the with-magnifier training group performed 4.3 times as well (gain index 0.84). Moreover, after this period more than half of the children in the with-magnifier training group attained a perfect score (53% vs 21% in the without-magnifier group).

**Table 2. Measures reflecting the qualitative task performance at the group level**

| | Proportion correct [a] | | | | Perfect scores [c] | |
|---|---|---|---|---|---|---|
| **Training group** | **pretest** | **Post-test** | **Ratio pre/post** | **Gain index[b]** | **pretest** | **Post-test** |
| Without magnifier (n=14) | 0.28 | 0.69 | 2.5 | 0.61 | 1 | 3 |
| With magnifier (n=15) | 0.20 | 0.86 | 4.3 | 0.84 | 0 | 8 |

[a] The proportion correct is given by the ratio of the number of correct end locations and the number of trails that were tried (average of only the children who had improved after training).
[b] The gain index is defined as (y-x) / (1-x), where x is the proportion correct in the pretest and y is the proportion correct in the post-test.
[c] A perfect score means that every trail that was tried resulted in the correct end location being found (i.e. proportion correct equals 1).

To analyse the changes in qualitative task performance more thoroughly, an analysis of covariance, with training group (with or without magnifier) as intersubject factor and age and pretest proportion correct as covariates, was performed on the arcsine-transformed post-test proportion-correct data. This analysis revealed a main effect for training group after correcting for initial performance level or age: $F(1,27)=5.23$, $p=0.03$.

## DISCUSSION

We studied the effect of a low-vision aid, specifically a stand magnifier, used by young children with visual impairments and the effect of specific training for the use of this magnifier. In a repeated-measures matched-groups design, children were divided into two groups to be trained either with or without the magnifier. To enable us to measure magnifier use objectively before and after the training, we developed a task that met several criteria. Importantly, the task closely resembled the dynamics of most real-life magnifier use. That is, it was designed to evoke some of the magnifier movements involved in scanning pictures or drawings and reading. The task enabled us to compare the differences in progress between the training groups.

Results demonstrated that the task provides an objective and effective instrument that can be helpful for assessing magnifier use in young children. Moreover, the training proved to have a positive effect on children's performance in the task, both quantitatively and qualitatively. Virtually all children followed a larger number of trails after the training; this was not the case, however, for the children younger than 3 years 6 months. In contrast to the number of paths followed, the quality of task performance differed significantly between the two training groups. Children who had trained with the magnifier became more effective in using it than the children who had trained without the magnifier. Both groups found a larger proportion of correct end locations, but in the with-magnifier training group this increase was about twice as large as in the without- magnifier training group.

From a clinical point of view, these are interesting results that can have important consequences for the prescription of low-vision aids to young children with a visual impairment, as well as for related training programmes. First of all, a basic finding of the present study is that our visual-attention training had a positive effect on children's performance in a demanding perceptuomotor task involving a visual aid. More specifically, this study shows that the training, which was based on the task, helps children to improve their capabilities in using the magnifier. We believe that, because of the close correspondence between the experimental task and the actual magnifier-demanding situations that children encounter, the increased effectiveness in using the magnifier can be extrapolated to their everyday practice. This study, therefore, supports the opinion that the prescription of a visual aid to children at a young age can be successful, provided that proper training is given. However, starting the present training before 3 years 6 months of age is not a good idea, although more study is needed to assess the factors that determine this minimum age.

### Magnifier use

A stand magnifier enlarges the object under its lens and provides a stable image. Nonetheless, in general, a visual aid can be beneficial to its user only once he or she has learned how to use it adequately and has learned to interpret and use the enhanced flow of visual information. This is a matter not only of the properties of the visual aid itself but also of the characteristics of the task and the user. The task that was used in this study was developed such that it could be performed successfully only when the magnifier was used properly.

Handling a stand magnifier is particularly challenging for young children with a visual impairment. Studies (e.g. Reimer et al.)[19] have shown that children with visual impairments not only have a lower level of vision, but generally show an overall impediment in their motor development as well, compared with their age-matched peers. From a social and a research point of view, this poses an interesting issue. On the one hand, because of their lower level of vision, they might benefit from the use of a magnifier. On the other hand, their poorer motor skills might render the actual use of the device difficult. This study suggests that there is progress to be made in this area of children's rehabilitation.

Finally, in a dynamic magnifier-aided task such as the one used in this study, non-visual factors play an important role in determining task performance. The visual abilities and motor-control skills of a child have to combine into a task-specific behavioural organization that enables him or her to perform the task effectively and efficiently[20]. Such a perception–action organization is what low-vision professionals often label as viewing behaviour, a term as yet without a clear definition. The quality of the viewing behaviour is what ultimately determines a child's functional vision[7]. In conclusion, more insight into the perception–action organization that underlies viewing behaviour, and its development in young children with visual impairments, is necessary.

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# Chapter 6

## Improvement of fine motor skills in children with visual impairment: An explorative study

Annemieke Reimer
Ralf Cox
Ria Nijhuis-van der Sanden
Nienke Boonstra

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**ABSTRACT**

In this study we analysed the potential spin-off of magnifier training on the fine-motor skills of visually impaired children. The fine-motor skills of 4- and 5-year-old visually impaired children were assessed using the Manual Skills test for children (6-12 years) with a Visual Impairment (ManuVis) and Movement Assessment for Children (Movement ABC), before and after receiving a 12-sessions training within a 6-weeks period. The training was designed to practice the use of a stand magnifier, as part of a larger research project on low-vision aids. In nystagmus this study, fifteen children trained with a magnifier; seven without. Sixteen children had nystagmus. In this group head orientation (ocular torticollis) was monitored. Results showed an age-related progress in children's fine-motor skills after the training, irrespective of magnifier condition: Performance speed of the ManuVis items went from 333.4 s to 273.6 s on average. Accuracy in the writing tasks also increased. Finally, for the children with nystagmus, an increase of ocular torticollis was found. These results suggest a careful reconsideration of which intervention is most adequate for enhancing perceptuomotor performance in visually impaired children: specific 'fine-motor' training or 'non-specific' visual-attention training with a magnifier.

## INTRODUCTION

### Development of Motor Coordination and Visual Impairment

Visual information plays an essential and guiding role in the planning and execution of voluntary goal-directed movements, especially during development and learning[1-4].

The adaptive behaviour that enables us to maintain a fluent relation with our environment requires a continuous informational link with this environment by means of direct sensory contact. This contact is effectuated by perceptual subsystems, action subsystems, and in particular by their mutual dependence and development[4,5]. The visual system, in particular, is part of many perception-action couplings that allow us to meet the complex demands coming from a dynamic surrounding. Perception-action couplings can loosely be defined as temporary stable, softly-assembled synergies between perception and action subsystems that are functional in specific action contexts[6,7].

Children with visual impairment are partly or completely deficient in the input of one of the vital sensory subsystems. If the visual information is incomplete or impoverish, the information necessary for action becomes more dependent on the remaining senses, and, as a result, behaviour often becomes less effective and efficient. Recently, it has been shown that there are specific differences in sensorimotor control between children with visual impairment and children with normal vision[2]. Interestingly, these differences were not all caused directly by the poorer vision per se, but seem to result from poorer calibration of the sensory information necessary for task performance. Various aspects of children's everyday behaviour (e.g., goal-directed movements and spatial orientation), as well as the general cognitive and social/emotional development, are negatively influenced by this condition[1].

The motor development of children with low vision is qualitatively and quantitatively different compared to children with normal vision. This is true for both the fine-motor skills as well as for their gross motor abilities[1,8-11]. First and foremost, visual impairment has a negative influence on their general motor activity, and may lead to developmental delays[11,12]. The impact of visual impairment is different for each phase in the development: Motor milestones are generally reached later and are sometimes even traversed in a different order. The functional development in terms of the action repertoire (i.e., the capability for solving action problems) also shows impairment-specific trajectories in children with visual impairment. The size and shape of the deviation from normative development depends, amongst other things, on the cause and severity of the child's visual impairment (anamnesis), in ways that are largely unknown[3].

Children with visual impairment experience uncertainty and insecurity with respect to the position and movements of their own body in space, of their own limbs with respect to their body, and of other people and objects in a room. This deficient spatial connection has a detrimental effect on the development of important gross-motor qualities such as adequate postural stability and control (e.g., sitting and standing), as well as on the acquisition of different modes of locomotion (e.g., crawling and walking)[13]. In addition to this, it takes them more effort to complete tasks involving fine-motor skills, such as object manipulation, object-oriented play and tool use. In a comparison study on the fine motor skills in children with visual impairment, aged six to ten, it was found that the performance was slower and more at one side of the body than in children with normal vision[14].

The developmental delays with respect to gross-motor skills and fine-motor skills might be related, because the former provide a stable platform for the development of the latter. For instance, stable erect sitting elicits a cascade of developmental processes with respect to reaching and grasping in young children, simply because the arms become available for actions at a larger radius, especially of importance when visual information is reduced[15-17]. This makes it possible to grasp nearby objects and touch surrounding surfaces, in order to actively explore them and perhaps employ them in the pursuit of more distal action goals. However, children with low vision are at risk for insufficient development of their fine-motor skills and eye-hand coordination for more direct reasons as well[18,19]. An important drawback is that, because of their poorer eyesight, these children often lack the intrinsic motivation to explore small objects and are less aware of, and as a result show little interest in, the detail-information that things possess[20]. This, of course, has a negative effect on the amount of time they spend on performing fine-motor activities typically associated with exploration and manipulation of objects, compared to their normally-sighted peers.

**Ocular Torticollis**

Congenital nystagmus (CN) has been described as a 'fixation' nystagmus, implying an inability to fixate a visual target on the fovea. However, each cycle of CN contains a target-foveation period during which the eye movement velocity is lowest. Prolongation of foveation time, reduction of retinal image velocity and cycle-to-cycle foveation repeatability all contribute to increased visual acuity[21,22]. With CN fixation of a target is difficult, which complicates the accuracy with which fine-motor tasks can be performed[23]. Despite some claims that CN is caused by absent or 'reversed' smooth pursuit, people with CN hardly ever experience oscillopsia or exhibit any accompanying symptoms of such deficits in pursuit. They are able to master sports requiring tracking of rapidly moving small objects (e.g. racquetball or handball)[22].

A large number of children with visual impairment have nystagmus, which result at the behavioural level in strategies or adaptation mechanisms in order to decrease the lack of foveation possibilities[23-25]. So as a coping mechanism, children with nystagmus may develop a functional head posture, referred to as *ocular torticolles*[24,25] which diminishes the negative effects of these tremor-like eye-movements. The direction of gaze with minimal nystagmus is called the neutral zone, which is the gaze direction that is preferred by the child[21,22]. If this neutral zone is while looking straight ahead, the child doesn't turn his or her head. If the neutral zone is eccentric, a compensatory head-turn position is taken (i.e. ocular torticollis), in order to enforce as less nystagmus as possible, fixating with the eyes in the neutral zone[26].

