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Hand functioning in children with cerebral palsy

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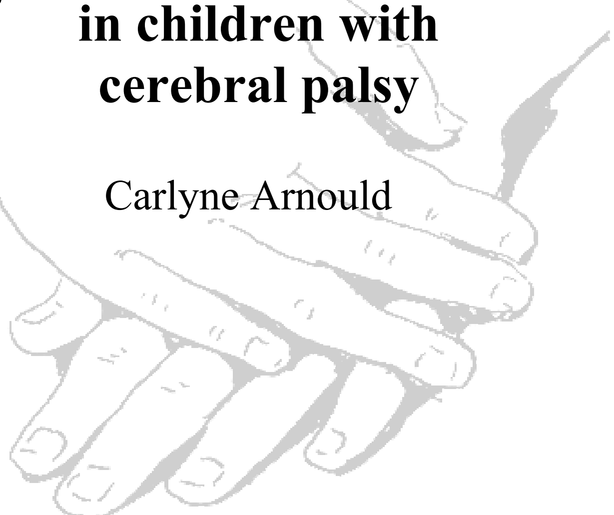

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Contents

Introduction

The hand is a marvelous organ which enables humans to carry out a enormous variety of actions in daily life. It is incredibly versatile and continually switches between executive, exploratory and expressive activities (Connolly, 1998). The hand can be used on its own as a platform, a hook, a tweezers, or a vice, or in combination with tools to grasp, transport and manipulate objects. It is capable of the strongest grasp such as in clasping a hammer or of the most delicate touch such as in holding a needle. The hand is also used to explore, perceive and recognize surfaces, objects and their properties and to express emotions, or communicate with others. Accordingly the human hand is a very complex organ with highly skilled functions that are very difficult to recreate with artificial electromechanical structures. All attempts in the construction of an artificial hand comparable in functioning to the human hand have been disappointing up till now (Connolly, 1998).

The phylogenesis of the hand functioning

The hand functioning has reached its highest level in humans. The anatomical structures and the biomechanical properties may explain, at least partially, the superiority of the human hand (Flanagan & Johansson, 2002). For instance, the human thumb which is much longer, relative to the index finger, than the chimpanzee thumb allows smaller objects to be grasped with a greater precision. Similarly, the more individuated muscles and tendons may explain the greater independence of finger movements observed in humans as compared to monkeys. However, the highly skilled functions of the human hand cannot be simply explained by differences in anatomical structures. Primates whose hands have similar musculoskeletal structure and biomechanical properties may present very different motor skills (Phillips, 1971). For instance, the rhesus monkey can pick up small objects between the index finger and the thumb whereas the squirrel monkey, whose hand and fingers shape is almost identical, can execute this task only by closing the whole hand around them. The reason for this difference is that the squirrel monkey does not have the central nervous system control mechanisms which make this refined task possible, whereas the rhesus monkey does (Lawrence & Hopkins, 1976). Rather than changes in biomechanical conditions, it is the evolution in cortical control mechanisms that mainly contributes to the different levels of hand functioning observed between the species. Compared to the lower mammals, the primates have evolved toward an increasingly dominant role played by the motor cortex and the corticospinal tract (i.e., a massive bundle of fibers which transmits the motor commands required to control the voluntary movements from the cortex to the spinal cord) in controlling hand movements. Direct monosynaptic connections between neurons in the motor cortex and spinal motoneurons have also emerged (Phillips, 1971; Heffner & Masterton, 1983; Porter & Lemon, 1993; Flanagan & Johansson, 2002). These major changes in the cortical system controlling highly skilled movements have reached their greatest development in the human being (Phillips, 1971; Porter & Lemon, 1993; Wiesendanger, 1999). Several studies (Lawrence & Kuypers, 1968a and 1968b; Porter & Lemon, 1993; Lang & Schieber, 2003 and 2004) have demonstrated the critical role of the primary motor cortex and the corticospinal tract (especially the direct corticomotoneuronal connections) in the capacity to move the hand and the fingers with skill which cannot be substituted by any other structures (Pehoski, 1995). However, skilled movements also require sensory feedback. It is therefore not surprising that the dorsal column nervous pathway, which conducts tactile and proprioceptive information from the periphery to the thalamus and the somatosensory cortex, has evolved in parallel with the motor corticospinal pathway and has reached its highest level of development in humans (Mountcastle, 1984). In addition, the cortical areas involved in the integration of motor and sensory information (e.g., posterior parietal lobe, premotor and supplementary motor areas) have also expanded to cope with more complex acts (Pehoski, 1995).

Cerebral Palsy: a disruption of the hand functioning

Hand functioning requires the integrity of the central nervous system and therefore may be disturbed by several brain disorders. Cerebral palsy (CP) which results from early brain lesions constitutes the most prevalent form of physical disability in childhood (Rosenbaum, 2003), occurring in 2 to 2.5 per 1,000 live births (Stanley et al., 2000). Cerebral palsy is a broad term that refers to “all non-progressive but often changing motor impairment syndromes secondary to lesions or anomalies of the brain arising in the early stages of its development” (Mutch, 1992). The severity and the type of motor impairment syndromes widely vary according to the time of appearance, the location and the degree of cerebral damage. Major lesions are commonly observed in the motor cortex and the corticospinal pathway (Uvebrant, 1988; Yokochi et al., 1992). As a result, skilled hand movements do not develop normally and several daily activities requiring the use of the hand(s) are difficult or even impossible to carry out.

The International Classification of Functioning, Disability, and Health

The impact of CP on child’s hand functioning may be formalized using the International Classification of Functioning, Disability, and Health (ICF) (World Health Organization, 2001). The ICF has been developed within the framework of the World Health Organization's (WHO) conception of health as a state of physical and psychosocial well-being. Three separate but related dimensions of functioning are defined: body functions and structures (body dimension), activity (individual dimension), and participation (social dimension). Body functions are the physiological or psychological functions of the different body systems, and body structures refers to the anatomic parts of the body such as organs, limbs and their components; activity is the child's ability to execute a task or an action generally considered as essential for his/her everyday life; and participation is the child's involvement in life situations such as his/her attendance at school. Problems in each dimension of functioning are respectively designated as impairments, activity limitations, and participation restrictions; and are encompassed under the umbrella term of “disability”. Impairments refer to anomaly, defect, loss or other significant deviation in body functions and structures; activity limitations occur when the child experiences some difficulties in executing daily activities; and participation restrictions are the problems the child may experience in the involvement in life situations.

The impact of CP on a child's hand functioning is illustrated in Figure 1 according to the three dimensions of functioning and disability. Cerebral palsy implies "lesions or anomalies of the brain arising in the early stages of its development" (Mutch, 1992). These brain lesions (e.g., corticospinal pathway) may affect other body structures such as hand and its components (e.g., muscles, joints, bones) as well as several body functions. Hand functions are frequently impaired and constitute the main problem in many children with CP (Uvebrant, 1988). Depending on the location and the degree of cerebral damage, both motor and sensitive hand functions may be impaired. Hand motor impairments which may be encountered in CP include, for instance: a diminution in mobility (e.g., passive and active range of motion), a reduction in muscle strength (e.g., grip strength), a lack of control in rapid coordinated movements (e.g., gross manual and fine finger dexterity), the presence of involuntary movements (e.g., synkineses), and an exaggeration in muscular tone (e.g., spasticity). Hand sensory impairments may be observed for example in tactile pressure detection, tactile spatial resolution (i.e., perception of spatial features of objects and surfaces), thermal sensation, stereognosis (i.e., recognition of common objects and shapes), and proprioception (i.e., position and movement sense of hand or fingers).

Cerebral palsy may also limit the achievement of daily activities requiring the use of the hands such as eating, drinking, grooming (e.g., toothbrushing, hairbrushing, washing), dressing (e.g., fastening, buttoning, putting on/taking off clothes), and schoolwork (e.g., writing, drawing, do-it-yourself). Manual ability refers to the child's capacity to use the hands and upper limbs in managing such manual activities of daily life (Penta et al., 2001). Manual ability is different from hand functions (e.g., grip strength, dexterity, tactile pressure detection) in the sense that the child's actions are performed for the purpose of doing a specific activity (McDougall & Miller, 2003). On the opposite, hand functions refers to skills that have no specific purpose. For instance, the grip strength measured on a dynamometer is a hand function which simply reflects the child's grasping skill while the capacity to execute daily activities such as grasping a jar of jam to open it or grasping a fork to cut meat reflects the child's manual ability. More details about the distinction between dexterity and manual ability are given in the conclusion and perspectives section.

Finally, CP may restrict the participation of the child in school life, familial life, and leisure activities. Participation restrictions represent the problems the child experiences in the fulfilment of social roles that are regarded as normal considering his/her age, sex, and the society and culture in which he/she lives. Social roles of a CP child consist, for instance, of being a pupil, a brother/sister, a friend, or a play companion. Participation restrictions may include difficulties to take lessons, inability to take care of his/her little siblings, difficulties to join in housework, and problems to communicate and have good relationships with peers.

Drawing distinctions between hand impairments (body dimension), manual ability (individual dimension), and participation (social dimension), as proposed by the ICF, is far from being futile as a problem may occur in one dimension but not in the others. For instance, a CP child presenting a grip strength significantly lower than normal (functional impairments) may carry out manual activities without any difficulty (manual ability). Conversely, a CP child without any hand impairments may be limited in the achievement of manual activities due to intellectual and/or visual disorders. A tetraplegic child with severe hand disability (hand impairments and manual ability limitations) may take an active part in his/her school life (participation) as the special school he/she attends provides facilities and teaching/supervisory staffs that favour the child's participation; conversely, an hemiplegic child with slight hand disability may be severely restricted in the participation in his/her mainstream school life. As illustrated here, the participation dimension refers to the impact of CP on a child's social functioning, and thus oversteps the frame of this work about the hand functioning in children with CP. Hence this dimension will not be considered further in this work.

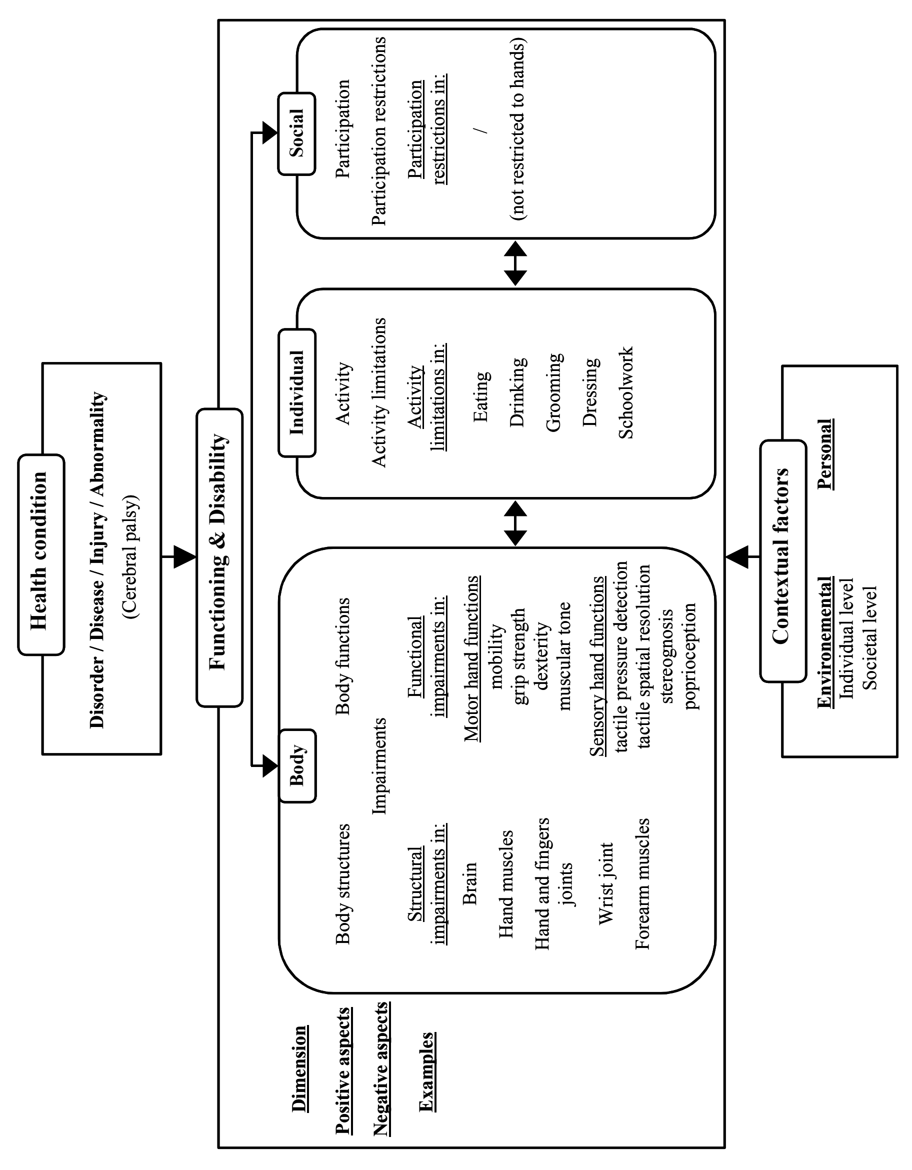

Figure 1. Impact of cerebral palsy on a child's hand functioning according to the ICF dimensions (modified from WHO 2001).

Figure 1 shows that there is a bidirectional interaction between the three dimensions of functioning. Changes in one dimension have the potential to influence and modify the other dimensions but not always in a predictable unequivocal relationship. For instance, hand impairments may be responsible for the difficulties a child has in executing manual activities. However, the child may compensate his or her hand impairments by learning adapted strategies (McCuaig & Frank, 1991; Wade, 1997; Penta et al., 2001) such as breaking down a bimanual activity into several unimanual sequences. The three dimensions of functioning may also be influenced by contextual factors that represent the background of the child's life. Contextual factors include both environmental and personal factors. Environmental factors are external features of the physical, social, and attitudinal environment in which the child lives. They are organised on two different levels: the individual level concerns the immediate environment of the child such as the home or school settings (e.g., physical environment, family, peers, school education type) while the social level includes all formal and informal social structures, services, and systems in the society that have an impact on the child (e.g., health services, social security, transportation services, cultural attitudes). Personal factors are the internal features of the child which are not part of a health condition. They may include age, gender, lifestyle, habits, motivation, personality, adaptability, educational and social background. Both environmental and personal factors can facilitate or hinder the child's functioning at the body, individual, or social level. For instance, low incomes of the child's parents may prevent the child from receiving treatments which are expensive but effective in reducing hand impairments; the use of assistive devices may reduce activity limitations; and the fear of peers' opinion may restrict the child's participation.

To summarize, the hand functioning of a child with CP may be conceptualized as the result of a dynamic interrelationship between hand impairments and manual ability which both may be influenced by environmental and personal contextual factors. Although the strength of the relationship between hand impairments and manual ability is not documented in the ICF and remains a question open to empirical testing, hand impairments are generally thought to be largely responsible for the difficulty experienced in manual activities (Fedrizzi et al., 2003). Based on this belief, most of the therapeutic interventions implemented in CP children aim to reduce the hand impairments (Koman et al., 2004). One objective of the present work is therefore to empirically test the relationship between hand impairments and manual ability in children with CP.

