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ORIGINAL ARTICLE

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

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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 to 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.

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## Background and Relevance of This Study

Brambring (2006) defined primary and secondary functions of vision in the acquisition of motor skills (Brambring 2006). 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 (Brambring 2006). 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 (Warren 1994). 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 (Brambring 2006). 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 (Smits-Engelsman et al. 2003), 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 (Boonstra et al. 2012). 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 (Gibson 1979; Reed 1982). It is well known that motor and perceptual development are strongly intertwined, working in unity rather than being separate processes (Berger and Adolph 2007; von Hofsten and Ronnqvist 1988). 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 (Adelson and Fraiberg 1977; Houwen et al. 2009; Troster and Brambring 1992). 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 (Haber et al. 1993; Papadopoulos and Koustriava 2011), and during the movements to control the actions in such a way that goals are reached (Brambring 2006). 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 (Brambring 2006). 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 (Bouchard and Tetreault 2000; Celeste 2002; Haibach et al. 2014; Houwen et al. 2007, 2008, 2009; Levtzion-Korach et al. 2000; Liebrand-Schurink et al. 2015; Navarro et al. 2004; Reimer et al. 1994; Troster and Brambring 1992). 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 (Houwen et al. 2009). In a recent study Haibach et al. (2014) used the Test of Gross Motor Development (TMGD-2; Ulrich 2000) 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 (Houwen et al. 2009). This is not what might be expected: during typical child development, motor coordination gradually improves with increasing age (Bayley 1969; 1993; Henderson and Sugden 1992; Ulrich 2000; Wiegersma

1988) 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 (Brambring 2001; Gringhuis et al. 2002; Sleeuwenhoek et al. 1995). 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 (Tadic et al. 2014).

## 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 (Bishop 1991; revised 1996). 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 (Aki and Atasavun 2007). 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 (Reimer et al. 2008) such as handwriting (Aki et al. 2008). 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 (Cox et al. 2009; Lewis et al. 2000). 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 (Webber et al. 2008). 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 (Caputo et al. 2007). Houwen and colleagues (2008) 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 (Houwen et al. 2008). 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 (Houwen et al. 2009) 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 (Terwee et al. 2012). 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 (Houwen et al. 2014) 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 (APA 1999; Bayley 1969; 1993) has been adapted for children with low motor performance and low vision (Visser et al. 2013, 2014). Comparable research had been done with the Dutch second edition of the Bayley –II (Ruiter et al. 2011). 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 (Visser et al. 2014), and a larger study found that the adaptations resulted in a higher score on the cognition scale, but not on the motor scale (Visser et al. 2013). 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 and 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 (Houwen et al. 2010). 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 (Reimer et al. 1999, 2008; Schurink et al. 2012; Smits-Engelsman et al. 2003). 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 Fig. 1) is based on items adapted from existing valid test assessments: the Movement Assessment Battery for Children version I (MABC I; Henderson and Sugden 1992), and the General Movement Coordination Test (GMCT; Wiegersma 1988). The inter-rater reliability of the Movement ABC (Dutch version ranged from 0.95 to 1.00 (Smits-Engelsman et al. 2008). The test-retest reliability of the manual dexterity items of the MABC was measured in young children (4–5 years) and varied from 0.88 to 0.98 (Van Waelvelde et al. 2007; Croce et al. 2001). 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 items. (GMCT; Wiegersma 1988).

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 and 11 years, and to investigate the test–retest reliability and inter-rater reliability of the ManuVis.

## 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.

**Table 1** Description of all six items and corresponding procedures of the ManuVis (Smits-Engelsman et al. 2003) compared to the original Movement ABC (MABC) (Henderson and Sugden 1992) and General Movement Coordination Test (GMCT) (Wiegersma 1988)

*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.*

**Table 1** (continued)

*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.