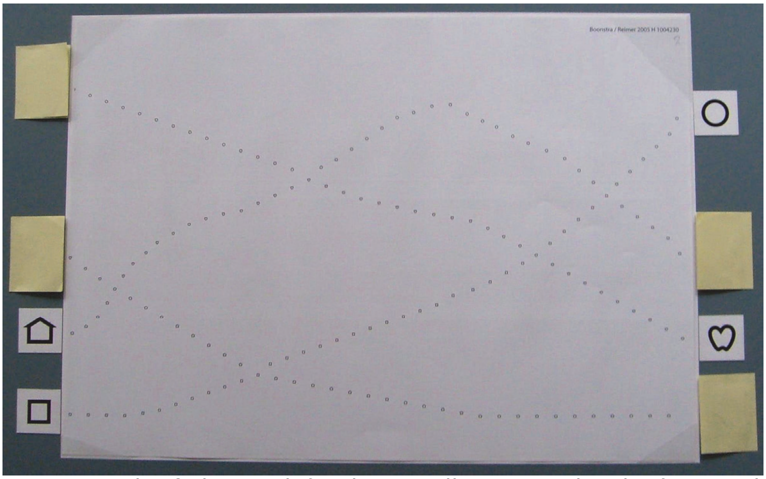
Ocular torticollis, therefore, denotes the tendency to keep the head in an oblique orientation during the performance of visually demanding activities. This is, for instance, observable in tasks with high-accuracy constraints or demands such as writing, or performing the items of a fine-motor test for that matter. It is unknown when children with nystagmus start developing this ocular torticollis. In the care of visually impaired children it is often assumed that children develop ocular torticollis when they go to school. At that time, performing high-precision tasks becomes a more prominent part of the daily routine of children. In any case, it is reasonable to suspect that there is a causal, or at least a temporal, relation between the onset of ocular torticollis and the demands on fine-motor skills.

**Earlier Study: Magnifier Training**

The present study is part of a research project funded by the Netherlands Organisation for Health Research and Development (ZonMw, program InZicht). The larger research project was a study with matched groups (matched on visual acuity and age), with the purpose of investigating the effectiveness of training with a stand magnifier in young children with a visual impairment. The results of this experimental part of the research project (i.e., on the effectiveness of the magnifier training) were reported elsewhere[20,27]. Below a concise description of the main features of the experimental design and training will be given, so as to give the reader an idea of its rationale, set-up and main results.

The training that was especially developed for this project involved 12 half-hour sessions within a six-week period. All training sessions were performed under the supervision of a personal trainer who instructed, guided and motivated the child in his/her performance of the task. The training material consisted of eight different A3-sized (42 cm x 29.7 cm) sheets, each with four trails of small symbols printed on them. An example of one such sheet that was used in the training is shown in Figure 1a. The trails crossed each other several times. Their orientation varied on different sheets (horizontal, vertical and circular), so as to induce trail-following movements in different directions. Each trail consisted of one specific type of small symbol from the LEA-optotype set[28]. The LEA-optotypes are four symbols (heart, circle, house and square) forming a standard for visual acuity testing in children[29].

**Figure 1. Materials used in the magnifier training**



(a) An example A3-sheet with four horizontally orientated trials of LEA-symbols.


(b) The stand magnifier used in the study.

Children were instructed to follow the trails meticulously, from a picture that marked its beginning to a corresponding picture marking its end. Half of the participating children trained with a stand magnifier (Figure 1b), whereas the other half used no visual aid (but instead used their finger to follow the trail during the training). Both groups performed the same task however, and had an equal number of training sessions. The effectiveness of the training with respect to magnifier use was assessed in a pre-test/post-test design. All children used the stand magnifier in the pre-test and post-test, in which they performed a similar trail-following task as during the training. The size of the symbols used in the training and pre and post tests was individually scaled, that is, based on each child's individual threshold M-value at a self chosen distance near visual acuity, as determined with a standard test[29]. If a magnifier was used (i.e. in the pre-test and post-test for all children and in half of the group during the training), the symbol size (M-value) was three logmar units below the child's threshold near visual acuity, making the use of the magnifier crucial for accurate task performance. If no magnifier was used (i.e., in half of the group during the training), the symbol size was precisely at the child's threshold M-value. Setting the symbol size for each child individually in this way ensured that performing the task was similarly demanding and difficult for all children, in both training groups. In other words, it required much visual attention and concentration, regardless of threshold M value, and regardless of whether you trained with or without the magnifier.

The results of the study showed an overall improvement of task performance across all children after the training[20,27]. For the group that trained with the magnifier the quality of task performance, as measured by the number of correctly followed trails (i.e. correctly found pictures at the end), increased more compared to the (control) group that trained without the magnifier. Importantly, the results of the study demonstrated that all children benefitted from the training.

**The present study**

In the context of the larger research project described above, children's fine-motor skills were assessed using standard instruments for manual dexterity in visually-impaired children, i.c. the ManuVis[14] and the writing task of the Movement ABC[30]. In order to enable us to detect a potential spin-off effect of the training on fine motor skills we decided to administer the test items twice; at the pre-test as well as at the post-test. Although we have not measured a control group that received no training at all, some general conclusions and interesting observations might still be possible. In the present paper the results of this repeated measurement of fine-motor skills in the participating four- and five-year-old children with visual impairment are reported for the first time. It is important to emphasize again that the training was non-specific with respect to the majority of items in the fine-motor tests, but nevertheless entailed an intensive and demanding visual-attention task.

6

Due to the intensive character of the training with respect to visual attention and eye-hand coordination, we expected an improvement in the performance of the fine-motor tests, as well as an increase in ocular torticollis in the children with nystagmus, in both training groups. There is no clear basis to expect differences between the training groups (i.e. with magnifier versus with finger), since the training is designed to be equally demanding in both groups.

## METHOD

### Subjects

A group of 8 girls and 14 boys participated in this study. They were assigned to one of the two experimental trainings groups, with or without magnifier (see Table 1). There were 15 (68 %) 4-year-old children (10 boys and 5 girls) and 7 (32 %) 5-year old children (4 boys and 3 girls). All children were visually impaired, and were selected on the basis of having a near visual acuity between 0.05 and 0.3 (Snellen equivalent: 20/400–20/67)[29]. Exclusion criteria were progressive eye disease, hemianopia, mental retardation, prematurity, dysmaturity, perinatal complications or delay in motor development. Nystagmus was present in a total of 16 of the included children (see Table 1).

Finally, the inclusion criteria required children to have a developmental level in accordance with the age-norms for this group, so as to ensure that children were able to understand the task and the instructions. This was assessed using the Reynell-Zinkin Development Scale for Young Visually Handicapped Children[31].

### *Material, Design and Procedure*

In this study children performed the bicycle trail test of the Dutch version of the Movement ABC[30] and the complete ManuVis test[14] twice. The tests were administered by or under the supervision of an experienced professional paediatric physical therapist. The validity and reliability of the Movement ABC was established by Henderson[30]. The Dutch version of the Movement ABC showed no differences in norm scores with the original norm scores[32]. The ManuVis is especially developed to measure and quantify manual dexterity of children with visual impairment in the age range of 6 to 12 years. This test is also proven valid and reliable, and has norm scores for this age range.

The fine-motor items of the ManuVis provide quantitative measurements of different aspects of motor behaviour for each test item separately (i.e. motometric data), and produce an overall score for each individual child when combined. The writing task of the ManuVis also measures movement speed, whereas the writing tasks of both ManuVis and Movement ABC allow for the assessment of the quality of performance.

**Table 1. Data of 22 children concerning diagnosis, age, sex, training (with or without magnifier), visual acuity, motometric and motoscopic data**

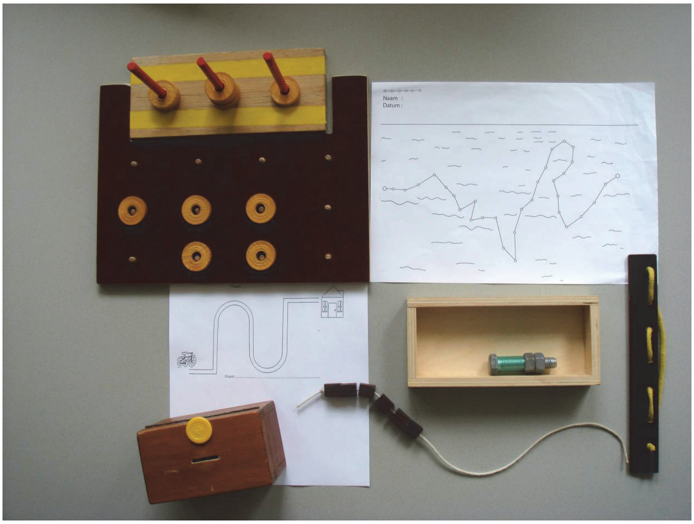
| Child Number | Diagnosis | Age | Sex | Training Group | Visual Acuity | Item 1-5 ManuVis (s) | | Motoscopic data | |
|---|---|---|---|---|---|---|---|---|---|
| | | | | | | Pre-test | Post-test | Pre-test | Post-test |
| 1 | Achromatopsia | 4:01 | F | With | 20/80 | 356 | 242 | 14 % | 42 % |
| 2 | Albinism | 4:00 | M | With | 20/100 | 360 | 312 | 16 % | 19 % |
| 3 | Albinism | 4:04 | M | With | 20/200 | 254 | 276 | 20 % | 35 % |
| 4 | Albinism | 4:06 | M | With | 20/200 | 302 | 227 | 44 % | 49 % |
| 5 | Albinism | 5:01 | M | With | 20/100 | 371 | 331 | * | * |
| 6 | Albinism | 5:03 | M | With | 20/200 | 296 | 256 | 24 % | 47 % |
| 7 | Albinism | 5:06 | M | With | 20/200 | 231 | 231 | 32 % | 43 % |
| 8 | Cong cataract | 4:07 | F | With | 20/200 | 285 | 321 | 20 % | 19 % |
| 9 | Cong cataract | 5:11 | F | With | 20/60 | 248 | 199 | 7 % | 48 % |
| 10 | Cong nystagmus | 4:02 | M | With | 20/60 | 399 | 330 | 12 % | 43 % |
| 11 | Cong nystagmus | 4:05 | M | With | 20/100 | 346 | 269 | 22 % | 43 % |
| 12 | Cong nystagmus | 4:10 | M | With | 20/100 | 197 | 184 | 34 % | 46 % |
| 13 | Retinoschizis | 5:07 | F | With | 20/60 | 285 | 260 | 27 % | 34 % |
| 14 | Albinism | 4:03 | F | Without | 20/200 | 426 | 327 | 21 % | 6 % |
| 15 | Albinism | 5:11 | F | Without | 20/200 | 235 | 245 | 26 % | 42 % |
| 16 | Retinoschizis | 4:10 | M | Without | 20/120 | 233 | 267 | 6 % | 12 % |
| 18 | Cong cataract | 4:06 | F | With | 20/60 | 231 | 227 | | |
| 21 | Retinoblastoma | 4:05 | F | With | 20/270 | 317 | 209 | | |
| 17 | Cong cataract | 4:08 | M | Without | 20/60 | 516 | 355 | | |
| 19 | Hypermetropia | 4:00 | M | Without | 20/60 | 586 | 331 | | |
| 20 | Moebius | 4:07 | M | Without | 20/50 | 539 | 408 | | |
| 22 | Retinoschizis | 5:10 | M | Without | 20/100 | 321 | 206 | | |

Data for participant 5 is missing because of technical videotape problems.