The measurement of the hand functioning

The measurement of both hand impairments and manual ability in children with CP is crucial to determine the complete impact of CP on a child's hand functioning. This should give the health professionals the information required to identify the therapeutic needs of the children, to plan and implement interventions, to assess the effectiveness of the interventions, and, if necessary, to adjust the interventions. Consequently, it is essential to have high-quality measurements.

A wide variety of instruments are available to measure hand impairments (Thonnard et al., 1994; Barbier et al., 2003). Most of them express the hand impairments in physical units. For instance, joint mobility can be measured in degrees with a goniometer, grip strength can be measured in Newtons with a dynamometer, and tactile pressure detection can be measured in g/mm^2 with calibrated monofilaments. The physical measures obtained with such instruments satisfy the requirements of an objective measurement as defined by the Institute for Objective Measurement (2000): "objective measurement is the repetition of a unit amount that maintains its size, within an allowable range of error, no matter which instrument, intended to measure the variable of interest and no matter who or what relevant person or thing is measured". For instance, the Newton unit can be used to measure either the grip strength of a child or the force exerted by a cyclist on the pedals of a bike, and allows them to be compared quantitatively. So, instruments using physical units to measure hand impairments allow quantitative comparisons to be made across people and over time provided that the testing procedure supplies reproducible measures. Despite the existence of such instruments, hand functions and the extent to which they are impaired are not frequently investigated in children with CP. Moreover, the few studies looking into hand impairments are generally performed on small samples (Jones, 1976; Eliasson et al., 1991 and 1995; Cooper et al., 1995; Gordon & Duff, 1999a and 1999b; Krumlinde-Sundholm & Eliasson, 2002; Duqué et al., 2003), are mostly restricted to children with hemiplegia (Tizard et al., 1954; van Heest et al., 1993; Cooper et al., 1995; Gordon & Duff, 1999a and 1999b; Krumlinde-Sundholm & Eliasson, 2002; Duqué et al., 2003), are focused on just one aspect of hand functions (Wilson & Wilson, 1967a; Lesny, 1971; Jones, 1976; Lesny et al., 1993; Eliasson et al., 1991 and 1995; Gordon & Duff, 1999b), or do not compare the measures obtained in CP children with normative data accounting for age, sex, and handedness (van Heest et al., 1993; Yekutiel et al., 1994). Additional information about hand impairments in various types of CP children is therefore required.

Contrary to hand impairments, there is a lack of appropriate instruments measuring manual ability in children with CP. Indeed, most instruments developed for CP children are focused on the child's capacity to use the lower limbs (Russell et al., 1989; Boyce et al., 1992). Other pediatric instruments were developed to measure the child's ability to execute manual activities but they mainly involve unimanual activities (Johnson et al., 1994), are time-consuming (Haley et al., 1992), or are not validated for children with CP (Haley et al., 1992; Young et al., 2000). It was therefore deemed useful to develop a new measure of manual ability in children with CP.

Manual ability as a construct is observed indirectly through particular manifestations of the ability in carrying out specific tasks with the hands. Manual ability is therefore considered as a latent variable concealed within the child in the same sense as pain, intelligence, or anxiety (Thurstone, 1959; Rasch, 1960; Penta et al., 2001). Its measurement is possible through the performances on selected tasks and is based on the general idea that any measurement implies the conceptualization of an underlying continuum representing the variable that is being measured (Thurstone, 1959; Rasch, 1960). For instance, when measuring the length of an object, we refer to a continuum which is only partially represented by the ruler actually used. The obtained measure is generally represented as a point along the ruler and its location indicates the “amount” of length the object possesses. Manual ability can also be conceptualized as a continuum representing an infinity of levels from “less able” to “more able”. Measuring the manual ability of a CP child is equivalent to determining the child's location along the underlying scale of manual ability. This measurement scale can be materialized by situations or activities, called “items”, which require some amount of manual ability to be performed. Although there may be an indefinitely large number of manual activities, only a sample of activities is in practice included in a test or questionnaire.

The items selected in a questionnaire are in fact the operational definition of the variable (i.e., the graduations of the measurement scale). Therefore, it is important that they cover the whole range of manual ability that we want to explore. As illustrated in Figure 2, the more difficult the item, the higher the manual ability level it requires to be performed successfully. The less difficult items can be successfully achieved by the less able children while the more difficult items can only be successfully achieved by the more able children. For instance, child A presents a very low manual ability since he/she is expected to successfully achieve only the four easiest items; child B has an intermediate manual ability that should enable him/her to succeed approximately half of the items; and child C has a high manual ability and is supposed to succeed all but the most difficult item.

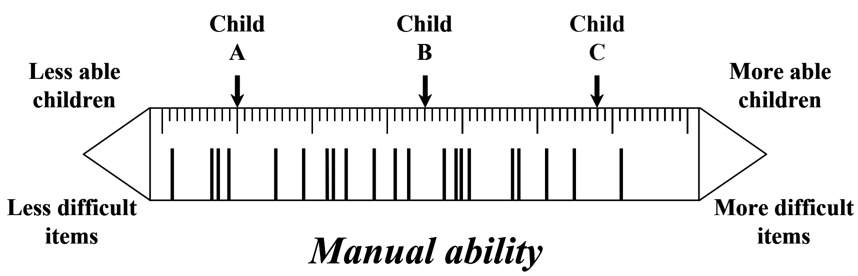

Figure 2. Representation of the manual ability continuum. Thick lines indicate, from left to right, the location of manual activities (or items) of increasing difficulty. Arrows indicate the location of children A, B, and C on the manual ability continuum.

Thus, the measurement of a latent variable such as manual ability starts with the counting of the number of items succeeded (as when we count the number of metric graduations which separate two extremities of an object in order to measure its length). This measurement process can be applied regardless of the response format of the questionnaire used to record the responses. Briefly, the dichotomous response format, success or failure, is the simplest response format as it includes only two response categories (e.g., the child is able or unable to perform each manual activity) while the polytomous response format allows a more subtle response as it includes more than two ordered categories (e.g., each manual activity is scored as either "impossible", "difficult", or "easy").

The operational definition of the manual ability continuum occurs differently depending on the response format used. In the dichotomous response format (see Figure 3), the "graduations" of the underlying manual ability scale are the item difficulty locations. The item difficulty locations are defined as the level of ability that gives an equal chance of success and failure, and are the thresholds between the failure and success categories. In the polytomous response format (see Figure 4), the "graduations" of the measurement scale are the item threshold locations defined between successive response categories. The thresholds of each item correspond to the manual ability levels required to have an equal probability to endorse a response rather than the previous one. Note that the item difficulty merely represents the average value of its encompassed thresholds.

Whatever the response format used, a score is assigned to each response category in such a way that higher scores represent a higher manual ability (e.g., in the dichotomous response format: 0 = failure, 1 = success; in the polytomous response format: 0 = impossible, 1 = difficult, 2 = easy) and a total score is subsequently computed by summing the child's scores to each item. If the successive response categories really represent increasing levels of manual ability as postulated a priori, the "amount" of manual ability a child possesses can be inferred from his or her total score: the higher the total score, the higher the manual ability. However, the total score is not sufficient to make quantitative comparisons as a unit progression in total score does not necessarily entail the same progression in manual ability throughout the continuum. Such comparisons require a measurement unit that is constant and reproducible throughout the range of the variable measured (Merbitz et al., 1989; Wright & Linacre, 1989; Wright, 1997). Measurement models are therefore required to translate the ordinal total score observed in a questionnaire into a linear manual ability measure.

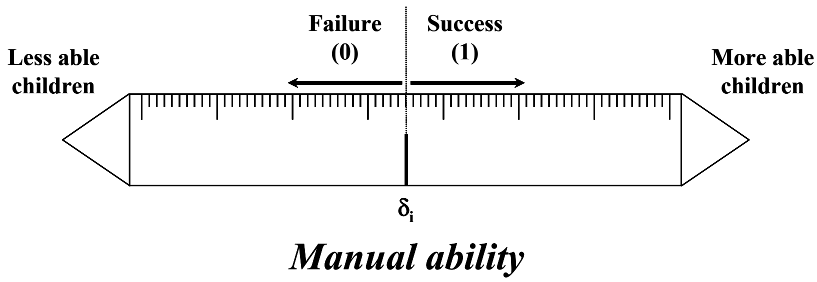

Figure 3. Dichotomous response format. The manual ability continuum is materialized by the item difficulty (thick line; δ_i). Children with a manual ability level lower than the item difficulty (i.e., located to the left of the item) are expected to fail the item; children with a manual ability level higher than the item difficulty (i.e., located to the right of the item) are expected to succeed the item.

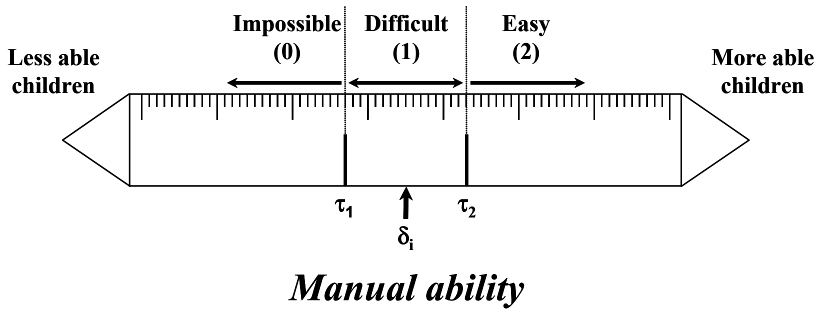

Figure 4. Polytomous response format. The manual ability continuum is materialized by the item thresholds (thick lines). The first threshold (τ_1) is located at the manual ability level required to respond "difficult" rather than "impossible" while the second threshold (τ_2) is located at the manual ability level required to respond "easy" rather than "difficult". The item difficulty (arrow; δ_i) represents the average value of its encompassed thresholds. Children with a manual ability lower than the first threshold are expected to fail the item; children with a manual ability located between the two thresholds are expected to perform the item with difficulty; and children with a manual ability higher than the second threshold are expected to perform the item easily.

The Rasch model

Several measurement models called "Item Response Theory" (IRT) models (Hambleton et al., 1991; Bertrand & Blais, 2004) have been developed to transform, through a probabilistic framework, the responses observed in a set of items into manual ability measures. They are based on the assumption that subjects with a higher manual ability should have a higher probability, relative to subjects with a lower ability, to successfully achieve any item. Almost all IRT models are descriptive since they aim to describe the observed responses the most exactly possible. Such models are generally used to find the model which best explains the observed data but they do not satisfy the requirements of an objective measurement (Hobart, 2002; Andrich, 2004). On the contrary, the Rasch model (Rasch, 1960) is a prescriptive rather than a descriptive model in the sense that it formulates the requirements of an objective measurement and makes it possible to verify if these requirements are satisfied in the observed data (Rasch, 1960; Andrich, 1989; Hobart, 2002; Andrich, 2004; Penta et al., 2005).

The Rasch model (Rasch, 1960) is a probabilistic model developed by the Danish mathematician Georg Rasch in the 1960s to construct an intelligence scale. It was progressively and successfully applied to psychology, sociology, educational sciences, medicine, and many other disciplines in the human sciences. The Rasch model prescribes that the probability to succeed a given item solely depends on the subject ability and the item difficulty. This model was originally developed for dichotomous response formats. Other models of the same family, i.e. Rasch models, have subsequently been derived to comply with polytomous response formats (Andrich 1978a, 1978b, and 1979; Masters, 1982). These models prescribe that the probability of endorsing any response category to an item solely depends on the subject ability, the item difficulty, and the threshold difficulties. In the case of manual ability measurement, no other attribute of the subjects or the items than manual ability is theorized to account for the probability of endorsing a response. This requirement called "unidimensionality" is essential to achieve an objective measurement as found in physical sciences (Andrich, 1988; Wright & Linacre, 1989; Bond & Fox, 2001). For instance, a ruler measures only the length of the objects and should not be biased by other attributes such as its temperature, weight, volume, texture, or color. Similarly, the items of a questionnaire intended to measure manual ability should provide a measure relating to only the manual ability of the subject, unbiased by other attributes of the subject or of the items.

> ***Example:*** *Consider a questionnaire made of 10 dichotomous items related to manual ability and 10 dichotomous items related to locomotive ability. A score of 0 is attributed when the item is failed and a score of 1 when the item is succeeded. If we consider all of the 20 items, what would mean a total score of 10? It could mean that the subject presents a moderate manual and locomotive ability. But it could also mean that the subject has a high manual ability but a poor locomotive ability, or vice versa.*

This example shows that, when a total score is related to more than one attribute of the subject it is impossible to infer relevant information about the ability of the subject. Actually, unidimensionality is a theoretical concept which is never entirely met in practice as the complete isolation of one attribute from the others is extremely difficult (Andrich, 1988). However, getting as close as possible to this ideal is important to obtain a measurement scale which allows the same attribute (e.g. manual ability) to be quantitatively compared across different subjects (Wright & Linacre, 1989). The metric requirement of unidimensionality may be considered to be satisfied when the responses to the items are influenced in a dominant way by the latent variable purported to be measured (Hambleton et al., 1991). Rasch analysis can be used to verify the extent to which the responses of each subject to each item fit the requirement of unidimensionality (Andrich, 1988). The observed responses are contrasted with the responses expected by the model which formulates this requirement. The degree of similarity between observed and expected responses is reported through different fit statistics and indicates the extent to which unidimensionality is satisfied in the observed data (Rasch, 1960; Wright & Panchapakesan, 1969; Wright & Stone, 1979; Wright & Masters, 1982; Andrich, 1988; Smith et al., 1998; Smith, 2000). Fit statistic analyses are particularly useful during the construction of a questionnaire to select items that line up on a unidimensional continuum. Provided that the data fit the requirement of unidimensionality, the scores observed to any particular item should not be influenced by other factors of the subjects than just the variable of interest. Hence, subjects of the same ability should obtain the same score to any item, regardless of their other characteristics (e.g., age, gender, handedness, gross motor and hand functions). If this is not the case, the item is biased or, according to a more recent terminology, the item presents a differential functioning (Holland & Wainer, 1993). The Rasch model allows the invariance of the item functioning to be tested across various subgroups of subjects using differential item functioning tests (Wright & Stone, 1979; Smith, 1992; Andrich et al., 2004).

The methods allowing to quantify how the observed data fit the requirement of unidimensionality can be applied to both dichotomous and polytomous response formats. A supplementary verification is however required in the case of polytomous items. When constructing a polytomous rating scale, a score is assigned to each response category so that higher scores represent a higher amount of the variable purported to be measured, for instance: (0) impossible, (1) difficult, and (2) easy. The order of the response categories is postulated a priori based on the common sense that a subject performing an activity easily should have a higher ability than a subject performing the same activity with difficulty, and so on. In practice, the polytomous rating scale is not always used by the respondents as intended by the test constructor (Roberts, 1994). It is therefore important to examine whether or not the order of the response categories postulated a priori is verified in the observed data (Andrich, 1996a). The Rasch model makes it possible to verify whether the successive response categories of each item represent increasing levels of ability and whether the thresholds between successive response categories are located on the continuum in the anticipated order (Andrich, 1996b).