## 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 (WHO 1997). The inclusion criteria for children with VI in this study were adopted from the rehabilitation protocol (Boonstra et al. 2012): visual

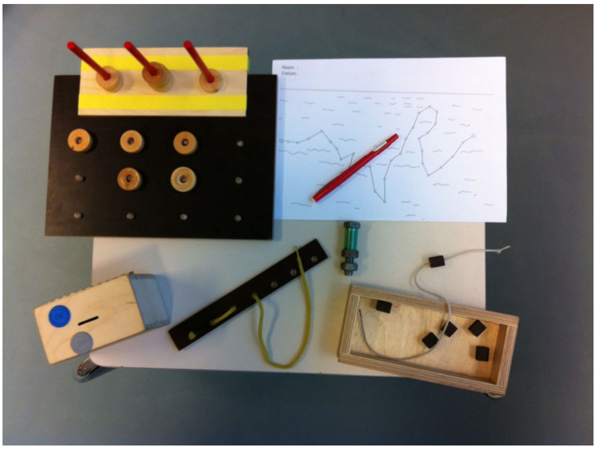

**Fig. 1** Photo of all materials used in the ManuVis for all six items: (1) money box, (2) wooden board with twelve rings, (3) nuts with bolt, (4) beads with cord, (5) board with lace, (6) paper for the writing task and (7) open container

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. The eye disorders underlying the visual impairment in the participants are described in detail in Table 2.

The NS children in the control group attended regular schools in the neighbourhood of the Dutch city of Utrecht and were matched on age. 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 (Smits-Engelsman et al. 2003) 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.

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) aged 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 (YS / RC) during the test, and the other (AR) was scored later from video recordings.

**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 | Congenital stationary night blindness (CSNB) | 13 |
| 6 | Aniridia | 11 |
| 7 | Cone- rod dystrophy | 9 |
| 8 | Optic nerve defects / optic nerve atrophy | 7 |
| 9 | Hyperopia (>4D) | 7 |
| 10 | Congenital glaucoma | 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 |

**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 (Smits-Engelsman et al. 2003)

| 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 |

## 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, (Henderson and Sugden 1992) and in 2007 a second version was presented (MABC II; Henderson et al. 2007). 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 test items of the ManuVis with the same instructions and procedures in all age groups in contrast to the MABC. In this latter test the items change over age groups resulting in an increasing task 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 (Wiegersma 1988). 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 Fig. 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 min, 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 (Portney and Watkins 2000). 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 [$SEM = SD \times \sqrt{1 - ICC}$] (Nunnally and Bernstein 1994). As a criterion for acceptable precision of the SEM, a value SD/2 was used (Wyrwich et al. 1999). 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 ($n=14$): $M=52.5$ s, $SD=23$ s; new group ($n=13$): $M=43.7$ s, $SD=7.4$ s. 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 ($n=20$): $M=38.6$ s, $SD=7.5$ s; new group ($n=9$): $M=36$ s, $SD=3.5$ s. 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.

### 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.

### 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 Fig. 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*.

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 Fig. 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 and 6 years and 5 and 6 years. Differences between the VI group and NS group are largest in the younger age groups.

**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 | 39 | 42 | 32 | 37 | 40 | 29 | 36 | 39 | 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

There were also significant Group by 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 0.007. It can be concluded that ManuVis reliability is sufficient.

## 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 and 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 Fig. 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