Below a description of the individual items of the tests will be given: There are two unimanual items, three bimanual items, and two items for eye-hand coordination. A picture of all the material used in the tests is shown in Figure 2.

In addition, for each item, children's head orientation and working distance (motoscopic data) was scored. These aspects of task-related behaviour were chosen for their relevance in visually impaired children[26]. Details of this scoring are also described below. All scoring was done from the videotaped sessions.

**Figure 2. Materials used in the ManuVis and the Movemement ABC: (1) Money box, (2) Wooden board with twelve rings, (3) Nuts with bolt, (4) Beads with cord, (5) Board with lace, (6) Open container, and (7) the two papers for the writing tasks**



***Unimanual items***

*Putting coins in a moneybox*. Ten coins are presented in an open container, so that their starting position is relatively fixed, and have to be picked up one-by-one and pushed into the slit of a moneybox. The task is carried out with both the left and the right hand separately, starting with the child's preferred hand.

*Putting rings on rods*. Twelve wooden rings must be placed on three vertical round rods, standing side-by-side. The rings are positioned on a wooden board in three columns of four rings, right in front of the rods. The task is to place the first ring on the first rod (on the left), the second one on the middle rod, the third on the right rod, and so on, starting with the left rod again and continuing in the same order. The rings may be picked up in any order. This task is also performed with the left hand and the right hand, starting with the preferred one.

***Bimanual items***

*Screwing nuts on bolt*. Two nuts must be attached on a bolt (nut: 1 cm. in diameter; bolt: 3 cm long) using a sort of counter-movement of the hands, and then screwed on using fingers and thumbs. The bolt is kept in one hand, while the nut is in the other hand.

*Threading beads*. Six octagonal beads (1.2 cm in diameter and 1.5 cm long) in an open container must be threaded onto a piece of cord of approximately 38 cm. The beads must be picked up one at the time, and the cord’s end must be carefully manipulated into the opening and shoved through completely.

*Threading lace*. A 40 cm long (shoe)lace is attached to the back of a wooden board (22 cm by 3 cm, and 3 mm thick). The lace must be threaded through each consecutive hole in the board, from the back to the front and back again. So, when the lace comes out through the hole on one side, it must be pushed back through again to the other side at the immediate next hole. There must be no loops over the edge of the board.

***Eye-hand coordination items***

*Drawing dots (ManuVis)*. This item requires children to carefully place dots with a felt-tip pen in small circles. There are 32 circles on a trail from the left-side to the right-side of an A4-sized paper. This item has been designed to measure eye-hand coordination, which constitutes an obvious difficulty for visually impaired children. The task quantifies the relative degree of success and difficulty of this type of coordination, by counting the correct dots and the speed.

*Bicycle trail (Movement ABC)*. Here children have to draw a single continuous line on a track, without crossing the delineations. The track is approximately 33 cm long and 0.4 cm broad, and has four alterations of movement direction; two right angles and two round curves. When the pen is lifted up from the paper, the child is allowed to continue from the same point. The paper may be rotated up to 45 degrees with respect to the edge of the table, so as to facilitate task performance. Number of crossings of the delineations is scored afterwards from the drawing, and speed is recorded as well.

***Motoscopic (quality of movements) measurement***

*Working Distance*. Children's overall working distance was scored during the performance of each of the five fine-motor items. During the one-handed items the child was not allowed to lift part of the material. She could get closer, however, by bending forward. During the two-handed items the child could lift all the material, in order to bring it closer to the eyes. Since it was not possible to reliably score working distance in centimetres, scoring was done categorically. The following categories were used: (a) larger than 20 cm, (b) 20-15 cm, (c) 15-10 cm, (d) 10 cm -5 cm, and (e) less than 5 cm.

*Ocular Torticollis*. Children's head orientations of 15 children were scored at critical instances (see below) during task performance. This was done for each of the five fine-motor items of the ManuVis, during both the pre-test and post-test measurement. For the two unimanual items the head orientation was scored for each hand separately. An observer scored whether the head was not on the midline (i.e. upright) for at least two seconds, so whether the head was either rotated or in lateroflexion (both more than 10 degrees). Lateroflexion is defined as the situation where one eye is above a certain horizontal line whereas the other eye is below that same line. Rotation is defined as the situation where the nose is left or right of the body midline.

The critical instances were pre-defined as those moments during each particular item where visual control was essential for adequate task performance: when a ring was placed on a rod (2 hands; 12 rings each), when a coin was put in the moneybox (2 hands; 10 coins each), when the bead was pushed on the cord (6 beads), when the lace was pushed in the gap of the board (6 gaps), and at the start and finish (2) of connecting the nuts onto the bolt (2 nuts). The maximum possible score of 'ocular torticollis' was 60.

### Scoring and Data Analysis

For the motoscopic measures, 20 % of video-recordings were observed by two scorers. The inter-rater reliability was at least 0.8 of each item. Preliminary analysis has shown no differences in task performance between the sexes, and the data for girls and boys have therefore been pooled in what follows. All the data were analysed in the statistical program SPSS (Version 16.0). Correlation analyses of variance and t-tests were performed on the data, using a significance level (alpha) of .05.

## RESULTS

Below we will subsequently report performance measures and statistical analyses of (1) the quantitative data of the ManuVis, (2) the writing tasks of the Movement ABC and ManuVis, and (3) the motoscopic data (i.e. working distance and ocular torticollis) during the execution of the items.

### Motometric data: ManuVis

In this section the analyses of the five fine-motor items of the ManuVis will be presented for the pre-test and the post-test. The main performance measure is the time (i.e. duration in seconds) a child needed to finish the test items, individually and combined [14]. A lower duration score corresponds to a higher item performance speed, which requires more developed fine-motor skills, and vice versa. An overview of the total-test duration score for each child (raw data; see below) as well as additional subject data is presented in Table 1, for both measurement instances separately.

The mean performance duration for each individual test item, of all children and for the children with versus without magnifier, are presented in Table 2, for the two phases in the original study[1]. (Footnote 1)

[1] Due to a procedural error in the execution of the item 'screwing nuts on bolt' made by one of the testers during the post-test, a recalculation was necessary for 36 % of the scores (i.e. eight children). The analysis presented here are with the recalculated durations for this particular item. However, when the scores for this item are excluded from the analysis, this does not lead to differences in the reported results. The total score in the pre-test without this item was 277.1 s versus a total score in the post-test of 229.5 s.

**Table 2. Mean scores (mean times in seconds (SD)) for each item of the ManuVis and the eye-hand coordination tasks of ManuVis and Movement ABC.**

| Item | Total Pre-test | Post-test | Without magnifier Pre-test | Post-test | With magnifier Pre-test | Post-test |
|---|---|---|---|---|---|---|
| Threading beads | 73.6 (36.2) | 59.5 (19.7) | 87.1 (48.3) | 69.8 (20.5) | 64.2 (22.4) | 52.5 (16.2) |
| Putting coins in moneybox | 51.4 (11.8) | 49.1 (7.6) | 51.0 (12.2) | 53.1 (7.5) | 51.6 (11.9) | 46.4 (6.7) |
| Putting rings on rods | 71.6 (26.1) | 60.0 (11.5) | 82.89 (36.8) | 59.0 (11.0) | 63.7 (11.3) | 60.7 (12.2) |
| Threading lace | 73.1 (45.4) | 57.0 (22.9) | 97.4 (54.7) | 68.9 (22.2) | 56.2 (29.3) | 48.7 (20.2) |
| Screwing nuts on bolt | 63.7 (23.6) | 47.7 (17.5) | 62.2 (16) | 51.4 (15.9) | 64.7 (28.2) | 45.08 (18.7) |
| Item 1-5 combined | 333.4 (105.9) | 273.3 (58.5) | 380.7 (138) | 302.2 (62.6) | 300.6 (63.9) | 253.3 (48.2) |
| **Writing tasks** | | | | | | |
| ManuVis Duration | 94.0 (47.4) | 70.5 (21.7 ) | 106.6 (65.8) | 66.7 (19.5) | 83.5 (6.7) | 73.3 (23.6) |
| ManuVis correct dots | 6.4 (7.0) | 12.6 (8.0) | 5.9 (5.0) | 6.7 (8.2) | 12 (5.4) | 13.1 (9.5 |
| Movement abc score | 2.3 (1.9) | 0.9 (1.2) | 3.2 (2.1) | 0.89 (1.2) | 1.7 (1.5) | 0.85 (1.2) |

Table 3 displays the correlations between item durations: The upper-right part for the pre-test and the lower-left part for the post-test.

**Table 3. Correlations between the duration scores (performance measure) on all fine-motor items in the ManuVis: The upper-right part shows the correlations in the pre-test phase, the lower-left part those for the post-test (cursive)**

| Item | Threading beads | Putting coins in moneybox | Putting rings on rods | Threading lace | Screwing nuts on bolt |
|---|---|---|---|---|---|
| Threading beads | - | .58** | .56** | .65** | .37 |
| Putting coins in moneybox | *.57*** | - | .06 | .36 | .32 |
| Putting rings on rods | *.40* | *.44** | - | .40 | .13 |
| Threading lace | *.43** | *.53** | *.40* | - | .20 |
| Screwing nuts on bolt | *.49** | *.45** | *.24* | *.32* | - |

* p < .05; ** p < .01

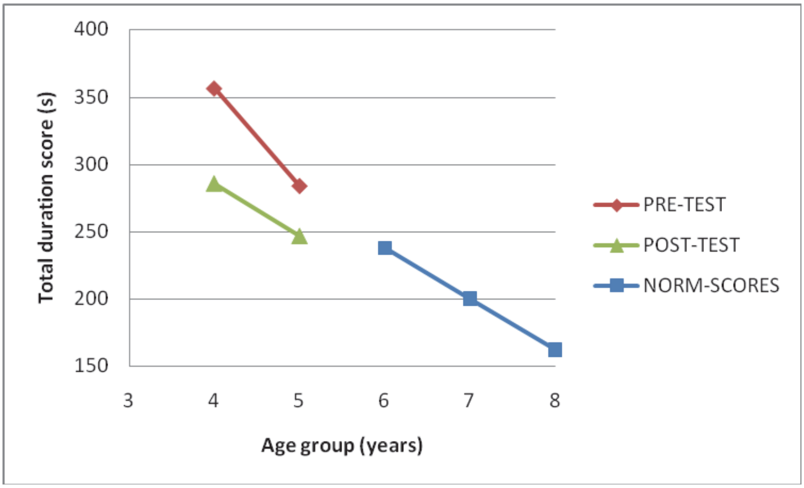
A one-factor (training group: with versus without magnifier) multivariate analysis of variance was performed on the durations scores of all five fine-motor skill items of the ManuVis, with age in months as a covariate[2]. This yielded a significant main effect, $F(5,15) = 4.31$, $p = .012$ (*Wilk's lambda* = 0.41), revealing a difference in total score on the ManuVis between the children who had trained with the magnifier compared to those who had trained without magnifier. None of the individual items were significantly different between the groups, with a Bonferroni corrected alpha-level. The same analysis performed on the item durations in the pre-test did not yield a significant difference between the groups, revealing that performance differences were not present before the start of the training.