Once the observed responses fit the model's prescriptions, the Rasch model estimates the ability of each subject and the difficulty of each item or threshold on a common linear interval scale (Rasch, 1960). Moreover, the location of each subject is estimated independently of the relative difficulty of the particular item set used to collect the responses and, similarly, the location of each item (or threshold) is estimated independently of the ability of the particular sample assessed (Rasch, 1960). These two properties known as the “linearity” and the “specific objectivity” are essential to obtain objective measures like those found in physical sciences (Wright & Stone, 1979; Bond & Fox, 2001; Hobart, 2002; Andrich, 2004).

Linearity implies that the unit of the measurement scale is constant throughout the scale so that identical intervals represent the same amount of the variable purported to be measured (Wright & Linacre, 1989). Linearity is not obtained when we work with ordinal total scores since they rely on counts of potentially unequal units (Merbitz et al., 1989; Wright & Linacre, 1989; Wright, 1997). Actually, the use of raw total scores has several limitations especially when quantitative comparisons are made across subjects or over time. Firstly, the ordinal scores assigned to the response categories of polytomous items are separated by unknown distances (Merbitz et al., 1989; Wright & Linacre, 1989). To illustrate, suppose a questionnaire measuring manual ability on the three-level scale: (0) impossible, (1) difficult, and (2) easy. It is impossible to know a priori if responding “difficult” rather than “impossible” to a given item represents the same progress in manual ability than responding “easy” rather than “difficult” although the score has increased by one point in both cases. Similarly, a progression from “impossible” (scored as 0) to “easy” (scored as 2) represents greater progress than from “impossible” (scored as 0) to “difficult” (scored as 1), but not necessarily a double progress. Consequently, the total scores allow subjects’ manual ability to be compared in terms of “higher than”, “lower than” or “equal to”, but they fail in determining “how much higher” or “how much lower”. Secondly, obtaining the same score to different items represents not necessarily the same amount of the latent variable (Bond & Fox, 2001; Penta et al., 2005). For instance, responding “easy” to the item “Buttoning up a shirt/sweater” requires a higher manual ability than responding “easy” to the item “Switching on a bedside lamp”. The total scores must therefore be converted into linear measures before quantitative comparisons can be made. The Rasch model uses a logistic transformation to convert the ordinal total scores into linear measures expressed in “logits” (i.e., log-odds units). The logit is a probabilistic unit defined as the natural logarithm of the odds of success (i.e., the pass/fail probability ratio) of a subject to an item. This unit is constant throughout the measurement scale. At any level of the measurement scale, a 1-logit difference in subjects' ability implies a constant ratio of their odds of success ($e^1 = 2.71$) to any given item; a 2-logit difference always represents the odds of success in a ratio of $e^2 = 7.39$, and so on. Consequently, the measures obtained by the Rasch model are linear; they can be used to compare quantitatively the ability of different subjects or to follow their ability over time.

Specific objectivity implies that the subjects measures are independent of the relative difficulty of the particular items used, and similarly, that the relative item difficulties are independent of the ability of the particular sample who answered them. This property is essential to obtain measures that maintain their quantitative status regardless of the particular context in which they occur (Wright & Linacre, 1989). Consider the well-known metric system, the length measure of an object is independent of which ruler is used and the calibration of a ruler is maintained irrespective of what it is measuring. Similarly, the subjects measured by a questionnaire must retain the same ability regardless of the relative difficulty of the particular items encountered and the items must maintain their difficulty regardless of the ability of the respondent. This requirement is not satisfied when using the total scores of a questionnaire. Indeed, the total score of a subject depends on the difficulty of the particular items used in the questionnaire (Rasch, 1960). It will be high if the questionnaire includes many easy items and will be low if the questionnaire includes a great proportion of difficult items although the subject's ability has remained the same. The total score which may be computed for each item also depends on the ability of the particular sample used (Rasch, 1960). Due to its specific logistic formulation, the Rasch model transforms the total score of each subject into a measure independent of the relative difficulty of the particular items considered and, similarly, it transforms the total score of each item into a measure independent of the ability of the particular sample considered. This is possible because the Rasch model defines the relative item difficulties, that is, it is the difference between item difficulties that is invariant. Then, by fixing an arbitrary origin, person measures can be estimated independently of the difficulty of the particular items encountered. There is currently no other mathematical formulation that allows the subject measures and the item difficulties to be estimated independently of one another (Wright & Stone, 1979). That's why the Rasch model is the only model to date that approximates objective measurement in health sciences provided that the observed data fit the model's requirements (Rasch, 1960).

The specific purposes of the study

The purpose of the present work is to study hand impairments and manual ability in children with CP as well as to clarify their relationship. To reach these objectives, the normal development of manipulative functions was firstly studied to provide normative data required to appraise the performance of children with CP. A new scale measuring a child's manual ability was secondly developed using the Rasch model and its invariance was tested across relevant demographic and clinical subgroups of CP children. Finally, hand impairments in children with CP were described and their relationship with manual ability were investigated.

Chapter 1 examines the development of fine finger dexterity and gross manual dexterity in 500 healthy children and adolescents. The results show that fine finger dexterity significantly increases with age until 10 years old and gross manual dexterity until 17-18 years old. The improvement of manipulative functions is thought to reflect the maturation of neuromotor systems, especially the corticospinal tract, as well as the development of more efficient motor strategies. The influence of handedness and sex on dexterity is also addressed. This first study provides all normative data required to adequately appraise the disruption in manipulative functions accompanying CP according to age, sex, and handedness. Chapter 1 is presented as it has been submitted for publication in *Developmental Medicine Child Neurology*.

Chapter 2 presents the development and the validation of ABILHAND-Kids, a Rasch-built measure of perceived manual ability in children with CP. The ABILHAND-Kids questionnaire was submitted to about a hundred children with CP and their parents, and resubmitted to both groups after one month. A Rasch analysis shows that parents had a finer perception of their children's manual ability than the children themselves, leading to a scale with a wider measurement range and a higher reliability. ABILHAND-Kids was therefore exclusively built on the parents' perceptions. The implementation of the Rasch model on parents' responses provided both a measure of the difficulty hierarchy of the 21 ABILHAND-Kids activities and a measurement of the manual ability of each CP child. The validity and reproducibility of ABILHAND-Kids was also assessed. The development and the validation of ABILHAND-Kids is presented as it was published in *Neurology*. A second section of chapter 2 examines the invariance of ABILHAND-Kids through differential item functioning tests. The difficulty hierarchy of the 21 ABILHAND-Kids activities was compared across children's subgroups (e.g., boys vs. girls, younger vs. older, etc.). Overall, the item difficulty hierarchy was invariant across various demographic and clinical subgroups of children with CP. This means that the ABILHAND-Kids questionnaire can be used to measure manual ability in children with CP whatever their age, sex, handedness, school education, type of CP, gross motor function, manual ability, and hand impairments.

Chapter 3 describes hand impairments in children with CP and investigates their relationship with manual ability as measured with ABILHAND-Kids. The results show that motor functions were more commonly impaired than sensory functions. Grip strength, gross manual dexterity, fine finger dexterity, and stereognosis of both hands were significantly but moderately correlated with manual ability measures. In contrast, tactile pressure detection and proprioception were unrelated to manual ability in children with CP. The gross manual dexterity on the dominant hand and the grip strength on the non-dominant hand were the best independent predictors of the manual ability of children with CP. However, this combination of hand functions predicted only 58% of the variance in manual ability measures. This indicates that the manual ability of children with CP cannot simply be inferred from their hand impairments but must be measured per se. Chapter 3 is presented as it has been submitted for publication in *Neurology*.

Chapter 1
Development of fine finger and gross manual dexterity in healthy children and adolescents.

Abstract

The present investigation was conducted to investigate the developmental changes occurring with age in fine finger and gross manual dexterity as well as the influence of handedness and sex on these two types of dexterity. The fine finger dexterity of 335 healthy subjects from 6 to 14 years old (mean age ± SD, 10 ± 2 years; 161 males) was measured with the Purdue Pegboard Test. The gross manual dexterity of 509 healthy subjects from 6 to 20 years old (mean age ± SD, 13 ± 4 years; 239 males) was measured with the Box and Block Test. Fine finger dexterity significantly increased with age until 10 years old and gross manual dexterity until 17-18 years old. On average, the dominant hand presented a higher dexterity than the non-dominant hand. Females had a better fine finger dexterity than males but only for the dominant hand. The gross manual dexterity of males and females was not significantly different. The knowledge of the developmental changes occurring with age in fine finger and gross manual dexterity as well as the influence of handedness and sex gives health professionals the opportunity to determine properly the extent to which a child's or an adolescent's dexterity deviates from normal.

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Introduction

Dexterity can be defined as "the manual skill that requires rapid coordination of fine and gross voluntary movements based on a certain number of capacities developed through learning, training, and experience" (Poirier, 1987). In 1949, Super (1949) distinguished fine from gross dexterity stating that the first mainly involved wrist and finger movements while the second involved arm and hand coordination. Two factor analytic studies have also suggested that finger and manual dexterity are different psychomotor abilities (Fleishman, 1954; Fleishman & Ellison, 1962). More recently, Desrosiers et al. (1994) differentiated fine finger dexterity (also called fine, finger, or digital dexterity) from gross manual dexterity (also called manual dexterity) in terms of the fineness of the movements. According to them, fine finger dexterity involved fine interdigital movements while gross manual dexterity involved more global movements of the hand and the fingers.

Fine finger and gross manual dexterities are crucial in the achievement of almost all activities in daily living (Exner, 1990; Penta et al., 2001) but may be impaired at any age because of brain lesions, peripheral neuropathies or other aetiologies. Therefore, it is important to measure the actual level of dexterity dysfunction in order to set treatment goals, establish effective rehabilitation programs, and follow the efficiency of the treatments intended to improve dexterity. Over the years, several instruments were developed to measure fine finger and gross manual dexterity dysfunctions. Among these instruments, the Purdue Pegboard Test (PPT) (Tiffin & Asher, 1948) and the Box and Block Test (BBT) (Cromwell, 1960), which are frequently used in rehabilitation to measure respectively fine finger and gross manual dexterity, have been established as reliable and valid (Tiffin & Asher, 1948; Cromwell, 1960; Mathiowetz et al., 1985). Moreover, they were reported as sensitive enough to identify dexterity dysfunctions in various impaired populations (Goodkin et al., 1988; Desrosiers et al., 1994; Penta et al., 2001), including children (Cromwell, 1960; Gardner & Broman, 1979; Duqué et al., 2003). The measurement of dexterity dysfunctions in impaired children needs to take the developmental changes occurring with age into account since manipulative skills improve as children grow up. The development of children's fine finger dexterity as measured with the PPT has been examined in several studies (Costa et al., 1964; Gardner & Broman, 1979; Wilson et al., 1982; Brito & Santos-Morales, 2002) by using a one-trial administration which is less reproducible than a three-trial administration (Tiffin & Asher, 1948; Buddenberg & Davis, 2000). Only one study (Mathiowetz et al., 1986a) has applied the three-trial administration of the PPT to establish normative data in adolescents aged between 14 and 19 years old. The investigation of the development of fine finger dexterity as measured with the PPT is therefore required for children younger than 14 years old. Similarly, the development of gross manual dexterity as measured with the BBT was never investigated for subjects younger than 20 years old except for children aged between 7 and 9 years old (Smith, 1961). Sex and handedness may also influence the children's dexterity and, therefore, should be taken into account when measuring the actual level of dexterity dysfunction in impaired children.

The present study was conducted to investigate the developmental changes occurring between 6 and 14 years old in fine finger dexterity and between 6 and 20 years old in gross manual dexterity as well as the influence of sex and handedness so that health professionals can differentiate the real dexterity dysfunctions of impaired children from their normal difficulties attributable to age, sex, and handedness.

Methods

Participants

The study was authorized by the ethics committee of the Université catholique de Louvain, Faculty of Medicine, Brussels, Belgium. All subjects were recruited in various sports centres, primary schools, and colleges located in the center and the south of Belgium. After having the authorization of the establishments, all children of the sport centers were assessed on the occasion of their sports course; only primary school children whose parents gave their written informed consent were assessed; and only undergraduates contacted at the end of an academic course and prospective undergraduates contacted during a pre-enrolment meeting who accepted to participate in the study were assessed. All participants were tested individually by the same examiner in a quiet room from January 2003 to March 2005.

The fine finger dexterity was assessed in 335 healthy subjects (161 males, 174 females) aged between 6 and 14 years old and the gross manual dexterity in 509 healthy subjects (239 males, 270 females) aged between 6 and 20 years old. All children who were assessed for fine finger dexterity test were also assessed for gross manual dexterity. Eighty-six percent of the subjects were right-handed and 14% left-handed. All subjects were free from disease or injury that could affect their upper limb dexterity.

Instruments and testing procedures

The subjects were tested individually in a quiet room. They were instructed how to perform each test. Both hands were measured, starting with the dominant hand. Handedness was determined by writing hand preference.

Fine finger dexterity

The fine finger dexterity was measured with the Purdue Pegboard Test (Lafayette Instrument Model 32020) (Tiffin & Asher, 1948) according to the procedure described by Mathiowetz et al. (1986a). The subjects were instructed to pick up the pegs (length: 2.5cm; diameter: 2.5mm), one at a time, from a cup and to place them into the holes (diameter: 3mm) of a board as quickly as possible within a 30-second time period. The subjects were allowed to pick up and place three or four pegs for practice before the test began. They subsequently performed the test three times with each hand, alternating the dominant hand (DH) and the non-dominant hand (NDH). The PPT score was determined for each hand as the mean value of the maximum number of pegs picked up and placed within 30 seconds that was observed during the three trials.

Gross manual dexterity

The gross manual dexterity was measured with the Box and Block Test (Cromwell, 1960) according to the procedure described by Mathiowetz et al. (1985). The subjects were instructed to grasp the blocks (cube side: 2.5cm) individually from one compartment of a box, to transport them over a partition, and to release them into the opposite compartment of the box as quickly as possible within a 60-second time period. The subjects were allowed to transport the blocks within a 15-second time period for practice before the test began. They subsequently performed the test one time with each hand, starting with the DH. The BBT score is the maximum number of blocks transported within one minute.

Statistical analysis

Repeated measures analyses of variance (RM-ANOVA) with one within-subjects factor (handedness) and two between-subjects factors (sex and age) were conducted to detect the main and interaction effects of handedness, sex and age. Subsequently, significant main and interaction effects detected by the RM-ANOVA were investigated through paired t-tests (for handedness factor), t-tests (for sex factor), and pairwise multiple t-test comparisons (for age factor). The alpha level of significance was set at 0.05 for all statistical tests except for pairwise multiple t-test comparisons which required a Bonferroni's adjustment of the alpha level of significance ($0.05/36 = 1.38^{-03}$ and $0.05/105 = 4.76^{-04}$, respectively for PPT and BBT) to ensure that the overall Type 1 error rate remains at 0.05.

Results

Fine finger dexterity

Table 1 presents the results of the RM-ANOVA on the PPT data. Significant differences in the PPT scores were observed for handedness, sex, and age. Moreover, a significant interaction between handedness and sex was also found. All other interaction effects were not statistically significant. The results of the RM-ANOVA indicated that the fine finger dexterity normative data of the 335 healthy subjects needed to be stratified by handedness, sex, and age. Table 2 reports the mean, the standard deviation (SD), the lowest and the highest values (Low and High), and the 95% of confidence interval ($CI_{95\%}$) of the PPT scores for each age group according to handedness and sex.