**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 |

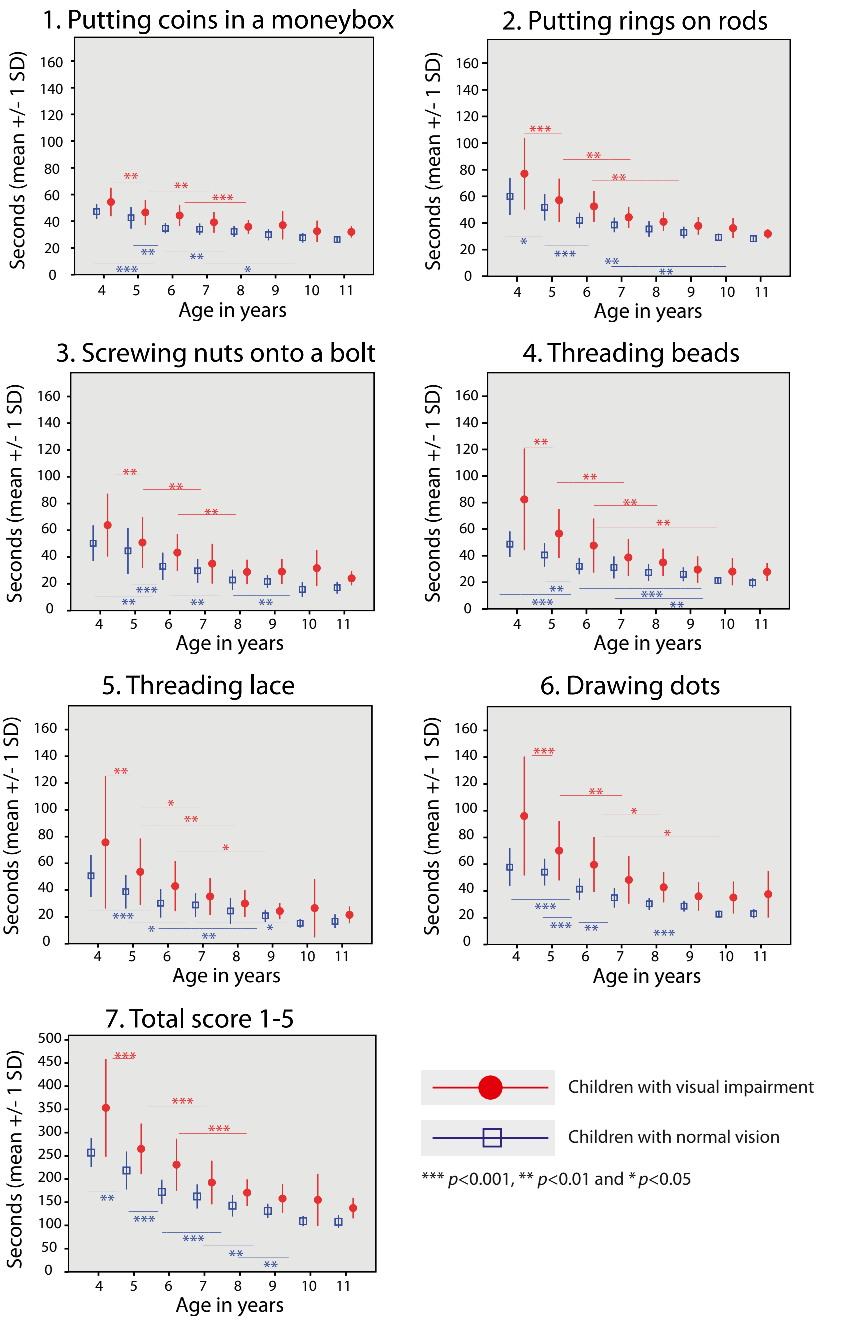


**Fig. 2** Mean (M) and standard deviation (SD) in seconds of the scores per group for each item and for total score 1–5

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

**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

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 (APA 1999; Bayley 2006) adapted for children with low motor performance and low vision (Visser et al. 2013, 2014) or the previous version of the Dutch second edition of the Bayley – II (Ruiter et al. 2010, 2011). 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 (Houwen et al. 2014) 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 (Houwen et al. 2010). The influence of age was confirmed in a factor analysis. In a recent study (Haibach et al. 2014) of the TMGD-2 (Ulrich 2000), 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 (Bayley 1969; 1993; Henderson et al. 2007; Henderson and Sugden 1992; Ulrich 2000; Wiegersma 1988), 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 (Houwen et al. 2009). 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 (Gibson 1979; Reed 1982), and motor and perceptual development are strongly intertwined (Berger and Adolph 2007; von Hofsten and Rosblad 1988), 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 (Rogow 1992). Only weak evidence is found for a relationship between the degree of VI and gross motor development and manual dexterity (Houwen et al. 2009) 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 (Liebrand-Schurink et al. 2015; Reimer et al. 2008) especially seen in the results of more complex tasks such as handwriting (Aki et al. 2008). 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 (Haber et al. 1993; Papadopoulos and Koustriava 2011) 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 Fig. 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 Fig. 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 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 (Reimer et al. 1999). 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. 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 do 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- 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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