## Total score of the ManuVis: Age differences

[2] Although group sizes were rather small and unequal, this analysis seems to be the appropriate one. A check of Box's M test revealed that the covariance matrices were equal for the dependent variables for the two groups.

The total score for the fine-motor skills part of the ManuVis for each child is given by the sum of his/her duration scores on the five individual items. This total performance duration constitutes the main measure of this assessment instrument, for which norm-scores are reported in the age range of 6- to 12-year-old children[14]. Figure 3 presents the mean total scores, that is, the total performance duration of all fine-motor items together, for the two age groups in this study.

**Figure 3.** **Graphical presentation of the mean total duration scores (mean times in seconds) of the fine-motor items of the ManuVis for different age groups. The data-points of the 6-, 7-, and 8-year-olds are published norm-scores[14]. The double data-points of the 4- and 5-year-olds come from the present study.**




From Figure 3 at least two interesting observations can be made. First, during the pre-test, older children were faster in performing the fine-motor items of the ManuVis than younger children (283.9 s versus 356.5 s, respectively; $t(20) = 2.02$, $p = .058$). This pre-test difference is to be expected since the instrument is devised to assess fine-motor skills in (young) children and monitors its changes on a developmental timescale in terms of the increase on this duration measure. Unfortunately, norm-scores for four- and five-year-olds are not reported in the literature yet. Extrapolating from the published norm-scores for older children however, we feel confident that pre-test performance of the younger children in the present study is certainly reasonable and reliable.

The difference in performance duration between older and younger children is no longer present in the post-test (246.9 s vs 285.7 s, respectively; $t(20) = 1.49$, $p = .152$). This relates to the second observation, which is that of a large reduction in performance duration in both the four-year-olds (70.8 s or 19.9 %) compared to five-year-olds (37.0 s or 13.0 %). Both age groups are faster after the training, $F(1,20) = 11.20$, $p < .003$, and the progress seems slightly larger in the younger group.

**Eye-hand coordination: Writing tasks**

In what follows we will report the analysis on the data of the two eye-hand coordination tasks performed in this study: the drawing of dots belonging to the ManuVis and the bicycle trial of the Movement ABC. Similar to the motometric data above, before analyzing the writing data some corrections are necessary in case of errors made by the child during task performance, which will be described below. Several different (corrected) results related to these items are given in Table 2, for each of the two measurement instances.

***Drawing dots (ManuVis)***

The time children needed from drawing the first dot to the last one was recorded. For each mistake, a penalty addition was made to the duration score, according to ManuVis instructions (speed-accuracy trade-off factor;[14]). After these corrections, there proved to be a significant overall difference in item performance duration between the pre-test and post-test, according to a paired-samples t-test, $t(21) = 2.47$, $p = .023$. Another measure of performance for this item is the accuracy score, which is given by the amount of correctly placed dots (i.e. exactly within the small circle; from a total of 32), and which increased after training, $t(21) = -3.49$, $p = .002$. For both measures there was no difference between the age groups or between the training groups.

***Bicycle trail (MABC)***

The prominent measure for this item is the computed error score (see Movement ABC manual[30]). Children received a score between 0 and 5, where '0' is given for perfect task performance and '5' when he/she completely failed to perform the task. The error score for the bicycle trail was significantly lower in the post-test than in the pre-test, $t(21) = 3.42$, $p = .003$. In addition to this, the time it took children to complete the bicycle trail, also decreased significantly, $t(21) = 3.39$, $p = .003$. Finally, as converging evidence of the fact that task performance has improved, we note that the mean distance over the delineation went from 4.6 cm in the pre-test to 2.0 cm in the post-test, and the mean number of occasions that the trail continued for 12 mm outside the delineation went from 1.4 to 0.5, in the pre-test and post-test, respectively. Both are an indication of increased accuracy of task performance. Again no differences were found between groups.



**Motoscopic data**

The motoscopic data gathered in this study consisted of the (overall) working distance and the (accumulated) head turns during the five fine-motor items, recorded both in the pre-test and in the post-test. Results for working distance will include all 22 children; head orientation only the 15 children with nystagmus.

### *Working Distance*

The overall working distance, averaged over all children and over all fine-motor items, showed a slight increase between the pre-test en post-test, $t(15) = 3.16$, $p = .006$. This means working distance during the pre-test was 1.4 cm (closer to 15 cm) and 1.1 cm (closer to 20 cm) during the post-test. Interestingly, there was no correlation between the mean working distance and the total score of the ManuVis. Furthermore, none of the individual fine-motor items showed an increase or decrease in working distance after the training.

### *Ocular Torticollis*

The results of the head-orientation scores in the nystagmus group are presented in Table 1. Within this group, the amount of ocular torticollis at critical instances in the task had increased significantly after the training, $t(10) = 5.11$, $p < .000$. The mean scores, of a maximum of 60, were 21.8 (36.3 %) and 40.9 (68.2 %), for the pre-test and post-test, respectively.

## DISCUSSION

The purpose of this study was to investigate the potential enhancement of fine-motor skills in young children with visual impairment after twelve sessions (six weeks) of intensive visual training. This training was carried out in the context of a larger research project. The training was especially designed to acquire and practice magnifier-using skills by means of a trail-following task. As has been reported elsewhere[20], the training yielded a considerable improvement of children's skills and willingness to use a stand magnifier, and to engage in a demanding visual attention task. The results reported in the present paper reveal a parallel improvement in children’s fine-motor skills due to the training (regardless of using a magnifier) as demonstrated by significant overall increases in performance speed and accuracy across items in the tests. In addition, during the post-test the children with nystagmus more often preferred to keep their head in an oblique orientation. This phenomenon called ocular torticollis entailed that they increased the number of head turns at critical instances during task performance, in order to utilize the neutral zone to acquire optimal fixation.

Regarding the possible mechanisms that might have contributed to the progress in fine-motor skills, it is hard to be very precise and specific. No definite statements are warranted, since no experimental manipulations were carried out for this purpose (except for the magnifier training, which was not specifically a training in fine motor skills) and the sample was too small to construct groups based on child-related factors (only partly for nystagmus). However, the prominent role of visual attention as discussed in the previous section clearly points in the direction of perceptuomotor processes underlying manual control. In an earlier study of our research group we have put forward the idea of an impoverished integration and calibration of sensory and motor subsystems in visually-impaired children[2]. Accordingly, this group experiences disadvantages in the coupling of executed (finger) movements and the visual and proprioceptive feedback coming from that. Given that the training provided a large amount of experience with respect to the coupling of sensorimotor information, this might have triggered a catch up in their learning. The precise underlying mechanisms by which this catch up was established deserve further investigation, which should focus on selecting specific subgroups of children and designing experimental manipulation based on relevant aspects of the task.

With respect to the intervention goal, it is well-known that children with low vision generally display different developmental pathways with respect to fine-motor skills, compared to normally-sighted peers. Still, to our knowledge there are no evidence-based training or intervention programs that aim at targeting these particular problems in the low-vision group. Amongst other things, this hiatus reflects a lack of knowledge about which kind of intervention is most suitable for improving manual dexterity in visually-impaired children. Professional therapists and researchers in the field must determine which perceptual-motor training is most adequate for this group: Specific training of fine-motor skills or non-specific (i.e., with respect to fine-motor skills) visual attention training. The results of the present study make an interesting and compelling case in support of the latter. Further investigation of different types of training and their combination is needed however.

To continue, the overall goal of vision training for children with visual impairment is to maximize the use of available (residual) vision so as to promote optimal development of cognitive, motor, communicative and social functioning[33]. As mentioned in the introduction, in general, these children need to be motivated and stimulated to actively explore their environment for attaining sensory (visual) information and to engage in fine-motor activities. This stimulation at an early age is very important and has a positive effect on the development of fine-motor skills[34,35]. Unless delays are detected and treated at an early age, the resulting problems in many cases are permanent. The positive outcomes of low-vision aid training for children with visual impairment are a novel feature in this field, serving two related but separate goals: improvement of magnifier-using skills and fine-motor skills.

Additionally, in this context, it is important to know whether a visual-impaired child is functioning at a satisfactory level or whether the child is delayed, compared to normally-sighted peers as well as to visually-impaired peers. Since children with visual impairment are at greater risk of less-than-optimum development of manual dexterity, it is essential to monitor these children during their development and to record their performance regularly, starting at a young age, using an objective and standardized measuring instrument. If development is delayed, the availability of a specific measuring instrument can help in the planning and evaluation of early intervention strategies for these children. It is therefore essential to develop an assessment instrument especially for very young children with low vision. Such an instrument and corresponding age-norms are currently lacking.

A major weakness of the present study is the rather small sample size. Although the results are significant and clear, generalization to the entire population of visually-impaired children is difficult. There was no control group (i.e. with no training at all), because of the limited availability of children in this target group. Further investigation, of the development of ocular torticollis in a larger sample of visually-impaired children would be valuable, for instance. The same is true for an explicit comparison of the motoscopic data during fine-motor tasks between children with and without nystagmus.

Another weakness of this study is the absence of norm-scores of the ManuVis for children of four and five years old. However, as can be seen in Figure 3, the present results of total duration scores during the pre-test measurement nicely extrapolate from the published norm-scores for six- to eight-year-olds. In addition, in a manuscript from our research group that is currently in preparation[36], norm-scores for the ManuVis will be reported for children in the age of four years and five years. The total duration scores reported here are also perfectly in line with the data already gathered for that study. Moreover, notwithstanding the current absence of norm-scores, children in this study have made an exceptional and unexpectedly large progress with respect to their fine motor skills. This positive spin-off effect of the training on fine-motor skills in this group of four- and-five-year-olds is remarkable, since the focus of the training was on magnifier use and not on aspects of manual dexterity specifically. Considering these facts, it is likely that this leap is related to the training that was done in this period, rather than being (completely) caused by natural development. Developmental impact is present of course, but after a training period of six weeks there was more progress than is to be expected.