Table 1: Repeated measures ANOVA on the PPT data

Factors	F-statistic	df	*P*
Handedness	314.94	1	< 0.001*
Sex	13.65	1	< 0.001*
Age	65.77	8	< 0.001*
Handedness by Sex	6.21	1	0.013*
Handedness by Age	0.71	8	0.680
Sex by Age	0.61	8	0.768
Handedness by Sex by Age	0.38	8	0.933

* Significant effect

Table 2: Normative data on the PPT (N = 335)

		Males						Females					
Age	Hand	N	Mean*	SD	Low	High	$CI_{95\%}$	N	Mean*	SD	Low	High	$CI_{95\%}$
6	DH	15	11.6	1.31	9.3	14.3	10.8 - 12.3	15	12.5	0.96	10.0	14.3	11.9 - 13.0
	NDH		10.6	0.95	9.0	12.0	10.0 - 11.1		10.9	1.20	8.7	12.7	10.2 - 11.5
7	DH	15	12.6	1.08	10.7	14.7	12.0 - 13.2	16	13.6	1.42	11.7	16.7	12.8 - 14.3
	NDH		11.6	1.64	8.7	14.3	10.7 - 12.5		11.9	1.45	10.0	15.0	11.1 - 12.7
8	DH	15	13.9	1.09	12.3	16.7	13.3 - 14.5	19	14.4	1.18	12.0	16.7	13.9 - 15.0
	NDH		12.3	1.24	10.0	14.7	11.7 - 13.0		12.6	1.36	10.7	16.0	11.9 - 13.2
9	DH	15	14.6	0.72	13.0	16.0	14.2 - 15.0	32	15.1	1.22	12.7	18.0	14.7 - 15.6
	NDH		13.2	1.19	11.3	15.0	12.5 - 13.9		13.6	1.48	10.0	17.0	13.1 - 14.1
10	DH	26	15.5	1.08	13.0	17.0	15.1 - 16.0	23	15.8	1.34	13.3	18.0	15.2 - 16.4
	NDH		14.6	1.18	12.3	18.3	14.1 - 15.1		14.2	1.47	11.3	17.3	13.6 - 14.9
11	DH	27	16.0	1.50	12.7	19.0	15.5 - 16.6	24	16.5	1.37	13.3	18.7	15.9 - 17.0
	NDH		14.8	1.49	12.0	17.3	14.2 - 15.3		15.1	1.47	11.7	17.0	14.5 - 15.7
12	DH	18	16.5	1.02	14.7	18.3	16.0 - 17.0	15	16.9	1.78	12.7	19.7	15.9 - 17.8
	NDH		15.3	1.68	11.7	19.0	14.4 - 16.1		15.5	2.16	12.0	19.0	14.3 - 16.7
13	DH	15	16.0	1.70	11.7	18.0	15.0 - 16.9	15	16.9	1.54	15.0	20.7	16.0 - 17.7
	NDH		15.1	1.44	12.7	18.3	14.3 - 15.9		15.6	1.30	12.3	17.7	14.9 - 16.3
14	DH	15	15.9	1.24	13.7	18.3	15.2 - 16.6	15	17.1	1.53	13.3	19.0	16.3 - 18.0
	NDH		15.0	1.14	13.0	18.0	14.4 - 15.6		15.9	1.38	12.3	18.0	15.1 - 16.6

* Average of three trials.
DH = dominant hand; NDH = non-dominant hand; N = number of subjects; SD = standard deviation;
$CI_{95\%}$ = 95% of confidence interval.

A significant difference between the DH and the NDH was observed for males (DH: mean = 14.9, SD = 1.97; NDH: mean = 13.8, SD = 2.05; $p < 0.001$) and females (DH: mean = 15.4, SD = 1.94; NDH: mean = 13.9, SD = 2.12; $p < 0.001$). The DH made it possible to pick up and place, on average, 1.1 extra pegs for males and 1.5 extra pegs for females when compared with the NDH. Females presented a slightly better fine finger dexterity than males. However, a significant difference between males and females was only observed on the DH (DH: $p = 0.02$; NDH: $p = 0.56$) accounting for the significant interaction effect observed between handedness and sex.

The development of fine finger dexterity from 6 to 14 years old is illustrated in Figure 1 according to handedness and sex. Pairwise multiple t-tests comparisons indicated that, generally, all age groups between 6 and 10 years old were significantly different ($p < 1.38^{-03}$) except with their adjacent age groups. On the contrary, all age groups between 10 and 14 years old were not significantly different ($p > 1.38^{-03}$). It emerges from these results that, for both the DH and the NDH, fine finger dexterity significantly increases in both males and females until the age of 10, and thereafter continues to improve until 12 years old although no statistically significant change occurred.

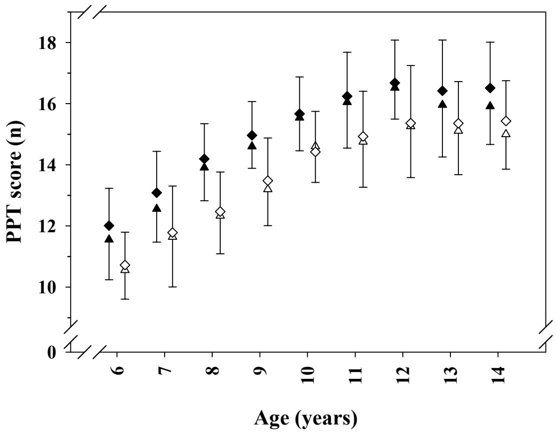

Figure 1. Development of fine finger dexterity from 6 to 14 years old. Mean PPT scores (symbols) and their standard deviations (bars) are plotted as a function of age. The normative data are separately reported for males (triangles) and females (diamonds) according to their handedness (dominant hand: black symbols; non-dominant hand: white symbols).

Gross manual dexterity

Table 3 presents the results of the RM-ANOVA on the BBT data. Significant differences in the BBT scores were observed for handedness and age. The gross manual dexterity of males (DH: mean = 64.0 , SD = 14.90; NDH: mean = 62.7, SD = 13.94) and females (DH: mean = 64.9, SD = 15.07; NDH: mean = 62.9, SD = 14.47) was not significantly different (DH: $p = 0.494$; NDH: $p = 0.909$). Moreover, no significant interaction effect was observed. Consequently, male and female normative data were combined. Table 4 reports the mean, the standard deviation, the lowest and the highest values, and the 95% of confidence interval of the BBT scores for each age group according to handedness.

Table 3: Repeated measures ANOVA on the BBT data

Factors	F-statistic	df	*P*
Handedness	50.22	1	< 0.001*
Sex	0.01	1	0.923
Age	177.95	14	< 0.001*
Handedness by Sex	2.90	1	0.089
Handedness by Age	1.52	14	0.098
Sex by Age	0.98	14	0.469
Handedness by Sex by Age	1.02	14	0.434

* Significant effect

The performance on the BBT was significantly better with the DH (DH: mean = 64.5, SD = 14.98; NDH: mean = 62.8, SD = 14.21; $p < 0.001$). Indeed, the DH made it possible to transport, on average, 1.7 extra blocks when compared with the NDH. The development of gross manual dexterity from 6 to 20 years old is illustrated in Figure 2 according to handedness. Pairwise multiple t-tests comparisons showed that, in general, all age groups between 6 and 10 years old were significantly different ($p < 4.76^{-04}$) except with their adjacent age groups. They also indicated that the BBT scores sequentially increased, from 10 to 17 years old, in three steps (1st step = 11-12 years old; 2nd step = 13-14 years old; 3rd step = 15-16-17 years old). Finally, all age groups after 17 years old were not significantly different ($p > 4.76^{-04}$). It emerges from these results that, for both the DH and the NDH, gross manual dexterity significantly increases until the age of 17, and thereafter slightly improves until 18 years old although no statistically significant change occurred.

Table 4: Normative data on the BBT (N = 509)

Age	Hand	N	Mean	SD	Low	High	$CI_{95\%}$
6	DH	30	41.5	5.49	33	53	39.4 - 43.5
	NDH		40.6	3.87	34	50	39.2 - 42.0
7	DH	31	44.9	6.67	34	64	42.4 - 47.3
	NDH		46.0	5.92	33	57	43.9 - 48.2
8	DH	34	50.1	5.82	39	61	48.1 - 52.2
	NDH		49.3	5.81	38	64	47.3 - 51.4
9	DH	47	52.8	6.09	41	68	51.0 - 54.6
	NDH		51.1	5.67	40	63	49.4 - 52.8
10	DH	49	56.4	7.05	40	72	54.3 - 58.4
	NDH		54.4	5.98	44	68	52.7 - 56.1
11	DH	51	61.2	8.54	41	84	58.8 - 63.6
	NDH		58.9	7.74	42	75	56.7 - 61.0
12	DH	33	61.4	7.63	46	74	58.7 - 64.1
	NDH		60.2	7.51	45	77	57.5 - 62.8
13	DH	30	68.6	7.59	51	83	65.8 - 71.5
	NDH		66.4	6.29	53	83	64.1 - 68.8
14	DH	30	68.6	6.59	55	81	66.1 - 71.0
	NDH		67.9	6.94	53	81	65.3 - 70.5
15	DH	32	76.7	6.51	65	90	74.4 - 79.0
	NDH		73.2	6.58	62	87	70.8 - 75.6
16	DH	30	76.9	5.80	67	89	74.7 - 79.1
	NDH		75.4	6.07	65	89	73.1 - 77.6
17	DH	27	78.6	5.26	69	92	76.6 - 80.7
	NDH		76.4	5.14	63	88	74.3 - 78.4
18	DH	28	82.9	6.60	69	94	80.4 - 85.5
	NDH		81.2	5.37	70	93	79.1 - 83.3
19	DH	28	83.5	4.21	73	91	81.9 - 85.1
	NDH		81.0	5.08	70	92	79.0 - 83.0
20	DH	29	84.4	6.71	70	98	81.8 - 86.9
	NDH		81.6	5.66	68	95	79.5 - 83.8

DH = dominant hand; NDH = non-dominant hand;
N = number of subjects; SD = standard deviation;
$CI_{95\%}$ = 95% of confidence interval.

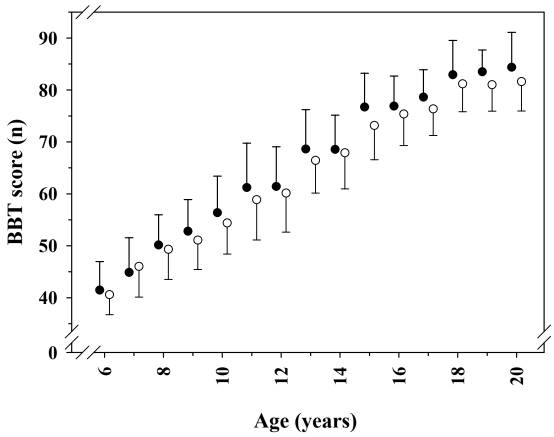

Figure 2. Development of gross manual dexterity from 6 to 20 years old. Mean BBT scores (symbols) and their standard deviations (bars) are plotted as a function of age. The normative data are reported according to subjects' handedness (dominant hand: black symbols; non-dominant hand: white symbols).

Discussion

The developmental changes occurring with age as well as the influence of handedness and sex on fine finger and gross manual dexterity were investigated from childhood to early adulthood using the PPT and the BBT, respectively. Age and handedness influence the performance on both fine finger and gross manual dexterity tests while sex affects only the performance of the DH on the fine finger dexterity test. Additionally, the current study has provided PPT norms for subjects aged between 6 and 14 years old and BBT norms for subjects aged between 6 and 20 years old allowing health professionals to differentiate dysfunctional from normal dexterity.

Age effect

Fine finger and gross manual dexterities gradually improve with age during childhood and adolescence, a finding supported by other studies (Costa et al., 1964; Gardner & Broman, 1979; Wilson et al., 1982; Brito & Santos-Morales). For both the DH and the NDH, the fine finger dexterity of males and females significantly improves until the age of 10 (see Figure 1). This is in agreement with a previous study (Costa et al., 1964) in which little change was found in the PPT scores above the age of 10 years old. Moreover, the level-off observed in fine finger dexterity at the age of 10 years old seems to be supported by several studies which found that some features of the corticospinal system reach adult values around 10 years (Koh & Eyre, 1988; Müller et al., 1991; Forssberg, 1999; Fietzek et al., 2000). The major role of the corticospinal tract in fine finger dexterity has been highlighted by phylogenetic studies (Porter & Lemon, 1993) as well as by lesion studies in animals (Porter & Lemon, 1993) and in humans (Porter & Lemon, 1993; Duqué et al., 2003; Lang & Schieber, 2004). The latter have shown that the ability to perform independent finger movements was severely affected by damage to the corticospinal tract. The maturation of the corticospinal tract is generally assessed in humans with transcranial magnetic stimulation (TMS). A significant relationship between the central motor conduction time (CMCT) as measured with TMS and the performance in a peg-transportation test was found in relaxed subjects aged between 2 and 13 years old (Müller & Hömberg, 1992). Relaxed CMCT decreases with age until around 10 years old when adult values are reached (Müller et al., 1991; Fietzek et al., 2000). This rapid decrease of the relaxed CMCT during the first decade probably reflects the maturation of the synaptic excitability at the cortical level and morphologic maturation such as the myelination of corticospinal axons and the growth of axon diameters (Müller et al., 1991; Fietzek et al., 2000). The stimulation threshold for TMS, which reflects myelination processes and synaptic efficacy within the motor cortex, has also been reported to decrease until around the age of 10 years (Koh & Eyre, 1988; Müller et al., 1991). Additionally, the density of the corticospinal connections gradually increases with age up to 10 years old (Forssberg, 1999). Although all of these studies support the level-off observed in fine finger dexterity at the age of 10 years old, further research is required to examine the relationship between the maturation of the corticospinal system and the acquisition of fine finger dexterity.

Gross manual dexterity of both hands gradually increases with age up to 10 years old and after that continues to sequentially improve until 17-18 years old (see Figure 2). It is not clear why gross manual dexterity reaches adult values later than fine finger dexterity. The performance on the PPT mainly involves distal muscles to execute independent finger movements which largely depend on the primary cortex (M1) and on its corticospinal projections (Forssberg, 1998). On the contrary, the performance on the BBT also involves proximal muscles which are mainly controlled by subcortical structures (Forssberg, 1998). Consequently, the level-off observed in gross manual dexterity at the age of 17 years old might reflect the maturation of other neuromotor systems than the corticospinal tract. The strategies used to perform the BBT task also become more efficient with age whereas the development of strategies with age was not observed for the PPT. In the BBT, young children turned their trunk during the transport of the blocks from one compartment of the box to the other and kept a visual control on the blocks during the whole movement. On the contrary, the adolescents moved only their upper limbs and focused their gaze on the compartment of the box in which the blocks were picked up. The development of appropriate strategies such as postural adjustments might be responsible for the latter level-off observed in the BBT.