Finally, our results emphasize the importance of visual attention and eye-hand coordination with respect to manual dexterity, and how the former can serve as a vehicle to boost the development of the latter. A recent study with kindergarteners also stresses this interaction between perceptual-motor development and attention development in young children in a different way[37]. In that study, researchers found a relationship in the other direction (i.e., fine-motor activities help increase attention), in a slightly older group of normally-sighted children (six-year-olds), and particularly in girls. Still, a relation between (visual) attention and fine-motor skills seems to be present in both studies, which offers an interesting and promising direction for future research as well as for the development of interventions.

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# Chapter 7

## General discussion

The general aim of the studies reported in the dissertation was to get an insight into the difference in the development of fine motor skills of children with visual impairment (VI) and normally sighted children (NS). As expected, it appeared that in children with VI this development differed from children with normal sight (NS). The differences in velocity and accuracy between children with VI and NS were greatest in the youngest age groups, while these differences decreased in children of older ages, but did not disappear.

Overall, children with VI performed one-handed and two-handed fine motor tasks more slowly than NS children and had more difficulty with the accuracy of goal-directed movements and prewriting tasks, and at the age of four and five years more procedural errors were made. In this chapter we reflect on the outcomes of the motor control study, the differences in fine motor skills between children with VI and NS, the results of the ManuVis-2 and the results of magnifier training for children with VI. Finally, because of the high relevance of motor skills in daily life and psychosocial participation, advice concerning prevention and intervention is presented not only with regard to fine motor skills but also with regard to ball skills and gross motor skills.

**The motor control study: goal-directed aiming movements**

In this study we used Fitts aiming tasks in normally sighted children and children with VI (mean age 8 years)[1,2]. The tasks required fast and accurate back-and-forth movements with a puppet between two predefined target areas. For this task adequate visual and proprioceptive information is necessary, which is particularly relevant at the start of the movement in order to detect information about distance and direction of movement and position of objects[3,4]. During the movements this information is necessary to control the actions in such a way that goals are reached[5]. The children performed two types of movements (discrete and cyclic), in two orientations (azimuthal and radial, i.e. along the viewing and lateral direction), and with two amplitudes (10 and 20 cm). Furthermore, the relative influence of the visual and proprioceptive subsystems on task performance was manipulated by making the target location visible during the first few trials and invisible afterwards. We found that the children with VI showed more end-point variability and less fluent movements than their NS peers. They had more problems with lateral movements than their NS peers, especially in the discrete condition and when the target was not visible. The diagnosis of children with VI participating in this study was albinism (with nystagmus). A head turn is often seen in children with nystagmus, possibly to reduce the effect of this nystagmus. Such a head turn may result in a limited lateral visual field, leading to less accuracy in the radial orientation, although this has not yet been investigated. Another explanatory factor for the difference between NS and VI children in the discrete condition could be that the radial movements rely more on visual information than on proprioceptive information[6]. The fact that the children with VI had more problems than their NS peers in the condition without vision of the target supports the hypothesis that they rely less on proprioceptive information. We supposed that several interacting factors and a lack of learning experience may be responsible for a lack of perceptual integration in the case of VI[7]. In a recent study in 2015, a similar task was used by Liebrand-Schurink[8]. The children with VI and infantile nystagmus syndrome were somewhat younger (aged 4–8 years) than in our study. They found that these younger children with VI performed the goal-directed hand movements more slowly, less accurately (especially in movements of 10 cm amplitude) and less fluently than their NS peers not only in the condition without vision of the targets but also in the condition with vision of the targets.

**ManuVis**

Valid and reliable instruments in the daily practice of rehabilitation for children with VI are scarce. We decided to expand the earlier version of the ManuVis[9] to make it reliable and applicable in clinical practice. By developing the ManuVis for children from the age of three, we wanted to provide reference scores for children with VI in order to make them ready for use in clinical settings and provide information on the fine motor skills of Ns children as well as Vi children at a preschool age[10]. Whether specific reference scores for tailored populations have additive value in clinical practice is a valid question. On the one hand, one can reason that in daily living children need to be able to perform tasks as adequately as typically developing children to increase their participation. On the other hand, it can be expected that adapting to a disability such as visual impairment will always lead to some loss in accuracy or velocity. The possibilities of compensating with other sensory and proprioceptive information will determine the magnitude of loss. Of course, we expected that fine motor skills development in children with VI would differ qualitatively as well as quantitatively from that of children with NS. Our findings suggest that in particular the younger VI children were not used to performing fine motor skills. So differences between children with VI and NS children at a younger age seemed to be at least partly the result of a lack of learning and practice. Reference scores of the group of VI children are useful in order to detect whether individual VI children are at least able to perform at the same level as their peers with VI. By monitoring development over time, combined with an in-depth anamnesis on contextual factors, timely intervention can be started to build up a playful learning environment to stimulate children to explore the use of adaptive strategies.

Only for the three-year-olds with VI were the original ManuVis tasks adapted qualitatively to make them attractive and doable. For the age bands from 4 to 11 years we decided to use the same items to avoid an increasing load on visual perception and to increase longitudinal comparability from age 4 to age 11[11]. In this study we compared VI and NS children and sampled norm references for VI children.

A leading rationale was that both simple and complex one-handed and two-handed and prewriting skills needed to be present in the test. We discussed the possibility of floor effects, but we judged that it would be an advantage if the task load on the visual system were comparable in different age groups. The results showed clearly that for the item *putting coins into a moneybox* the floor effect was most prominent. This hinders the validity of the test outcomes, especially at older ages. The results showed that performance increased in older children with VI. So apparently they are able to increase velocity and accuracy in simple fine motor tasks, but they seem to need more experience over time. The cross-sectional research design was not adequate to test the hypothesized relationship between training experience and outcomes, so in future more longitudinal training studies are necessary to get an insight into the influence of training on performance. Not only in the item *putting coins into a moneybox* were floor effects observed, but also in the other tasks, starting at different ages, which seemed to be dependent on the task difficulty. Therefore we must conclude that choosing the same set of items over the age groups is a weakness of the ManuVis study, because these floor effects influence diagnostic accuracy in the older children. Therefore in future research, tasks with greater difficulty and complexity should be included. Moreover, it would be of added value to sample data longitudinally over time instead of cross-sectionally to strengthen the insight into developmental trajectories. Because such studies are difficult to organize, we advise that centres for visually impaired children sample data over time in a systematic and standardized way in daily clinical practice.

**Prevention and intervention: what do the results of the magnifier training tell us?**
Prevention of fine motor problems can facilitate a normal start in school and can reduce the problems that children with VI experience in daily life. It is recommended that children with VI are referred to a developmental centre as soon as severe VI is suspected[12]. Preschool teachers of children with VI reported a comparable number of problems in activities involving looking at picture books, fine motor skills (handicrafts) and cognitive play (puzzles)[13]. Students with low vision had poorer handwriting performance, with lower legibility and slower writing speed[14]. Children with VI require more effort to learn fine and gross motor skills, not only as a result of lacking visual information but also due to less motor skills practice. The overall goal of training in, for instance, magnifier use for children with VI is twofold: 1) to maximize the use of available (residual) vision, and 2) to develop adaptive strategies to achieve optimal development of cognitive, motor, communicative and social functioning[15].

In this thesis an intervention is described for young children with VI involving a task with a magnifier[16,17]. In rehabilitation practice magnifiers are advised to facilitate the acquisition, and to increase the quality, of visual information. In our study we used the magnifier in visual tracking tasks, however we also found that fine motor skills performance improved. The results of this magnifier training study in Chapter 6 are promising. In this study it was shown that magnifier training for 6 weeks not only led to a specific effect – e.g. trail finding or making fewer faults at crossings – but also to an unspecific or 'spin-off' effect, namely a fast increase in fine motor skills in a relatively short time frame independently of whether the magnifier was used during the training. The result of this fine motor skill achievement can therefore not be related to the magnifier use, but it can be related to trail-following movements over 6 weeks and possibly more indirectly also to the development of viewing behaviour. This means that training should not only be focused on adaptive strategies but also on using the visual capacity as much as possible.

However, another 'spin-off' effect of this training was a significant increase in torticollis after 6 weeks' training in 15 children with nystagmus. This increase probably shows a rise in the use of visual capacity and possibilities leading to a (not desirable) adaptation, because torticollis may reduce the nystagmus amplitude and thereby the visual acuity[14].

Apparently, intensive perceptuo-motor training in a structured and challenging visual environment had a positive transfer effect on all fine motor activities. This raises the question of whether 'specific' fine motor training or 'non-specific' visual-attention and perception-action training or a combination of both is most appropriate in interventions. In another study, a relationship between (visual) attention and fine motor performance was also found[18]. In this study, training of fine motor activities increased visual attention in a slightly older group of normally sighted children (six-year-olds). Apparently, there is a strong relationship between the visual, perceptual and motor systems, and it is important to train both factors.

A young child with VI can profit from the use of a low-vision aid, but needs to be stimulated to explore and experience the additive value[19]. Such a magnifier can also be helpful when children look in picture books or start reading. In our study, we found that after the training the children were familiar with a magnifier, and its use in daily life and in a classroom was encouraged. However, a magnifier is not the only useful attribute for children with VI. Currently, smartphones, tablets and computers are also considered indispensable for children with VI. The opportunities, for example, with a computer or tablet to enlarge the font size or zoom in on pictures are very helpful for those children with VI. The use of these devices needs a considerable amount of fine motor skills and therefore VI children require training to use them in an appropriate way.

### Psychosocial effects of visual impairment

The influence of visual impairment is described from various viewpoints such as development, parenting, education and support[20]. The relationships between visual impairment and orientation, mobility and motor performance and social integration are described by Sleeuwenhoek[21]. Children with VI have to make more effort to learn activities of daily living[22] such as dressing, washing, eating, mobility and moving into unfamiliar places or using public transportation. Lewis and Iselin found that parents of children with VI (ages 6–9) reported that their children performed only 44% of daily-living tasks independently, while parents of typically developing peers reported that 84% of such tasks were performed independently. At school, the same difference was found: students who were blind or who had low vision were not able to perform 41% of the tasks, even with assistance, while their peers only needed help in 14.5% of the tasks[23]. Moreover, children with VI have more difficulty making appropriate comments and ending conversations appropriately, because they miss parts of the non-verbal communication. Finally, the degree of visual impairment may be a factor in determining self-esteem[24], and there is an interaction between motor development and self-esteem[25].

For parents it is difficult to estimate what a child can or cannot see, and this sometimes may lead to over- or underestimation of their ability to participate in activities. For parents it is important to understand the impact of VI on communication, self-esteem and attitudes[26]. Research showed that the educational level of parents is a predictor of developmental delay in communication, reading ability and socialization: the higher the educational level of parents, the better the performance and the lower the developmental delay[27,28]. Brown et al. summarized factors that may contribute to decreased movement and exploration in children with VI and identified strategies to facilitate movement and exploration[29] that may guide the goals and interventions in a multidisciplinary team.

### *Paediatric physical therapy assessment in children with VI*

The paediatric physical therapy assessment of children with VI should be focused on the identification of questions for help and problems in daily living and participation, characteristics of the visual impairment, and testing the level of motor skills performance. The Hypothesis-Oriented Algorithm for Clinicians II (HOAC-II) model provides a structure to identify the complex factors that play a role in children with VI and supports the paediatric physical therapy decision process[30].

### Measurement instruments adapted for children with VI

For young children up to 42 months, the Bayley Scales of Infant Development (BSID-III) are adapted for children with VI[31,32]. This test can be used for testing both cognitive and motor development. For fine motor skills the ManuVis-2 (3–11 years) is now available. At the time we started developing the ManuVis we partly selected items from the Movement ABC (MABC-1), which was available for ages 4–12 years. However, recently a second edition (MABC-2) was developed for the age range 3–16 years[33]. A lot of research has started worldwide to sample recent reference data for the MABC-2, however as far as we know no version for children with VI is being developed. The KTK test[34] is useful for testing gross motor skills in children from 5 to 14 years. For children with VI, research on this test was performed[35]. In order to carry out the subtests with limited sight in a correct way and as safely as possible, small adaptations have been made with respect to the test material. Also, this study pointed out that motor development in children with VI follows a different pathway to that of NS children, so specific reference scores would be valuable. The Test of Gross Motor Development[36] (TGMD-2) for the age range 3–10 years covers locomotor and object control skills. Research with the TGMD-2 pointed out that this test is also useful for children with VI [37,38]. The Paediatric Evaluation of Disability Inventory (PEDI)[39] (6 months–7.5 years) can be used as a parent report/structured interview instrument or as an observational instrument by professionals in a hospital, outpatient or educational setting to get insight into the domains functional skills/behaviours in daily life and to get an overview of the necessity of carer assistance and the need for environmental modifications and extra equipment. It would be advisable to adapt the PEDI as an instrument for standardized history taking in children with VI, which would be helpful for early detection of questions for help. The reliability of the modified Paediatric Evaluation of Disability Inventory (PEDI-NL)[40] for children with cerebral palsy and cerebral visual impairment has already been examined[41]. The Sensory Profile[42] (0–10 years) provides a standard method for measuring an infant's sensory processing abilities in everyday situations. This instrument could be helpful for testing systematically the sensory development of a child with VI, which helps in tailoring the interventions more systematically. A pilot study has already been performed with additional items for children with VI (3–10 years)[43].

**Indication for paediatric physical therapy assessment/intervention**

With low vision there is basically no immediate indication for referral to a paediatric physiotherapist. However, if there are problems in the domains of activity and participation, such as self-care, mobility, posture and movement insecurity in everyday life and/or at school during physical education and writing education, there is an indication for paediatric physical therapy assessment. In the anamnesis, data are sampled on the type and degree of visual impairment and other medical data. Information regarding parents, teachers and other people who are involved in the education of the child gives insight into the developmental trajectory and problem-solving behaviour of the child and the participation at school, in playing with peers and in sports or music. Based on the test outcomes using the above-mentioned measurement instruments, the observation of spontaneous motor activity and the information from the anamnesis, a decision will be made as to whether it is useful to train specific aspects of motor skills.

If the assessment indicates a delay in motor development or a risk of delay, paediatric intervention is indicated. Especially in visual impairment and a specific delay in motor development, physical therapy can be meaningful. If the motor development is harmonious and age adequate, it is sufficient to advise the parents in the form of a low-frequency monitoring.

**Physical education and participation in sports**

The level of gross motor skills performance of children with VI is lower than that of NS children[35,44]. Walking speed is lower, and there is a shorter stride length and a prolonged duration of stance and of double support. There are more adaptations for balance or strategies to allow the foot to probe the ground[45]. Intentional and specific instruction on motor skills at a younger age may be needed to enable the development of gross motor skills[46]. Parents and children should have more information about the benefits of, and opportunities for, physical activity[47-49]. Proper rehabilitation in order to learn to use the available vision can improve the level of motor skills[50]. Studies of body mass index (BMI) show that sufficient exercise should be stimulated in visually impaired and blind children[51]. BMI seems to be more elevated for both groups than in NS children. School-aged children with VI are less physically active than their peers without VI[52]. In a video surveillance with a pedometer at the Bartiméus school it was found that blind children and adolescents were less active than those with low vision. Promoting an active lifestyle in children with VI[53] has a positive influence on body image and BMI for children and adolescents with VI, thus it should be encouraged. Most children with VI need extra support and encouragement to increase mobility in different environments, for example independent shopping, walking or cycling in traffic and using public transport. For children with hemianopia, specific (pilot) training is developed to help them orientate themselves in different environments.

Every boy wants to learn to play football and many children are dropping out again. For a child with VI it may be a good experience to discover the sports in which they can participate fully. Appropriate sports for children with VI include athletics, guide running, gymnastics, fitness, powerlifting, swimming, skating, inline skating, skiing, judo, wrestling, horse riding, tandem cycling, rowing, showdown, goalball, golf, sailing, windsurfing, climbing and dancing. For each sport there are specific modifications or suggestions for training and competition.

Ball skills are important in order to be able to participate in the schoolyard and in many sports. It is plausible that children with VI require a lot of practice for skills learning and need to use adaptive strategies, for instance in goal-directed aiming. An intervention with children with learning disorders (aged 7–11) improved their ball skills significantly after six weeks' training[54]. Catching a ball, for the child with VI, will be more complicated[55], because visus is crucial in estimating the speed and size of the ball. However, participating in ball games is not impossible for children with VI and they give them a lot of fun. Prior to and during a ball game the child with VI benefits from extra information: for example, by adding a sound to the ball or to the playing rules (first bouncing then catching), or allowing them to discover how big the playing field is. During ball sports for children with VI it is relevant that the ball has a contrasting colour with the environment and that the team wears clothes with a contrasting colour to guarantee recognizability. Sports such as showdown and goalball are suitable.

**Suggestions for future research**

By studying the differences in (fine) motor skills between NS and VI children in different experimental and daily settings, large differences were detected, however studies on interventions are scarce. To develop an intervention that will reduce the gap between children with VI and children with NS as much as possible, an insight into the adaptive strategies is necessary as well as an insight into the technical support that will enhance such adaptations.

For future research a longitudinal research design monitoring the development of fine motor skills in individual children with VI is advisable, in order to gain more insight into individual developmental trajectories and to study the influence of personal and contextual factors. This will help us to understand why the inter-individual variability in the fine motor development of children with VI is so large, as well as the extent of the intra-individual variability in this group. Ideally such a set-up for research on fine motor skills could be combined with research of gross motor skills and an appropriate pupil-monitoring system for teachers.

Additionally, it is advisable to collect more ManuVis data about children with VI longitudinally to increase the reliability of the norm scores of the fine motor skills for each age band and also to compare these norm scores with the scores of NS children. For the items that showed a floor effect, another item may be developed to replace them. Moreover, increasing the task difficulty is advisable for some of the test items for children aged 10 and 11 years, and possibly for older children who were not part of the sample in this dissertation. For example, the item *putting coins in a moneybox* might be replaced by, for example, the item turning pegs. Future research should also focus on further connections of the ManuVis-2 with other measurement instruments for children with VI, such as the MABC-2. Further research with the Sensory Profile instrument[42] is also recommended.

To conclude, I think it is worthwhile carrying out investigations such as the one presented in this thesis and communicating the results to a wide audience, for the benefit of children with VI. It is important that in the future, (fine) motor assessment, as well as prevention and intervention, in children with VI should be based on adequate reference scores. More research should be performed on the relation between a child's poor eyesight and (fine motor) performance.

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# Summary

The central research questions of this thesis were: What is the influence of visual impairment on the performance of goal-directed movements? What is the level of the fine motor skills of children with visual impairment (VI) in comparison to children with normal sight (NS)? Is the ManuVis-2 a reliable instrument for discriminating between children with and without fine motor performance problems within the group of children with VI? Moreover, what would be the effect of magnifier training on fine motor skills in children with VI?

In Chapter 1 the characteristics of visual impairment and the consequences on the perceptuo-motor development are described. If the visual information is incomplete or impoverished, the information necessary for action becomes more dependent on other sensory information: tactile, auditory, vestibular, proprioceptive information and the sense of smell. Because of the importance of visual information for motor skills performance, visual impairment will have consequences for motor learning and motor performance. To be able to start the intervention and treatment of sensorimotor impairments as early as possible, research on typical motor development in children with VI is necessary. Moreover, we need reliable and valid tests with norm-referenced scores to determine whether there is a motor developmental delay in individual children with VI by comparing them with their peers.

In Chapter 2 we describe differences in the performance of goal-directed aiming between children with VI and children with normal sight (NS). For this, an experiment was carried out in which children performed repeated goal-directed aiming movements with a puppet between two targets that were both visible, and in the next experiment the ultimate targets were not visible anymore. Participants had to move the puppet over two distances, 10 cm and 20 cm, between the starting circle and target circle, in two orientation conditions (azimuthal and radial) and in a discrete and continuous mode. Discrete movements and movements over large distances were less fluent in both groups, but especially in the children with visual impairment. The results showed that the children with VI were less accurate and were slower in performance when the target was not visible anymore, specifically along the lateral direction and in the discrete condition. We discussed whether this was due to the reduced vision or to a lack of moving experience in children with VI. If the latter hypothesis is true, more practice would be needed in order to calibrate the movement to the non-visible target.

In Chapter 3 we describe the differences in the performance of fine motor skills between children with VI and NS in the age range from 4 to 11 years using the ManuVis-2. The children with VI needed significantly more time than NS children to perform all test items. Moreover, the writing tasks were performed with less accuracy. The differences between children with VI and NS were larger at younger ages. Performance

time significantly decreased in both groups of children from the younger to the older age groups in all tasks, while an interaction effect indicated that differences between groups were largest in the younger age groups. Also, children with VI aged 4 and 5 years made more procedural errors. The results suggest that children with VI apparently need more time to learn strategies adapted to their VI in order to perform fine motor skills in an adequate way. The decrease in movement time seems to be the effect of motor learning. In the more complex unimanual and bimanual tasks, it was found that the curves were steeper in children with VI. Visual information at the end of a movement sequence seems to play an important role. Moreover, we found that test-retest reliability for the ManuVis items varied from moderate to excellent and inter-rater reliability was excellent. The ManuVis differentiates between typical and atypical fine motor performance of children with VI between 4 and 9 years of age, and is useful for monitoring fine motor skills development in children with VI aged 4 to (at least) 11 years. Moreover, reference values are provided to enable determination, in individual children with VI, of whether motor skills performance is deviant compared to their peers with VI.