Handedness effect

The better performance observed with the DH in tasks involving fine finger and gross manual dexterity confirms previous reports (Wilson et al., 1982; Brito & Santos-Morales, 2002; Duqué et al., 2003) and is consistent with the asymmetrical functions of the hands observed in daily activities, with the DH playing a manipulative role and the NDH playing a postural role (Guiard, 1987). Both PPT and BBT measure the manipulative skill rather than the postural skill favoring the DH over the NDH. Furthermore, the dominant upper limb also demonstrates a more efficient control of intersegmental dynamics than the non-dominant upper limb (Bagesteiro & Sainburg, 2002). The asymmetrical functions of the hands as well as the asymmetry in the dynamic control of the upper limbs have been suggested to reflect asymmetrical neural mechanisms (Hammond, 2002). Several studies have shown (at least for right-handers) that, when compared with the hemisphere controlateral to the NDH, the hemisphere controlateral to the DH has a larger M1 and a slightly larger corticospinal tract (Amunts et al., 1996; Hammond, 2002; Reilly & Hammond, 2004). Cytoarchitectonic techniques have also demonstrated a microstructural asymmetry in M1. Indeed, the dominant hemisphere presents a smaller cell volume density in M1 with a corresponding greater neuropil volume containing axons, dendrites, and synapses (Amunts et al., 1996 and 1997). The larger neuropil volume within the M1 of the dominant hemisphere suggests a greater number of intracortical connections which play an important role in the reorganization that occurs within M1 as a result of motor practice (Hammond, 2002; Reilly & Hammond, 2004). Consequently, a particular amount of motor practice should lead to more effective fine and gross voluntary movements (i.e., more effective dexterity) of the DH than of the NDH (Hammond, 2002; Reilly & Hammond, 2004) due to the more extensive connectivity observed in the dominant hemisphere and the related richer experience-based reorganization in M1. In agreement with this theory, one study (Garry et al., 2004) has shown that only the corticomotor excitability changes (induced by a motor practice) of the hemisphere controlateral of the DH were related to the performance improvement observed in the PPT. A magnetoencephalographic study (Volkmann et al., 1998) has also reported an asymmetrical topographical organization of hand movement representation in the motor cortex. Indeed, the hand representation in M1 was larger in the dominant hemisphere than in the non-dominant hemisphere. This finding is consistent with the study of Nudo et al. (1992) who used intracortical microstimulation techniques on adult squirrel monkeys to obtain the representational maps of their distal forelimb movements in M1. The digit and wrist representations of the monkeys were larger, contained a greater number of discrete sites, and were topographically more complex in the dominant hemisphere. It may be suggested that the enlarged representation of hand movements in the dominant hemisphere would provide the neural substrate for a "more refined motor skill repertoire of the DH" (Volkmann et al., 1998).

Sex effect

Consistent with a study performed in elderly people (Desrosiers et al., 1994), the gross manual dexterity of males and females is not significantly different. On the contrary, an effect of sex is observed in the task involving fine finger dexterity. This is in accordance with a rhesus monkey study showing that females were significantly faster than males in a candy retrieval task from complex rods (double-S-shaped rod, question mark-shaped rod) but not from a simple rod (straight rod) (Lacreuse et al., 2005). Consequently, it is possible that the BBT which requires less fine interdigital movements than the PPT is not difficult enough to detect subtle sex differences.

The effect of sex in tasks involving fine finger dexterity is inconsistently reported in the literature. Some authors have reported a significantly better performance for females than for males (Mathiowetz et al., 1986a; Brito & Santos-Morales, 2002) or stated that females have a slightly better fine finger dexterity than males without giving any information about the existence of a statistically significant difference (Sattler & Engelhardt, 1982). Other investigators have found no significant sex effect (Costa et al., 1964; Wilson et al., 1982). These contradictory findings probably suggest small sex differences. In addition, almost none of the studies has investigated a possible interaction effect between sex and handedness. The current study shows that females present a significantly but slightly better fine finger dexterity than males for the DH but not for the NDH. This finding is similar to the PPT results observed in a recent longitudinal study (Takser & Dellatolas, 2002). Sex differences in brain have been studied very little in humans. However, sex differences were observed in the cortical neuropil volume which is larger in females than in males (Rabinowicz et al., 1999). This suggests a more extensive connectivity in females' brain that results in a more effective experience-based reorganization in brain (Hammond, 2002; Reilly & Hammond, 2004). The same amount of daily motor practice should therefore lead to more efficient voluntary movements in females than in males. This females' advantage may particularly concern the DH as manipulative skills are achieved day after day by this hand (Guiard, 1987; Reilly & Hammond, 2004) which thus benefits from a greater amount of motor practice than the NDH. This may explain why the manipulative performance assessed with the PPT is better in females than males for the DH but not for the NDH. However, further investigations are required to examine in practice the functional significance of the sex differences observed in the cortical neuropil volume.

Chapter 2
ABILHAND-Kids: a measure of manual ability in children with cerebral palsy

2.1. Development and validation of ABILHAND-Kids

Abstract

Objective: To develop a clinical tool for measuring manual ability (ABILHAND-Kids) in children with cerebral palsy (CP) using the Rasch measurement model.

Methods: The authors developed a 74-item questionnaire based on existing scales and experts' advice. The questionnaire was submitted to 113 children with CP (59% boys; mean age, 10 years) without major intellectual deficits (IQ>60) and to their parents, and resubmitted to both groups after 1 month. The children's and parents' responses were analyzed separately with the WINSTEPS Rasch software to select items presenting an ordered rating scale, sharing the same discrimination, and fitting a unidimensional scale.

Results: The final ABILHAND-Kids scale consisted of 21 mostly bimanual items rated by the parents. The parents reported a finer perception of their children's ability than the children themselves, leading to a wider range of measurement, a higher reliability (R = 0.94) and a good reproducibility over time (R = 0.91). The item difficulty hierarchy was consistent between the parents and the experts. The ABILHAND-Kids measures are significantly related to school education, type of CP, and gross motor function.

Conclusions: ABILHAND-Kids is a functional scale specifically developed to measure manual ability in children with CP providing guidelines for goal setting in treatment planning. Its range and measurement precision are appropriate for clinical practice.

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Introduction

Cerebral palsy (CP) is the most common cause of physical disability in children (Rosenbaum, 2003). While the rates of perinatal and infant mortality have declined toward the end of the last century (Parkes et al., 2001), the rate of CP has remained at 2 to 2.5 per 1,000 live births (Stanley et al., 2000). Medical and technological advances have enabled a cohort of children with severe CP impairments to survive (Stanley et al., 2000). Despite the non-progressive nature of their motor impairment syndromes, their clinical picture may change over time (Kuban & Leviton, 1994). A variety of treatments (Boyd et al., 2001) are intended to improve the child's functioning in their relevant environments, usually at home or school (Young et al., 2000). However, the effectiveness of these treatments is debated (Pagliano et al., 2001). There is a need to quantify the efficacy of a treatment and to follow a child's status over time (Young et al., 1995). The first step in this process is to develop clinically relevant, valid, and reliable outcome measures (Rosenbaum et al., 1990).

Although impaired arm and hand function are the main problems in about half the children with CP (Uvebrant, 1988), there is a lack of appropriate instruments for measuring the ability of the children to use their hands in daily activities (Pagliano et al., 2001) since most scales are focused on the lower limb function. Moreover, the existing pediatric scales focused on fine motor functions (e.g., Pediatric Evaluation of Disability Inventory (Haley et al., 1992), Activities Scale for Kids (Young et al., 2000)) are not validated for children with CP. It is important to have evaluation instruments that are specifically applicable to the population being studied (Rosenbaum et al., 1990) and the purpose of this study is to develop the ABILHAND-Kids questionnaire, a measure of manual ability in children with CP.

Manual ability is a behavior. It can be defined as "the capacity to manage daily activities requiring the use of the upper limbs, whatever the strategies involved" (Penta et al., 2001). Manual ability is based upon upper limb function, but it also involves environmental (e.g., assisting devices, school education) or personal (e.g., motivational, cognitive and emotional status, compensatory behaviors) contextual factors (World Health Organization, 2001). Therefore, manual ability cannot be observed directly; but it can be inferred from a patient's perception of the difficulty of performing manual activities (Penta et al., 2001). Adult patients who are most familiar with their own functional limitations are commonly considered as the gold standard to report their health status (Pierre et al., 1998). The use of parents as valid proxy reporters is advocated not only for very young children but also for school children and adolescents (Vogels et al., 1998), even though children at these ages have the ability to adequately communicate their perceptions (Verrips et al., 2001). The ABILHAND-Kids questionnaire was submitted to both children and their parents, in order to compare the reliability of the reported perceptions.

Once the subjects' perceptions were collected, a linear measure (Merbitz et al., 1989) of manual ability was obtained according to probabilistic measurement models, the most promising being the Rasch model (Rasch, 1960). Provided that the behavioral data fit the requirements of the model, manual ability is measured as the log-odds of reported successful achievement in manual activities and is located on a linear scale. In this study, the ABILHAND-Kids questionnaire was developed to assess manual ability as perceived by children with CP or their parents. Its reproducibility was tested after a delay of approximately 1 month.

Subjects and methods

Subjects

The study was authorized by the ethics committee of the Université catholique de Louvain, Faculty of Medicine in Brussels, Belgium. The definition adopted for selecting children with cerebral palsy was "all non-progressive but often changing motor impairment syndromes secondary to lesions or anomalies of the brain arising in the early stages of its development" (Mutch, 1992).

Subjects older than 6 years were recruited to focus on children with mature manipulative skills in activities of daily life (Illingworth, 1975). Age, sex, handedness, level of school education, type of CP, and the Gross Motor Function Classification System (GMFCS) (Palisano et al., 1997) were included as independent demographic and clinical indices in the validation analysis. The children, recruited through seven centers specialized in CP, exhibited a wide range in each index. Moreover, given that ABILHAND-Kids was designed as an interview-based questionnaire, children with a major intellectual deficit (IQ < 60) were excluded[1]. As a result, 113 children with CP (67 boys and 46 girls; mean age, 10 years) were assessed by the same examiner. The sample description is provided in Table 1.

Table 1. Sample description (n = 113)

Age, y, mean (range)	10 (6-15)
Sex	
Male	67
Female	46
Handedness	
Right	55
Left	57
Ambidextrous	1
School education	
Mainstream	47
Type 1: mild mental retardation	1
Type 4: physical handicap	58
Type 8: learning disabilities	6
Home	1
Type of CP	
Tetraplegia/paresis	35
Diplegia	24
Hemiplegia/paresis	
Right	26
Left	28
GMFCS	
Level I: most independent motor function	50
Level II	26
Level III	12
Level IV	21
Level V: least independent motor function	4

CP = cerebral palsy; GMFCS = Gross Motor Function Classification System.

[1] As IQ tests are time-consuming, the IQs of CP children were not assessed within the framework of the study. They were obtained from the children's medical records and thus were, in most cases, solely available for children placed in special schools. However, it is reasonable to think that children placed in mainstream schools have no major intellectual deficit. Moreover, the IQs that were available came from different tests (e.g., WPPSI, WPPSI-R, WISC-III, WISC-R, KABC). It was therefore not possible to exploit the IQ data except for the sample selection.

Questionnaire development

The ABIHAND-Kids questionnaire was designed to cover the widest range of children's manual activities including both unimanual and bimanual activities. The preliminary questionnaire included 41 items derived from the ABILHAND questionnaire already validated for adult patients (Penta et al., 1998 and 2001). Thirty additional items were selected from various existing scales: the Pediatric Evaluation of Disability Inventory (Haley et al., 1992), the Activities Scale for Kids (Young et al., 2000), the Denver Developmental Screening Test, versions I (Frankenburg & Dodds, 1967) and II (Frankenburg et al., 1992), and the Klein-Bell Activities of Daily Living Scale (Klein & Bell, 1982). Finally, 48 items were devised to extend the range of activities explored by the questionnaire. The pool of 119 items was submitted to 27 experts on children with CP (8 physicians, 11 physiotherapists, 7 occupational therapists, and 1 educator). The experts were asked to 1) determine the relevance of the activities; 2) estimate the difficulty of each activity for a CP child with moderate disorder on a three-level scale (very difficult/difficult/easy); and 3) devise pertinent activities not included in the original item set. Fifty items were eliminated either because the experts considered them irrelevant (46 items) or because the analysis of the experts' responses through the Rasch model showed that they did not contribute to the definition of a unidimensional variable (4 items). Finally, five items were added to the test following the experts' suggestions.

The experimental version of ABILHAND-Kids involved 74 items. The questionnaire was submitted to children with CP and their parents. They were also asked to suggest activities considered relevant in daily life but not already included in the questionnaire. None of the activities proposed by the children or their parents was added to the questionnaire because the suggestions were not exclusively related to the upper limbs (e.g., swimming, bicycling) and thus could involve factors other than just manual ability.

Instrument

The experimental version of ABILHAND-Kids explored unimanual and bimanual activities completed without technical or human assistance. For each question, the children and their parents were asked to provide their perceived difficulty irrespective of the limb(s) actually used to perform the activity on a three-level scale: impossible (0), difficult (1), or easy (2). Activities not attempted in the last 3 months were not scored and were encoded as missing responses (4% of the data for the children and 5% for their parents).

Procedures

The French version of the questionnaire was presented separately to children with CP and their parents. The 74 items were randomly presented. Each item was presented verbally to the child by the examiner, while the parents filled in the questionnaire themselves in another room. Fourteen percent of the children attended boarding school so their activities were not observed daily by their parents. In these cases, the questionnaire was completed by the occupational therapists on behalf of the parents. The test-retest reliability was investigated by submitting the questionnaire a second time, after a delay of 25 ± 13 days, to the children and their parents or occupational therapists.

Data analysis

Children's and parents' responses were analyzed separately with the WINSTEPS Rasch analysis computer program (Linacre & Wright, 1998). The Rasch model (Rasch, 1960) verified that successive response categories for each item represented increasing levels of ability and that thresholds between successive response categories are located in the anticipated order (Andrich, 1996b). It required that the probability of endorsing any response category to an item depended solely on the patient's ability, the item difficulty, and the threshold difficulties. Patient measures, item, and threshold difficulties were then located on a single real-number line representing the measurement scale. The Rasch model can also be used to verify that all items line up on a unidimensional scale (Andrich, 1988). Given the location of the parameters on the linear scale, the model recalculates the response expected for each subject to each item. The similarity between the observed and expected responses to any item is reported by the software through two fit statistics (Smith, 2000): 1) the outlier-sensitive fit statistic (OUTFIT), and 2) the information-weighted fit statistic (INFIT). The INFIT is more sensitive to unexpected responses from patients with an ability level near the item difficulty. The OUTFIT is more sensitive to unexpected responses from patients with an ability level far from the item difficulty. Another statistic, the point measure correlation coefficient (RPM), indicates the coherence of each item with the rest of the questionnaire. It is computed as the correlation coefficient between all patients' responses to an item and their measures on the overall questionnaire except for that particular item. Positive RPM values are expected when each item is coherent with the other questionnaire items.

Item selection

Starting from the 74 experimental items, indexes reported from successive analyses were used to select the items that constituted the final ABILHAND-Kids scale. Any item that did not meet any of the following criteria was eliminated.

An ordered rating scale

The subjects were asked to report their perceptions on a three-level scale: impossible (0), difficult (1), or easy (2). If the anticipated order of response categories was verified, subjects with a higher ability ought to select a higher response to any given item and subjects selecting a higher response for a given item ought to present a higher ability. When these conditions were not met, the order of thresholds between successive response categories was skewed, indicating that the rating scale was not used as anticipated for the particular item (Andrich, 1996b). Only items having thresholds in the anticipated order were retained.