In Chapter 4 we compare the fine motor performance of three-year-olds with VI with three-year-olds with NS and provided reference scores of the ManuVis with items adapted for this age group. Also, at the age of 3 years children with VI needed more time on all tasks and performed the prewriting task less accurately than children with NS. Children aged 42–47 months performed significantly faster in two items and had better total scores than children aged 36–41 months. Moreover, it appeared that some children with VI aged 36–41 months could not perform two items yet (i.e. *screwing nut onto a bolt* and *threading beads*). Children with VI of this age probably have less incentive to practise fine motor skills and prewriting tasks.

In Chapter 5 we test the effect of magnifier training. A stand magnifier enlarges the object under the lens and provides a stable image; however, handling a stand magnifier is challenging for young children with VI. Several studies recommend the use of low-vision aids for young children with VI and a theoretical analysis by Schurink et al. in 2012 showed potential positive effects. In the experiment in this chapter children with VI had to follow trails visually, from a start location to an end location, with or without a stand magnifier. The results showed that there was an improvement in task performance after training with or without the magnifier, except for the youngest children. The with-magnifier training group had significantly higher scores in finding the correct end location and it was advised that young children should be trained to work with a magnifier.

In Chapter 6 we report the spin-off effect on fine motor skills performance of the study mentioned in Chapter 5. Results showed age-related progress in children's fine motor skills after the training, irrespective of the magnifier condition (i.e. training with or without the magnifier). It was shown that if children with VI have more experience at focusing visual attention during perceptuo-motor performance, this has a very positive effect on the speed with which they can perform fine motor tasks. The older children performed the items faster than the younger ones, but in this study we found an improvement in fine motor skills already after six weeks that would otherwise take about one year on average. For clinical practice this means that magnifier training around the age of 4 years is a valuable intervention for improving fine motor skills.

Chapter 7 provides a reflection on the overall findings of the studies. Extra attention to the fine motor functioning at a young age is an important issue. The additive value of tailored advice about toys, a playful learning environment and specific interventions taking into account individual needs was discussed. All study results underline the point that the sensorimotor development of children with VI requires extra attention. The identification of sensorimotor problems at the earliest possible age seems to be important to avoid developmental delay due to a lack of adequate practice. Extra integrated and tailored support to allow the development in various fields is essential. Further longitudinal research on fine motor skills should be combined with research on gross motor skills in early childhood to develop standard values for children with VI, and an appropriate pupil monitoring system for healthcare professionals and teachers is recommended.

# Samenvatting

Het belangrijkste doel van dit hoofdstuk is om professionals die werken met kinderen met een visuele beperking inzicht te geven in de uitkomsten van mijn onderzoek. De centrale vraag in dit proefschrift is in hoeverre slechtziendheid het fijn-motorisch functioneren beïnvloedt. We hebben daarbij specifiek gekeken naar de uitvoering van doelgerichte bewegingen en naar het niveau van het fijn motorisch functioneren van slechtziende kinderen in vergelijking met normaalziende kinderen. Daarnaast hebben we onderzocht of de ManuVis-2 een betrouwbaar en valide instrument is om bij kinderen met een visuele beperking vast te stellen of er sprake is van motorische problemen op het gebied van de fijne motoriek in vergelijking met leeftijdgenoten. In een interventie studie hebben we gekeken welke voordelen het gebruik van een loep kan hebben. Tenslotte zal aandacht besteedt worden aan de betekenis van de uitkomsten voor de begeleiding van slechtziende kinderen. Vanaf de geboorte zullen slechtziende kinderen minder gebruik maken van de mogelijkheid tot imitatie. Visueel worden zij minder uitgelokt om te bewegen, de omgeving te exploreren en om speelgoed te gaan onderzoeken. Doordat zij minder spelen met diverse materialen zal dit het begrip van voorwerpen (conceptherkenning) en de spelontwikkeling negatief beïnvloeden en verder zullen de ADL vaardigheden en schoolse vaardigheden extra aandacht vragen.

In hoofdstuk 1 wordt beschreven wat slechtziendheid is en welke gevolgen dit heeft voor de verschillende ontwikkelingsdomeinen. Indien de visuele informatie onvolledig is, zal de informatie die nodig is om in actie te komen meer afhankelijk zijn van de informatie van andere zintuigen: tactiel, auditief en vestibulair en proprioceptieve informatie en geur zal meer betekenis hebben. Onderzoek van de motorische ontwikkeling en bepalen of deze ontwikkeling vertraagd is of anders verloopt in vergelijking met hun slechtziende leeftijdsgenoten is belangrijk. Indien er sprake is van een vertraging dan is het is belangrijk om een passende interventie te geven. Interventie en behandeling van zintuiglijke beperkingen moet voor een optimaal effect bij voorkeur op een jonge leeftijd beginnen.

In hoofdstuk 2 was het doel te weten te komen of de wijze waarop een beweging verloopt, verschilt tussen de slechtziende kinderen ten opzichte van normaalziende kinderen. Hiervoor werd een bewegingstaak uitgevoerd met een cilinder. De cilinder werd bewogen over twee afstanden: 10 en 20 cm., in twee richtingen (horizontaal en verticaal) en op discrete c.q. continue wijze tussen twee doelen die eerst beide zichtbaar waren en later zonder dat het einddoel zichtbaar was. Het uitvoeren van discrete bewegingen en bewegingen over de grootste afstand verliepen minder vloeiend bij beide groepen, maar vooral bij de slechtziende kinderen. Verder bleek dat de slechtziende kinderen meer moeite hadden om indien het doel niet zichtbaar was accuraat deze beweging uit te voeren in het bijzonder wanneer de beweging naar lateraal (van het lichaam af) was. De vraag die ontstond was of dit kwam door de visus beperking of doordat de slechtziende kinderen minder bewegingservaring hebben en meer oefening nodig hebben om een dergelijk niet zichtbaar doel vast te leggen in hun geheugen.

De hoofdstukken 3 en 4 zijn gewijd aan het onderzoek van de fijne motoriek bij kinderen van 3 tot 11 jaar. Er is gebleken dat slechtziende kinderen bij alle opdrachten trager zijn en meer moeite hebben met de accuratesse.

In hoofdstuk 3 beschrijven we de verschillen in uitvoering van de fijne motoriek tussen slechtziende en normaalziende kinderen in de leeftijd van 4 tot 11 jaar met behulp van de ManuVis-2. De slechtziende kinderen hebben aanzienlijk meer tijd nodig dan normaalziende kinderen om alle test items uit te voeren. Bovendien werd de schrijf taak met minder nauwkeurigheid uitgevoerd, vooral op jongere leeftijd. De uitvoering per opdracht verminderde qua tijdsduur in beide groepen kinderen bij toename van de leeftijd. De verschillen tussen de groepen waren het grootst in de jongste leeftijdsgroepen. Ook maakten slechtziende kinderen van vier en vijf jaar meer procedurefouten. De resultaten suggereren dat slechtziende kinderen blijkbaar meer tijd nodig hebben om adaptatiestrategieën te leren om de fijne motoriek op een adequate wijze uit te voeren. De afname van bewegingstijd is het effect van motorisch leren. In de complexere eenhandige en tweehandige taken, bleek dat de curves steiler waren bij slechtziende kinderen. Vooral als visuele informatie aan het einde van de beweging een belangrijke rol speelt. Bovendien hebben we gekeken of de gebruikte ManuVis test ook een betrouwbaar en valide instrument is om de fijne motoriek te meten. De test-hertest en inter-beoordelaarsbetrouwbaarheid van de ManuVis test-items varieerde van voldoende tot excellent. De ManuVis test is in staat een atypische fijn-motorische ontwikkeling te meten bij kinderen tussen 4 en 9 jaar en kan gebruikt worden om de ontwikkeling over de tijd heen te monitoren bij slechtziende kinderen tot (minimaal) 11 jaar. Daarnaast zijn referentie waarden verzameld om bij individuele kinderen te diagnosticeren of de fijne motoriek afwijkt.

In hoofdstuk 4 vergelijken we de fijne motoriek van slechtziende en normaalziende kinderen van drie jaar en worden er referentie scores gepresenteerd van het onderzoek met behulp van de ManuVis. Slechtziende kinderen hebben meer tijd nodig voor alle taken en zij voerden de voorbereidende schrijfopdrachten minder nauwkeurig uit dan de normaalziende kinderen. Kinderen van 42-47 maanden voerden twee taken aanzienlijk sneller uit en hadden betere totale scores dan de kinderen van 36-41 maanden. Slechtziende kinderen van 36-41 maanden konden sommige taken (schroeven van een moer op een bout en kralen rijgen) nog niet uitvoeren. Verder zijn

slechtziende kinderen van deze leeftijd waarschijnlijk minder geneigd om bezig te zijn met teken- en schrijftaken.
Voor de praktijk betekent dit dat bij het vaststellen van het niveau van fijn motorisch functioneren van slechtziende kinderen aangetoond is dat zij hiervoor vergeleken dienen te worden met slechtziende leeftijdsgenoten.

In hoofdstuk 5 hebben we onderzocht of het gebruik van een loep door jonge kinderen toegevoegde waarde heeft. Een loep vergroot het object onder de lens en zorgt voor een stabiel beeld. Omgaan met een loep is echter wel een uitdaging voor jonge slechtziende kinderen. Verschillende studies raden het gebruik van low-vision hulpmiddelen aan voor jonge slechtziende kinderen en een review van Schurink in 2012 geeft aan dat dit aan te bevelen zou zijn. In het experiment in dit proefschrift kregen slechtziende kinderen de opdracht om sporen visueel te volgen, van begin tot eindlocatie, met of zonder loep. De resultaten toonden aan dat de prestatie verbeterden na de training met of zonder loep, behalve bij de jongste kinderen. De trainingsgroep die de opdrachten met een loep uitvoerde, had significant hogere scores in het vinden van de juiste eindlocatie en er werd geadviseerd om jonge kinderen te trainen om met een loep te werken.

In hoofdstuk 6 wordt de potentiële spin-off van een loep training op de fijne motoriek van slechtziende kinderen beschreven. De resultaten toonden een leeftijdsgebonden vooruitgang aan bij kinderen wat betreft de fijne motoriek na de training, ongeacht of zij wel of niet een loep gebruikt hadden. Het bleek dat slechtziende kinderen na deze training door het accent op de visuele aandacht in de training ook sneller fijne motorische taken konden uitvoeren. Op zich gaan kinderen sneller werken met toename van de leeftijd, maar nu werd er na zes weken een vooruitgang vastgesteld die anders pas gemiddeld na één jaar bereikt wordt. Voor de praktijk betekent dit dat de loeptraining rond de leeftijd van 4 jaar een waardevolle interventie is, indien er een achterstand is op het gebied van fijn motorisch functioneren.

Hoofdstuk 7 geeft een reflectie op de algemene bevindingen van de studies. Extra aandacht voor het fijn motorisch functioneren op jonge leeftijd is belangrijk. Gericht speelgoed advies en interventie is waardevol omdat uit de studies bleek dat motorische problemen deels ook een gevolg waren van te weinig leerervaring. Geconcludeerd kan worden dat de motorische ontwikkeling van slechtziende kinderen extra stimulans vraagt. Het vaststellen van motorische problemen op een zo vroeg mogelijke leeftijd is dus belangrijk en tevens is het goed de motorische ontwikkeling in de tijd goed te monitoren. Extra ondersteuning in gang zetten ten behoeve van de sensomotorische ontwikkeling is mogelijk essentieel. Vervolgonderzoek van de fijne motoriek aangevuld met actuele motorische testen van de algemene motorische ontwikkeling en een leerlingvolgsysteem voor leerkrachten om normwaardes te ontwikkelen voor slechtziende kinderen is aan te raden. Onderzoek van het gebruik van APP's voor het leren lezen en schrijven bij slechtziende kinderen is tevens de moeite waard om te onderzoeken.

# Dankwoord

Als eerste wil ik mijn partner Hans Wisman bedanken voor alle geduld en hulp. Zoveel steun en compassie is fijn om te hebben gedurende zo'n lange periode.

` Vanuit de eerste lijn begon ik zo'n 36 jaar terug te werken met slechtziende/blinde kinderen van Bartiméus. Toen bestond er nog geen opleiding kinderfysiotherapie. Om voldoende kennis te vergaren ging ik meelopen op revalidatie centra en bij collega's die met kinderen werkten en bijvoorbeeld de Oseretsky test konden afnemen. Om deze kinderen adequaat te kunnen behandelen was er bijscholing nodig op neurologisch gebied. Met een urgentieverklaring van Professor Dr. Paul Helders en de toenmalige huisarts Dr. Pim Bergman van Bartiméus duurde het 3 jaar voordat ik eindelijk geplaatst kon worden op de Bobath cursus. In eerste instantie kwamen de kinderen van Bartiméus onder begeleiding naar de fysiotherapiepraktijk op de Steniaweg toe, waarbij er contact met de groepsleiding iedere keer aanwezig was. Na verandering van de praktijksituatie enige jaren later werden de werkzaamheden voortgezet op het terrein van Bartiméus en nam het directe contact met de groepsleiding af, omdat de kinderen zelfstandig naar de oefenruimte toe konden komen. Er ontstonden overlegmomenten met de huisarts, gedragsdeskundigen en de groepsleiding. Deze momenten namen steeds meer tijd in beslag en er werd mij gevraagd door AVG arts Dr. Riet Niezen om een loondienstsituatie te overwegen. Eenmaal in loondienst van Bartiméus, bij de afdeling medische dienst, ging ik veel kinderen zien voor onderzoek en advies. Als enige in loondienst zijnde kinderfysiotherapeut heb je kans om veel kennis te vergaren, over te dragen en te delen. Veel contacten zijn er steeds met de oogheelkundige afdeling, de gedragsdeskundigen, maatschappelijk werk, de ambulante medewerkers, de leerkrachten en de groepsleiding. Maar het meest boeiend zijn toch steeds de kinderen. Na het uitvoeren van een kinderfysiotherapeutisch onderzoek bleef het een zoektocht naar de invloed van de visus op het motorisch functioneren.

Door het volgen van nascholing lag het op mijn weg om zoveel mogelijk evidence based te werken. Er werden aanvragen ingediend om wetenschappelijk onderzoek uit te voeren bij de kinderen van Bartiméus. De voorstellen om de algemeen motorische en de fijne motorische ontwikkeling bij een aantal kinderen te gaan onderzoeken was snel geregeld. Door bij de bestuursleden, Johan Gerritsen en Paula van Woudenberg, binnen te lopen en de onderzoeksvoorstellen aan hen voor te leggen. Er werd voor de eerste onderzoeken van de fijne motoriek door hen telefonisch contact gelegd met de andere instituten, Visio en Sensis. De ethische commissie bestond nog niet en een toestemmingsverklaring met de ouders werd telefonisch geregeld. In ieder geval wil ik beiden bedanken voor de mogelijkheden die ze mij toen gaven.

Tijdens het volgen van de MDB cursus bij Dr. Bouwien Smits–Engelsman en leerde ik kinderfysiotherapeut Marieke Siemonsma-Boom kennen. Met Marieke heb ik samen een aantal onderzoeken bij blinde en slechtziende kinderen uitgevoerd, bij voorbeeld de KTK test voor het algemeen motorisch functioneren. Bouwien was aangenaam verrast met deze eindopdracht als afsluiting van de cursus. Dit resulteerde in een werkstuk en het eerste artikel over het algemeen motorisch functioneren bij deze doelgroep. kinderen. Het artikel werd tevens gelezen door Professor Dr. Paul Helders. Hierna gingen Bouwien, Marieke en ik verder bedenken wat we wilden gaan onderzoeken en we kwamen toen uit op onderzoek van de fijne motoriek. Met Marieke samen heb ik deze onderzoeken uitgevoerd. Dit resulteerde in een artikel, testkoffer en een handleiding: ManuVis. Hierna werd ik gevraagd om een hoofdstuk Visus en Motoriek in het leerboek Kinderfysiotherapie uitgegeven door Elsevier te gaan schrijven, samen met Marieke heeft dit vorm gekregen. Bouwien bedankt voor de begeleiding in deze fase. Marieke hierbij wil ik je bedanken dat we zovele jaren al contact hebben, een luisterend oor hebben voor elkaar en tevens kritisch naar elkaar zijn en ik hoop dat dit in de toekomst zo zal blijven.

Om in loondienst te gaan vanuit de eerstelijns zorg was het nodig om zelf iemand in loondienst te nemen om de praktijk te continueren. Dit was Annemieke Gerrits, met wie ik erna een lange periode in een maatschap gezeten heb. Ook zij heeft een bijdrage geleverd aan de onderzoeken van driejarige kinderen. Annemieke bedankt voor de samenwerking gedurende deze jaren.

Steeds bleef ik geïntegreerd om zo evidenced-based mogelijk aan het werk te zijn. Het behalen van Master of Research (NICI) onder leiding van Professor Dr. Bouwien Smits–Engelsman en Professor Dr. Gerard Van Galen in Nijmegen was de volgende stap. Het was een mooie tijd samen met Anneloes Overvelde, Hilda Bloem en Addy van der Lint. Het experiment wat toen als studieopdracht werd uitgevoerd is later gepubliceerd, mede dankzij medeauteur Dr. Ralf Cox. Het afronden en insturen van dit artikel vond plaats in een vakantie tijdens mijn wandeltocht van Amsterdam naar Nice. Met enige regelmaat raadpleegde ik de mail in Frankrijk door met de auto soms wel 40 km verderop te rijden en bij een 'office de tourisme' of bibliotheek er de tijd voor te nemen.

Door de medewerking aan het loep- en mijlpalen project op Bartiméus kreeg ik de kans veel kinderen te testen op de fijne motoriek. Het is altijd fijn wanneer mensen in je geloven en je willen laten presteren. Zo heb ik mede dankzij Nienke Boonstra, oogarts, het onderzoek naar de fijne motoriek van slechtziende kinderen verder uit kunnen breiden en vorm kunnen geven. Tijdens het mijlpalenproject leerde ik Bianca Huurneman en Joyce Schurink kennen. Onder de stevige leiding van Loekie de Vaere werd er een groot aantal kinderen op een dag gezien voor verschillende onderzoeken. Dit verliep allemaal vlekkeloos. Hierna kon er een groot aantal kinderen toegevoegd worden aan het databestand van de kinderen vanaf 4 jaar. Hierna kwam het artikel tot stand van de fijne motoriek van de leeftijdsgroep 4–11 jaar in samenwerking met de (co)promotoren.

Intussen werd ik gestimuleerd door Nienke Boonstra om een promotietraject in te gaan. Zo kreeg het contact met Ria Nijhuis van der Sanden verder vorm. De inhoudelijke discussies tussen ons gaven steeds nieuwe inzichten. Mijn promotor Ria

Nijhuis-van der Sanden wil ik bedanken voor haar onuitputtelijke enthousiaste inzet. Mijn copromotor Nienke Boonstra wil ik bedanken dat zij als oogarts veel belangstelling had voor het fijnmotorisch functioneren bij slechtziende kinderen, waardoor er mede door haar inzet projecten aangevraagd en gehonoreerd werden. Mijn copromotor Ralf Cox wil ik bedanken voor de steun en vele telefonische prettige contacten op de vrijdagochtend in de afgelopen jaren.

Voor de nieuwe versie van de manual, ManuVis-2, zijn Ria Nijhuis-van der Sanden en Ralf Cox mede auteur. Mede dank zij jullie is deze versie tot stand gekomen. Verder wil ik Marieke Siemonsma en Samantha Kerkhof bedanken voor de hulp om deze versie vorm te geven qua tekst en fotomateriaal. Ook wil ik hierbij Chris Mulder van Pinta bedanken voor de layout.

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# Over de auteur

Annemieke Reimer is geboren en getogen in Rotterdam.
Na afronding van een 6 jarige HBS opleiding (diploma 1969) is zij aan het werk gegaan als computerprogrammeur bij verschillende werkgevers (1969-1972 accountantskantoor Rotterdam: Moret & de Jong & Starke, 1972-1973 scheepvaartkantoor IHC, Schiedam, 1973-1975 Zwolse Algemene, Utrecht).
In 1975 koos zij ervoor om de opleiding fysiotherapie te gaan volgen. Van 1979 tot 2015 werkte ze bij verschillende (kinder)fysiotherapie praktijken in Zeist (Fysiotherapie centrum Steniaweg, Fysiotherapiecentrum Sanatoriumlaan, Kinderfysiotherapie Zeist). Zij volgde diverse bij- en nascholingscursussen voor het werken met kinderen in de leeftijd van 0-18 jaar.
Vanaf 1995 was zij ingeschreven in het deelregister bevoegde Kinderfysiotherapeuten en vanaf 1998 in het Centrale Kwaliteitsregister bij het Koninklijk Nederlands Genootschap voor Fysiotherapie.
In 1998 werd zij gevraagd om als consulent kinderfysiotherapeut bij Bartiméus te gaan werken. Van 1998 tot 2015 combineerde zij de praktijkwerkzaamheden met deze deeltijdbaan.
Vanaf 2000 combineerde ze de werkzaamheden tevens met een studie aan het NICI (Nijmegen Institute for Cognition and Information) en in 2003 behaalde zij de titel Master of Research in Cognitive Neuromotor Science.
Haar belangstelling voor de invloed van de visus op de motoriek had verder tot gevolg dat er een promotietraject tot stand kwam. Vanaf 2008 werd zij promovendus bij het Radboud Institute for Health Sciences, IQ healthcare/Department of Rehabilitation in Nijmegen, met als onderwerp: Fine motor skills of visual impaired children.
Vanaf 2013 verzorgt zij lessen aan de Opleiding Master Fysiotherapie afstudeerrichting Kinderfysiotherapie van de Hogeschool Utrecht.
Vanaf 1977 woont Annemieke in Zeist, samen met Hans Wisman.

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