All items share the same discrimination

Though all items were answered according to the same three-level rating, the threshold locations (relative to the item location) could vary across items. In this case, the items are perceived with a different discrimination (Wright, 1999; Linacre, 2000a). The difference in discrimination from one item to another complicates the clinical interpretation of scores since a given response has a different relative weight across all items. Therefore, items presenting a discrimination significantly different from the average (Z-test) were removed.

All items fit a unidimensional scale

Fit statistics (INFIT and OUTFIT) were used to detect items that did not satisfy the model requirement of unidimensionality[2]. The acceptable range of fit statistics for a sample of 113 subjects is between 0.80 and 1.20 for the INFIT and between 0.40 and 1.60 for the OUTFIT (Smith et al., 1998)[3]. Items presenting an INFIT lower than 0.80 or an OUTFIT lower than 0.40 are not considered to be a major threat to unidimensionality (Linacre, 2000b).

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[2] Various types of indicators have been developed to assess the fit of the data to the Rasch model (Wright & Panchapakesan, 1969; Andersen, 1973). The currently prevalent approach is based on the Pearson's chi-square (Wright & Panchapakesan, 1969). A residual is computed for the response of each person to each item as the difference between the observed response and the response expected by the Rasch model. Each residual is then squared and standardised according to the variance of the expected response to produce a chi-square statistic. The individual chi-squares are then averaged across all subjects to produce an approximate mean-square fit statistic for each item. Two averages of individual chi-squares are implemented in the Winsteps computer program, leading to a pair of mean-squares for each item. The first, obtained through unweighted averaging, is the outlier-sensitive fit statistic (OUTFIT) and is more sensitive to unexpected responses from subjects with an ability far from the item difficulty. The second, computed by weighing each individual chi-square by its variance, is the information-weighted fit statistic (INFIT) and is more sensitive to unexpected responses from subjects with an ability near the item difficulty. Both fit statistics have an expected value of 1 if the data perfectly fit the Rasch model's prescriptions. Values lower than 1 indicate that the observed responses are more deterministic than expected by the model (i.e., too predictable); values higher than 1 indicates that the observed responses are more random than expected by the model (i.e., too unpredictable).

[3] Several rules of thumb have been suggested to interpret fit statistics. Mean-square values in the range of 0.70-1.30 are traditionally considered as acceptable. Linacre & Wright (1994) have also provided some reasonable ranges for item mean-square fit statistics according to the type of the applied test. For instance, they propose a range of 0.50-1.70 for clinical observations but a range of 0.60-1.40 for rating scale surveys. Several simulation studies have, however, shown that the mean-square distributions were affected by the type of statistics (INFIT or OUTFIT) and various characteristics of the data (Smith, 1991; Smith et al., 1998; Smith, 2000; Karabatsos, 2000). Indeed, the Type I error rate of the fit statistics has been found to increase when the OUTFIT was used instead of the INFIT, and when the sample size (and to a lesser extent the test length) decreased (Smith et al., 1998; Smith, 2000). This means that the use of a single acceptable range for different testing situations may lead to both the overdetection and the underdetection of misfit due to the inconsistency of the Type I error rate. Richard Smith and collaborators (1998) have therefore suggested to calculate different acceptable ranges for the mean-square statistics depending on the type of fit statistics (INFIT or OUTFIT) and the sample size (n). INFIT mean-square values in the range of $1 \pm (2/\sqrt{n})$ and OUTFIT mean-square values in the range of $1 \pm (6/\sqrt{n})$ were presented as acceptable. These formulas were applied in the present study with our sample size of 113 subjects: INFIT range was 0.80-1.20 and OUTFIT range was 0.40-1.60. Note that the acceptable OUTFIT range is wider than the INFIT range to offset its higher Type I error rate. In the same way, narrower acceptable ranges will be observed for higher sample sizes to offset their lower Type I error rates (e.g., with $n = 500$, the acceptable OUTFIT range would be 0.73-1.27 and the INFIT range would be 0.91-1.09).

All items presenting an INFIT higher than 1.20 or an OUTFIT higher than 1.60 were removed.

Scale validation

To validate the difficulty hierarchy of the selected activities, four occupational therapists were independently asked to classify each as either 1) unimanual or 2) bimanual. Bimanual activities were further classified as either (2A) normally done with two hands but also manageable in several unimanual steps when using an adaptive strategy; (2B) requiring one hand to stabilize an object - not involving any finger - and the other hand to complete the activity; or (2C) requiring digital activity from both hands. In addition, the patient measures were validated according to the relationship between ABILHAND-Kids measures and different demographic (age, sex, handedness, school education) or clinical (type of CP, GMFCS) indices. A Pearson correlation coefficient was computed for continuous indices, a t-test for two groups of nominal indices, and a one-way analysis of variance for more than two groups of nominal indices. Finally, the item difficulty hierarchy between the parents and the 27 experts was compared through a differential item functioning (DIF) test (Wright & Stone, 1979).

Scale reliability

A person separation reliability coefficient was determined as the ratio between the true measure variance (as expressed by the SD corrected for measurement error) and the observed (true + error) measure variance in the sample (Wright & Masters, 1982). This index is analogous to the traditional internal consistency coefficient, Cronbach's alpha (Cronbach, 1951). Moreover, test-retest reliability of the parents' responses was determined by the Pearson correlation coefficient. The invariance in the item difficulty hierarchy across the first and the second assessment was also tested through a DIF test (Wright & Stone, 1979).

Results

Children's versus parents' perceptions

The children's and parents' responses to the questionnaire were analyzed separately. The analysis of the children's responses resulted in a 13-item questionnaire because most of the items (n = 54) showed a disordered rating scale. Moreover, five items did not fit a unidimensional scale and two items did not share the common discrimination. The analysis of the parents' responses resulted in a 21-item questionnaire because most of the items (n = 36) did not share the common discrimination. Moreover, 15 items did not fit a unidimensional scale and two items showed disordered thresholds. The analysis showed that the retained items were different in both groups and also that the manual ability was better discriminated by the children's parents as compared to the children themselves.

The subject measures and the item thresholds distributions for both the parents' and the children's scales are presented in Figure 1. The manual ability scales are calibrated in logits (i.e., log-odds units), a probability unit that expresses the natural logarithm of the odds of success (i.e., the pass/fail probability ratio). At any given ability level, a 1-logit difference between two children indicates that their odds of successful achievement of any activity are 2.7:1 (i.e., e^1:1), 2 logits have 7.4:1 odds, and 3 logits have >20:1 odds. The items are well targeted on the subjects in both scales. While both scales are able to successfully discriminate the manual ability of the subjects, the parents' scale covers a wider range than the children's scale, indicating a finer perception of item difficulties. Subjects measures are estimated over a range of 10.38 logits by the parents (leading to an odds ratio of over 32,000:1, i.e., $e^{10.38}$:1, between the most able and the least able child) while they cover only 7.54 logits according to the children's perceptions (leading to an odds ratio less than 2,000:1). Consequently, the subject measures can be discriminated with a greater than a 16 times higher resolution when using the parents' perceptions rather than the children's. Both scales present comparable floor and ceiling effects.

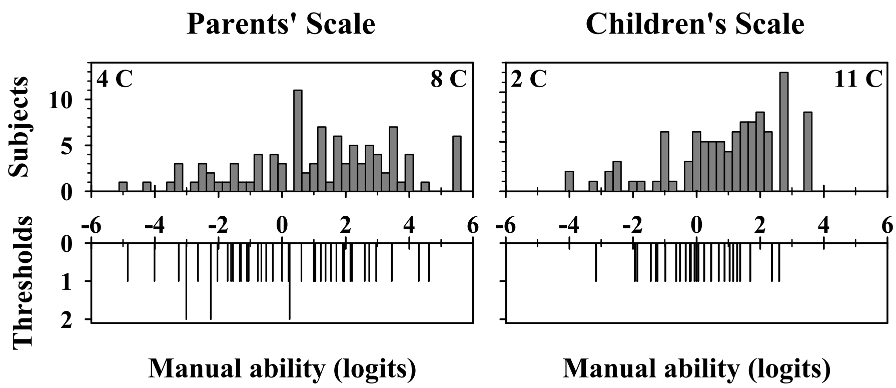

Figure 1. Manual ability scales as perceived by the children (13 items, 26 thresholds, right panel) and by their parents (21 items, 42 thresholds, left panel) and corresponding subjects distribution (top panels). The item threshold locations are well targeted on the subject measures on both scales. The floor and the ceiling effects are denoted by the number of children with extreme scores (either minimum or maximum) as marked on the upper corner of the distributions followed by a "C" (Children).

The children's lack of discrimination is emphasized even further when investigating the probability of each possible response ("impossible", "difficult", or "easy") to an item as a function of the child's ability. For any given item, a higher (i.e., easier) response is associated with a higher level of ability. However, the increase in manual ability required to answer "easy" rather than "impossible" is 2.66 times higher for the parents (3.24 logits) than for the children (1.22 logits). This indicates that the children's perception is more dichotomous; activities are perceived as either "impossible" or "easy" with very rare intermediate responses[4]. However, the parents report a finer perception on their children's manual ability. They allow a better separation of the subjects according to their manual ability and allow a more

4 Age related changes in children's perceptions were examined in our CP sample through the 74 items of the experimental questionnaire. Ordered thresholds, indicating that the intermediate response category is discriminated, were observed in 21 items for the 6-7 years old children (n = 26; average distance between the thresholds [$\bar{\tau}$] ± SD: 0.52 ± 0.60 logits), in 36 items for the 8-9 years old children (n = 27; $\bar{\tau}$ ± SD: 1.04 ± 0.82 logits), in 22 items for the 10-11 years old children (n = 31; $\bar{\tau}$ ± SD: 0.89 ± 0.75 logits), and in 41 items for the 12-15 years old children (n = 29; $\bar{\tau}$ ± SD: 1.21 ± 0.97 logits). The number of items presenting ordered thresholds as well as the average distance between the thresholds tend to increase with age suggesting that polytomous perception improves with age. One exception occurs for the 10-11 years old children which displayed a markedly more dichotomous perception than the 8-9 years old children. Currently, it is difficult to drawn solid conclusions about the age related changes in CP children's perceptions given the small number of children in each age subgroup. Further research with more children in each age subgroup is therefore required to specifically investigate this issue.

precise measurement as reported by the person separation reliability of 0.94 for the parents and of 0.87 for the children.

The final version of the ABILHAND-Kids questionnaire was therefore exclusively built on the parents' perceptions because of the higher discrimination of the three-level rating scale, the wider range of measurement, and the higher person separation reliability.

Metric Properties of ABILHAND-Kids

The calibration of the final 21-item ABILHAND-Kids scale is presented in Table 2. The items are sorted, from top to bottom, in order of decreasing difficulty (range: 3.00 to -3.23 logits). Higher logit values indicate more difficult activities. Table 2 also reports the standard error (SE) associated with each item difficulty (mean: 0.23 logits; range: 0.21 to 0.35 logits). The mean square fit statistics indicate that all 21 items contribute to the definition of a unidimensional measure of manual ability. Moreover, all RPM are positive (all values ≥ 0.55) indicating that each item is coherent with the rest of the questionnaire and contributes to the measurement of the manual ability.

Table 2. ABILHAND-Kids calibration for children with CP

Item	Difficulty (Logits)	SE (Logits)	INFIT (Mean Square)	OUTFIT (Mean Square)	RPM	Hands Involvement*
a. Buttoning up trousers	3.00	0.22	0.88	1.55	0.58	2 B
b. Buttoning up a shirt/sweater	2.67	0.22	0.77	0.65	0.58	2 A
c. Opening a jar of jam	1.82	0.22	1.09	0.97	0.55	2 B
d. Zipping-up a jacket	1.34	0.21	1.00	0.98	0.57	2 B
e. Rolling-up a sleeve of a sweater	1.12	0.21	1.14	1.42	0.57	2 A
f. Sharpening a pencil	0.98	0.22	1.05	0.98	0.58	2 C
g. Putting on a backpack/schoolbag	0.58	0.22	1.12	0.97	0.56	2 A
h. Zipping-up trousers	0.52	0.21	1.13	1.14	0.57	2 A
i. Fastening the snap of a jacket	0.33	0.21	1.13	1.03	0.57	2 A
j. Squeezing toothpaste onto a toothbrush	0.29	0.22	1.00	1.44	0.59	2 A
k. Unscrewing a bottle cap	0.08	0.22	1.07	1.33	0.57	2 B
l. Opening a bag of chips	-0.09	0.22	1.18	1.21	0.59	2 C
m. Opening the cap of a toothpaste tube	-0.41	0.23	0.80	0.62	0.61	2 A
n. Washing the upper-body	-0.61	0.22	1.13	1.06	0.60	1
o. Filling a glass with water	-0.62	0.23	1.08	1.17	0.58	2 A
p. Opening a bread box	-1.01	0.24	0.89	0.68	0.62	2 A
q. Taking off a T-shirt	-1.38	0.24	0.91	0.83	0.61	2 A
r. Putting on a hat	-1.38	0.26	0.83	0.70	0.63	2 A
s. Taking a coin out of a pocket	-1.63	0.26	0.66	0.45	0.64	1
t. Unwrapping a chocolate bar	-2.38	0.28	0.94	1.12	0.65	2 A
u. Switching on a bedside lamp	-3.23	0.35	0.84	0.51	0.70	1
Mean	0.00	0.23	0.98	0.99		
S.D.	1.52	0.03	0.14	0.30		

* 1 indicates unimanual activities; 2 indicates bimanual activities manageable in several unimanual steps (2A); requiring stabilization with one hand and digital activity with the other (2B); requiring digital activity from both hands (2C).

Description of ABILHAND-Kids

The definition and use of the ABILHAND-Kids scale is depicted in Figure 2. The top panel shows the distribution of manual ability measures of the children as perceived by the parents.

The manual ability measures of the children with CP are obtained by converting the ordinal total scores into linear measures. The bottom panel illustrates the ogival relationship between the finite total raw scores and the infinite manual ability measures. This relationship is approximately linear between total scores of 11 and 30. Outside this central range, however, a unitary progression in total score accounts for an increasing amount of manual ability measure. In the central range, the change in manual ability measure corresponding to a unitary increment in total score from 19 to 20 scores is equal to 0.17 logits. Outside this central range, it increases to 0.86 logits for the same increment in total score from 1 to 2. This fivefold difference denotes the non-linearity of the total score.

The middle panel shows the expected response to a given item as a function of the underlying manual ability measure. By comparing the ability of a given child to the difficulty of each item, it is possible to determine the expected score of the child to the item. According to the parents' perception, a child with an ability of 0 logits would be expected to perform without difficulty the two easiest activities, to perform with some difficulties the average activities, and to fail to perform the three most difficult activities. In summary, according to the distribution of subject measures, 52% of the children in our sample should successfully perform all the listed activities easily or with some difficulty. Twelve percent of the children should perform all activities easily and 4% should not be able to perform any of the 21 ABILHAND-Kids items. Therefore, the range of difficulties of the ABILHAND-Kids items fits the distribution of children's abilities.

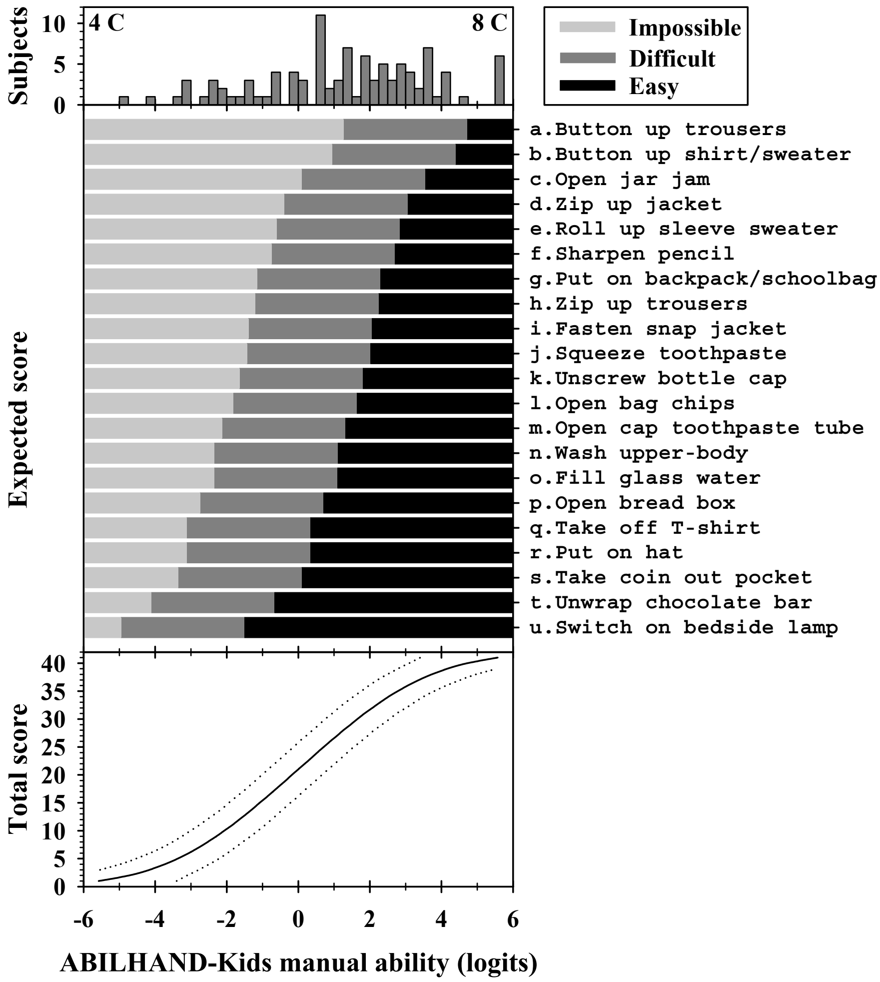

Figure 2. Top panel: distribution of manual ability measures of children with cerebral palsy according to their parents' perception. Twelve children with extreme scores cannot be measured by the ABILHAND-Kids scale because all activities were either impossible (4 C) or easy (8 C). Middle panel: a child's expected score to each item as a function of the underlying manual ability measure. A manual ability measure of zero is by convention set at the average item difficulty. Bottom panel: ogival relationship between total score and manual ability measure (solid line) and its 95% CI (dotted lines).

Scale Validation

The opinions of the four occupational therapists concerning the number of hands involved in each activity were consistent. The most frequently reported opinion is presented for each item in the last column in Table 2. Most of the ABILHAND-Kids items are bimanual activities (2A-2B-2C, 86%), most of which can be managed in several unimanual steps when using an adaptive strategy (2A, 67%). The activities requiring more bimanual involvement tend to be more difficult.

No significant differences in ABILHAND-Kids measures were observed across age, sex, or handedness. A significant difference in ABILHAND-Kids measures was observed as a function of school education ($t = 4.136$, $p < 0.001$), type of CP ($F = 9.621$, $p < 0.001$), and the GMFCS ($R = -0.640$, $p < 0.001$). A post hoc analysis of the indices having a significant effect on ABILHAND-Kids measures indicates that the children with CP who were placed in a mainstream school present a higher manual ability than the children following a special education program. Diplegic and hemiplegic/paretic children have a higher manual ability than tetraplegic/paretic children. Finally, a higher independence in gross motor function is associated with a higher manual ability.

The differential item functioning plot presented in Figure 3 compares the difficulty hierarchy of the items as perceived by the children's parents and by the experts. Most of the items lie within the 95% CI of the identity line indicating that the perceptions of the parents and the experts appear to be similar for the item hierarchy. There are four minor exceptions: "Zipping-up trousers" (h) and "Putting on a hat" (r) are estimated to be more difficult by the parents than by the experts, while "Unwrapping a chocolate bar" (t) and "Switching on a bedside lamp" (u) are estimated to be more difficult by the experts than by the parents.

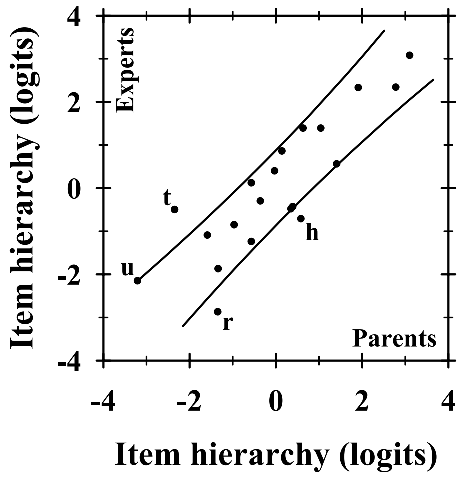

Figure 3. Differential item functioning plot of parents' and experts' perceived item difficulty hierarchy and 95% CI (solid lines) of the ideal invariance. Most difficult items are plotted in the top/right part of the figure. Items (dots) lying within the control lines have the same difficulty for both parents and experts groups. Four items lying outside the CI are identified by their labels.

Test-retest reliability

The test-retest reliability (delay: 25 ± 13 days) of the subject measures is presented in Figure 4. Children's measures perceived by the parents at the first and the second assessment are correlated ($R = 0.91$, $p < 0.001$). Most of the measures lie within the 95% CI of the identity line indicating that the parents tend to estimate consistently their child's ability over time. Moreover, the difficulty hierarchy of all 21 ABILHAND-Kids items is maintained between the first and the second assessment, indicating that the ABILHAND-Kids scale is invariant across time.

Discussion

We sought to develop a measure of manual ability in children with CP using the Rasch model. We also compared children's and parents' perceptions. The ABILHAND-Kids questionnaire was constructed from the parents' perception in order to cover a wider measurement range of manual ability than was possible using the children's perception. The 21 items retained for the final ABILHAND-Kids measure show an ordered rating scale, share the same discrimination, and fit a unidimensional scale.

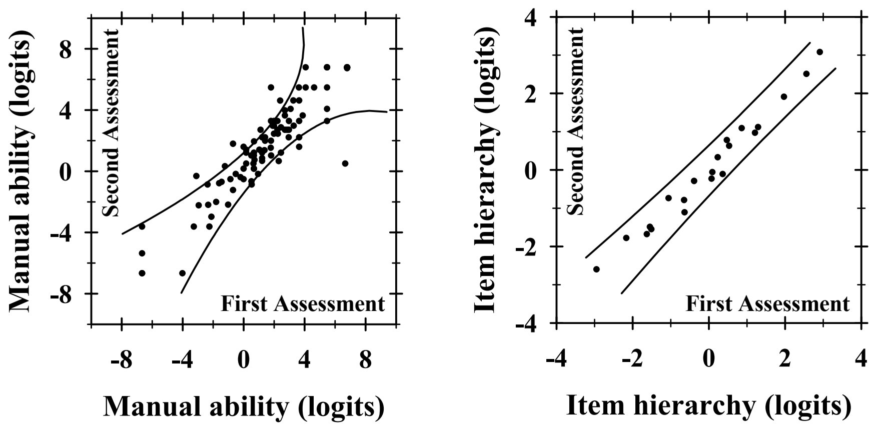

Figure 4. Left panel: relationship between the manual ability measures of the children as perceived by their parents at the first and the second assessment (delay: 25 ± 13 days) and 95% CI (solid lines) of the ideal invariance. More able children are plotted in the top/right part of the panel. Children measures (dots) lying within the control lines have the same estimated ability at the first and the second assessment. Right panel: differential item functioning plot of the item difficulty perceived by the children's parents across time and 95% CI (solid lines) of the ideal invariance. Most difficult items are plotted in the top/right part of the panel. All items (dots) are lying within the control lines indicating that they have the same estimated difficulty at the first and the second assessment.

The children's manual ability is better discriminated by the parents than by the children themselves. The activities are perceived by the children as either "impossible" or "easy" with very rare intermediate responses. The more dichotomous perception of the children is consistent with the Piagetian theory where young children typically engage in dichotomous thinking and may therefore focus only on the two extremes of Likert-type rating scales (Chambers & Johnston, 2002). The polychotomous parents' perception appears to be a more accurate source of information about manual ability than the dichotomous children's perception. However, the difference in discrimination between parents and children must be considered with caution, given the different modes of observation used in the two groups (Weinberger et al., 1996; Grootendorst et al., 1997). A face-to-face interview was used for the children and a written self-report for parents. A written self-report appears to be more appropriate for a clinical routine than a face-to-face interview where the interviewer is rarely the same. Moreover, face-to-face interviews may be influenced by the personality and the style of the interviewer, and the relationship with the subject (Verrips et al., 2001). The ABILHAND-Kids questionnaire was exclusively built on the parents' perception to reduce the error of measurement of manual ability as indicated by the higher person separation reliability observed in our sample (R = 0.94). In addition, the use of the parents' perception on behalf of the children's should allow, in the future, measurements of manual ability in all children with CP, including very young and severely impaired children and those with mental, psychological, emotional, attentional, or communicative disorders. The difficulty of just four items differs slightly between the parents and the experts. However, the differential item functioning of the four items is not high enough to compromise the clinical application of the scale. Parents might be better able than experts to judge which of the tasks are more difficult since they can observe their child's manual ability on a regular basis, capturing a sort of weighted average of the performance over long periods of time. Thus, two different modes of perception were used in the present study. Parents were asked to provide the observed difficulty of each activity for their child, while the experts were asked to provide the estimated difficulty of each activity for a "typical" CP child with moderate disorder. Nevertheless, further research will be needed to verify that parents' and experts' perceptions are really different for the four items, and to make any assumption about the reasons of these different points of view.

The 21 activities retained for ABILHAND-Kids involve both hands. Most of the unimanual activities included in the experimental version of the questionnaire were rarely perceived by the parents as "difficult", and consequently these activities were removed in the final version because the equal discrimination item selection criterion was not satisfied. This may be because the children either perform the unimanual activities easily using the less affected hand (usually the dominant hand) or cannot perform the activities because both hands are severely affected. In comparison, the activities requiring a higher bimanual involvement tend to be more difficult. The easiest bimanual activities performed every day by the children can often be managed in several unimanual steps using an adaptive strategy. The most difficult bimanual activities usually involve both hands, and sometimes involve bilateral digital activity. The perception of their difficulty might be complicated because alternative strategies might be adopted to complete these activities. Indeed, these activities might be performed with assisting devices, or by the parents instead of the child in order to prevent risk or save time (Sperle et al., 1997). Parents may also adapt their habits to facilitate some activities (e.g., by not overtightening a bottle). Nevertheless, all activities of the ABILHAND-Kids questionnaire retain the same discrimination and fit a unidimensional scale of manual ability, indicating that their difficulty was consistently perceived by the parents in our sample.

The 21 items of ABILHAND-Kids define a unidimensional measure of manual ability in children with CP and show a continuous progression in their difficulty. The standard errors associated with the item difficulties (mean: 0.23 logits) comply with the expectation for most variables (Linacre, 1994). Moreover, ABILHAND-Kids presents a good precision since the 21 items are well targeted on our sample expanding a wide range of functional states (R = 0.94). According to their parents, only 3.5% of the children with CP were not able to perform at least one item. All of these children were tetraplegic/paretic; they must be transported or use power mobility outdoors and in the community (GMFCS: Level IV) or were severely limited even with the use of assistive technology (GMFCS: Level V). Similarly, 7% of the children with CP were able to perform all items easily. All of these children were diplegic or hemiplegic/paretic and were at least able to walk indoors or outdoors on a level surface with assistive mobility devices (GMFCS: Levels I, II, III). The least measurable difference (Wright & Stone, 1979), which corresponds to the smallest difference in linear measure obtained by a unit increment in total score, is equal to 0.19 logits in the central range of the scale (range: -1.85 to 1.65 logits). This indicates that in the central range, the resolution of ABILHAND-Kids is sufficient to differentiate the ability of two subjects if one has 50% probability to succeed in performing a given item and the other 45%. The overall precision of the scale is summarized by a good person separation reliability in this sample (R = 0.94). The observed invariance in the item hierarchy after a delay of approximately 1 month indicates that the ABILHAND-Kids manual ability measures are reproducible over time. The metric properties of ABILHAND-Kids give to the scale the potential to measure any sensible change in manual ability induced, for example, by surgery, rehabilitation, biomedical treatment, or the use of assisting devices. However, the responsiveness and the predictiveness of ABILHAND-Kids need to be investigated further in a longitudinal study.

The analysis of the relationship between ABILHAND-Kids measures and demographic or clinical indices appears not only as a form of validation of the scale but also as a clinical end point. Although ABILHAND-Kids measures are not related to age, sex, or handedness, a significant relationship was found with school education, type of CP, and GMFCS. Children with more severe forms of CP are high-intensity users of physiotherapy services (Parkes et al., 2002) and are more likely to be placed in special schools which can better cope with treatment requirements. The significant relationship between ABILHAND-Kids measures and the type of CP confirms the previous reports (Azaula et al., 2000; Beckung & Hagberg, 2000 and 2002) that children with hemiplegia/paresis and diplegia are less disabled in their fine and gross motor functions than children with teraplegia/paresis. Finally, ABILHAND-Kids measures are significantly related to the levels of GMFCS; a higher manual ability relates to a higher gross motor function. A similar relationship between bimanual fine motor function and the levels of GMFCS was previously found (Beckung & Hagberg, 2002). However, in this study, the relationship to gross motor function is not perfect, indicating that fine and gross motor functions are two distinct but complementary variables. The relationship between ABILHAND-Kids measures and some demographic and clinical indices that relate to the severity of the pathology (i.e., school education, type of CP, GMFCS) is age-independent. This suggests that the questionnaire is sensitive to the pathological disruption of manual ability rather than to a maturation of manual ability, at least in CP children older than 6 years.

The Rasch model was used to construct and validate ABILHAND-Kids. It provides the calibration of the ABILHAND-Kids activities that can be sorted according to their estimated difficulty (Figure 2). The hierarchical nature of the scale identifies a child's spontaneous pattern of recovery given the current manual ability measurement. It can be used for goal setting in treatment planning. Furthermore, the

Rasch model has the ability to detect any discrepancies between the observed score to each item and the expected score, given the overall measure of the subject. More than a simple data quality control, it can be used to identify an idiosyncratic use of the questionnaire or diagnose behavioral peculiarities such as a misuse of adaptive strategies.

Finally, the Rasch model can be used to test the invariance of the manual ability variable defined by ABILHAND-Kids through differential item functioning tests (Wright & Stone, 1979). The current metric properties of ABILHAND-Kids make an encouraging starting point for further investigation of its invariance across demographic or clinical patient subgroups, and for its application across various pediatric diagnostic groups and cultures. If the invariance in the item hierarchy is also verified across treatment, then ABILHAND-Kids will also provide a responsive outcome measure to monitor patient status across time and recovery.

2.2. Invariance of ABILHAND-Kids across various demographic and clinical subgroups of children with CP

Abstract

The study was conducted to verify the invariance of ABILHAND-Kids through 19 demographic and clinical subgroups. The invariance was tested through differential item functioning tests which compare 1) the item difficulty hierarchy across two subgroups (e.g., boys vs. girls), and 2) the score observed at a given ability level in subjects from more than two subgroups (e.g., tetraplegics vs. diplegics. vs. right hemiplegics vs. left hemiplegics). Overall, the difficulty hierarchy of ABILHAND-Kids activities was invariant across various demographic and clinical subgroups of children with cerebral palsy, including age, sex, handedness, school education, type of cerebral palsy, gross motor function, manual ability, and hand functions. The invariance of ABILHAND-Kids across various demographic and clinical subgroups of children with cerebral palsy supports the clinical application of the scale.

This study is a supplement of the paper "ABILHAND-Kids: a measure of manual ability in children with cerebral palsy".

Introduction

ABILHAND-Kids is a manual ability scale developed in children with cerebral palsy (CP) using the Rasch model (Arnould et al., 2004). This measurement scale has been demonstrated to be valid, reliable in a sample of 113 CP children (R = 0.94), and reproducible over an average delay of 25 ± 13 days (R = 0.91). However, once satisfactory metric properties have been achieved, it is necessary to verify the invariance of the scale across different subgroups of CP children. Essentially, ABILHAND-Kids should work in the same way for boys and girls, right-handers and left-handers, etc. Provided that the data fit the requirements of the Rasch model, the score observed to an item solely depends on the manual ability of the CP child and the difficulty of the item (Rasch, 1960). Therefore, the scores should not be influenced by other demographic (e.g., age, sex, school education) or clinical (e.g., type of CP, gross motor function, hand functions) factors (Smith, 1992). Hence, children having the same manual ability are supposed to obtain the same score to any item, regardless of the other demographic and clinical factors. If this is not the case, the item is biased or, according to a more recent terminology, the item presents a differential functioning (Holland & Wainer, 1993).

Differential item functioning: uniform and non-uniform

The differential item functioning (DIF) approach is based on the item characteristic curves which express the score expected by the model to any given item as a function of children's manual ability. The item characteristic curves of two hypothetical items are presented in Figure 1 (Panels A1 and B1). The mean scores observed in four children's class intervals (CI) of increasing ability are also represented. Overall, the observed scores of both items fit the requirements of the Rasch model since the mean scores observed in each CI follow the evolution of the score expected by the model. If the children's responses are divided into two or more subgroups (e.g., boys vs. girls, tetraplegics vs. diplegics. vs. right hemiplegics vs. left hemiplegics), the mean scores observed in different CIs may also be represented for each subgroup. In Figure 1 (Panels A2 and B2), the mean scores observed in each subgroup (i.e., boys vs. girls) follow a different evolution as a function of children's manual ability, as if the scores of the two subgroups fitted two different items. Two types of DIF can be identified: the uniform and the non-uniform DIF. Uniform DIF is observed when the observed mean scores evolve in parallel in both subgroups (Figure 1, Panel A2). Given the manual ability of a CP child, the observed score for item A is systematically higher if the child is a girl rather than a boy and this systematic sex difference does not vary from one CI to another. In this case, the item A involves a constant higher challenge for boys than for girls as compared with the other items of the questionnaire.

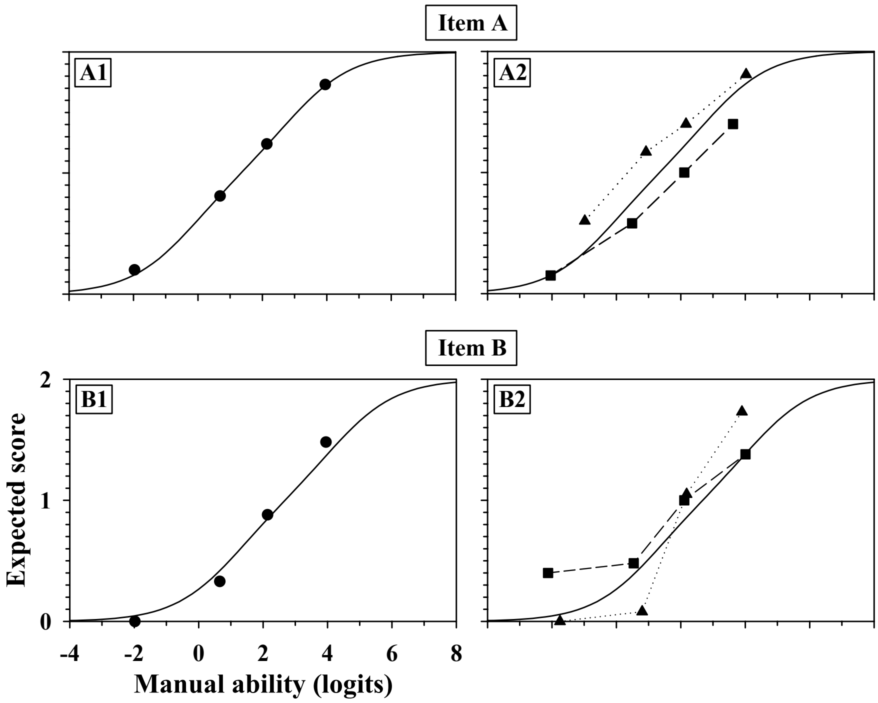

Figure 1. Item characteristic curves (solid line) of two hypothetical items A and B and mean observed scores (dots) of the entire sample for 4 class intervals (Panels A1 and B1). The mean scores observed in the entire sample fit the prescriptions of the Rasch model. When the mean observed scores are presented separately for the boys (squares, dashed line) and for the girls (triangles, dotted line), a differential item functioning appears (Panels A2 and B2). The item A presents a uniform DIF since the mean observed scores evolve in parallel in both subgroups (Panel A2). The item B presents a non-uniform DIF since the sex difference observed in mean scores varies from one class interval to another with manual ability (Panel B2).

Non-uniform DIF is observed when the observed mean scores do not evolve in parallel in both subgroups (Figure 1, Panel B2). Hence, the sex factor interacts with the manual ability level and this interaction depends on the child's location. Note that in the non-uniform DIF, the relative difficulty of the item is not necessarily different between the subgroups but the data observed in the different subgroups do not fit the requirements of the Rasch model (Smith, 1992).

Methods of detecting differential item functioning

In practice, two methods are mainly used to statistically test the extent of a differential item functioning.

The first method consists in computing a two-way analysis of variance (ANOVA) on the standardized residuals of the different CIs (Andrich et al., 2004). The residual of a particular CI is computed as the difference between the mean score observed in the CI (O_{CI}) and the mean score expected by the model (E_{CI}) considering the item difficulty and the mean manual ability of the children who are part of the CI. The standardized version of the residual is obtained by dividing the residual of the particular CI by the standard deviation of the mean expected score of the CI ($SD_{E(CI)}$):

$$\text{Standardized residual}_{CI} = \frac{O_{CI} - E_{CI}}{SD_{E(CI)}} \qquad \text{(Eq.3.1)}$$

The first factor of the two-way ANOVA is the person group of interest (e.g., sex) and the second factor is the CIs of increasing manual ability levels. This method detects uniform DIF if the standardized residuals change significantly from one subgroup to the other(s) as well as non-uniform DIF if the standardized residuals present a significant interaction between the subgroups and the CIs.

The second method consists in computing a t-test on the difference between the difficulties of each item estimated separately for two subgroups. This methods is based on the necessity to verify the requirements of an objective measurement (Smith, 1992). Indeed, the observed scores must fit the prescriptions of the Rasch model before they can be converted into linear measures. Consequently, the DIF of an item is merely envisioned as a difference in its relative difficulty between the subgroups. Independent analyses are performed for each subgroup and the difficulty of each item is estimated as well as the associated standard error. Given the pairs of item difficulties and the associated standard errors, a t-statistic can be computed:

$$t_i = \frac{\delta_{i1} - \delta_{i2}}{SE_{i1}^2 + SE_{i2}^2} \qquad \text{(Eq.3.2)}$$

where δ_{i1} is the difficulty of the item i estimated from the analysis of subgroup 1, δ_{i2} is the difficulty of the item i estimated from the analysis of subgroup 2, SE_{i1} is the standard error associated to the estimate of δ_{i1}, and SE_{i2} is the standard error associated to the estimate of δ_{i2}. This procedure is identical to the graphical method reported by Wright & Stone (1979) where the relative item difficulties of two subgroups are contrasted in a x-y plot according to a 95% confidence interval. The separate calibration t-test method presents therefore the advantage of providing, in just one graph, a very clear graphical interpretation of the uniform DIF of an item set, and thus was preferred in this study to the two-way ANOVA method. However, it fails to directly identify non-uniform DIF and, accordingly, requires the verification that data of each subgroup fit the prescriptions of the Rasch model (Smith, 1992). The two-way ANOVA method may also be used to verify the absence of non-uniform DIF before applying the separate calibration t-test method. Note that the t-test method can only be applied for the comparison of two subgroups. If there are more than two subgroups of interest (e.g., tetraplegics vs. diplegics. vs. right hemiplegics vs. left hemiplegics), two or more comparisons must be made. Multiple comparisons for a single item raise questions about the appropriateness of the Type 1 error rate (Smith, 1992). However, when there are more than two subgroups of interest, the whole sample can be split dichotomously by regrouping subjects into only two subgroups provided that the grouping makes sense. For instance, for the gross motor function criterion, it made sense to group the CP children with the lowest gross motor function levels together (GMFCS: Levels 2 to 5) and to compare the item calibrations estimated for this group with the one estimated for the less impaired children (GMFCS: Level 1). On the other hand, , for the type of CP criterion, it did not make sense to group diplegic children with either tetraplegic or hemiplegic children as each type of CP is characterized by specific clinical appearances.

Invariance of ABILHAND-Kids across various subgroups of children with CP

The invariance of ABILHAND-Kids was investigated across various demographic and clinical subgroups of CP children formed on the basis of the following criteria: (1) age (< 10 vs. ≥ 10 years old); (2) sex (girls vs. boys); (3) handedness (right-handers vs. left-handers); (4) school education (special vs. mainstream); (5) type of CP (quadriplegics vs. diplegics vs. right hemiplegics vs. left hemiplegics); (6) gross motor function (GMFCS < Level I vs. GMFCS = Level I); (7) manual ability (less vs. more able, split on the median manual ability measure); (8-19) hand functions on both the dominant hand (DH) and the non-dominant hand (NDH) (less vs. more hand function, split on the median score). Hand functions included grip strength (GS), gross manual dexterity (GMD), fine finger dexterity (FFD), tactile pressure detection (TPD), stereognosis (S), and proprioception (P). The assessment of these hand functions is described in Chapter 3.

Differential item functioning of the ABILHAND-Kids items was tested using the graphical method of Wright & Stone (1979) for all demographic and clinical criteria except the type of CP which included more than two subgroups of CP children. Instead, the two-way ANOVA method was used to test the invariance of ABILHAND-Kids across the four types of CP (i.e., tetraplegics vs. diplegics. vs. right hemiplegics vs. left hemiplegics). The results of the ANOVAs are shown in Table 1. No ABILHAND-Kids activities were found to have non-uniform DIF and only the item "s. Taking a coin out of pocket" was found to be significantly more difficult for tetraplegic children than for other types of CP.

Table 1. Differential Item Functioning across types of CP using the two-way ANOVA				
Item	Uniform (df = 3)		Non-uniform (df = 9)	
	F	p-value	F	p-value
a. Buttoning up trousers	0.06	0.98	0.54	0.84
b. Buttoning up a shirt/sweater	2.04	0.12	0.20	0.99
c. Opening a jar of jam	0.23	0.88	1.78	0.09
d. Zipping-up a jacket	1.56	0.21	0.86	0.56
e. Rolling-up a sleeve of a sweater	1.22	0.31	0.38	0.94
f. Sharpening a pencil	1.44	0.24	1.52	0.16
g. Putting on a backpack/schoolbag	2.59	0.06	0.77	0.65
h. Zipping-up trousers	0.61	0.61	0.49	0.88
i. Fastening the snap of a jacket	1.08	0.36	1.77	0.09
j. Squeezing toothpaste onto a toothbrush	1.53	0.22	0.10	1.00
k. Unscrewing a bottle cap	0.22	0.88	1.71	0.10
l. Opening a bag of chips	2.22	0.09	1.01	0.44
m. Opening the cap of a toothpaste tube	0.14	0.94	0.76	0.65
n. Washing the upper-body	0.66	0.58	1.20	0.31
o. Filling a glass with water	1.85	0.15	1.48	0.17
p. Opening a bread box	0.30	0.82	0.88	0.54
q. Taking off a T-shirt	2.60	0.06	0.14	1.00
r. Putting on a hat	0.15	0.93	0.61	0.79
s. Taking a coin out of a pocket	2.84	0.04*	0.61	0.79
t. Unwrapping a chocolate bar	1.89	0.14	0.39	0.94
u. Switching on a bedside lamp	0.75	0.53	0.69	0.72

* Significant DIF ($p < 0.05$)

For all other criteria the sample was split in two subgroups and the item difficulty hierarchy was estimated separately for both subgroups after potential non-uniform DIF was ruled out by verifying the fit to the model prescriptions in both subgroups[5]. The item difficulty hierarchy obtained in each subgroup was subsequently centred on the average item difficulty in either analysis and contrasted in a x-y plot according to the 95% confidence interval. Figure 2 presents the DIF plots for the 18 criteria dividing the sample into two subgroups. For each panel of the Figure 2, the dots represent the difficulty of the items estimated in abscissa for the CP children of one subgroup (e.g., children younger than 10 years old for the age criterion) and in ordinate for the CP children of the other subgroup (e.g., children aged 10 or more for the age criterion). More difficult items are plotted on the top/right part of the panels. The two curves represent the 95% confidence interval of the ideal invariance. The

[5] Similar results were obtained using the two-way ANOVA method and were therefore not reported here.

items lying within the 95% confidence interval have the same relative difficulty between the two subgroups. The items lying outside the 95% confidence interval have a relative difficulty significantly different from one subgroup to the other and are identified by their labels. Overall, most of the items lie within the 95% confidence interval indicating that the difficulty hierarchy of the ABILHAND-Kids activities is invariant across various demographic and clinical subgroups. There are few exceptions; for instance, "k. Unscrewing a bottle cap" appears to be more difficult for children aged 10 or more than for younger children, while "o. Filling a glass with water" appears to be more difficult for girls than for boys. Some adjustments can be made to allow such items displaying DIF to vary between the subgroups (Tennant et al., 2004). For instance, the item "s. Taking a coin out of pocket" which displays a uniform DIF according to the two-way ANOVA method may be split into two items, with one item calibrated for the tetraplegics and one item calibrated for the children with other CP types. Such adjustments were not implemented up until now as the DIF of the ABILHAND-Kids items observed in our sample seems not high enough to compromise the clinical application of the questionnaire.

Conclusion

The present study has shown that, overall, the invariance of ABILHAND-Kids is verified across various demographic and clinical subgroups of children with CP. The ABILHAND-Kids questionnaire can therefore be used to measure manual ability in children with CP whatever their age, sex, handedness, school education, type of CP, gross motor function, manual ability, and hand functions.

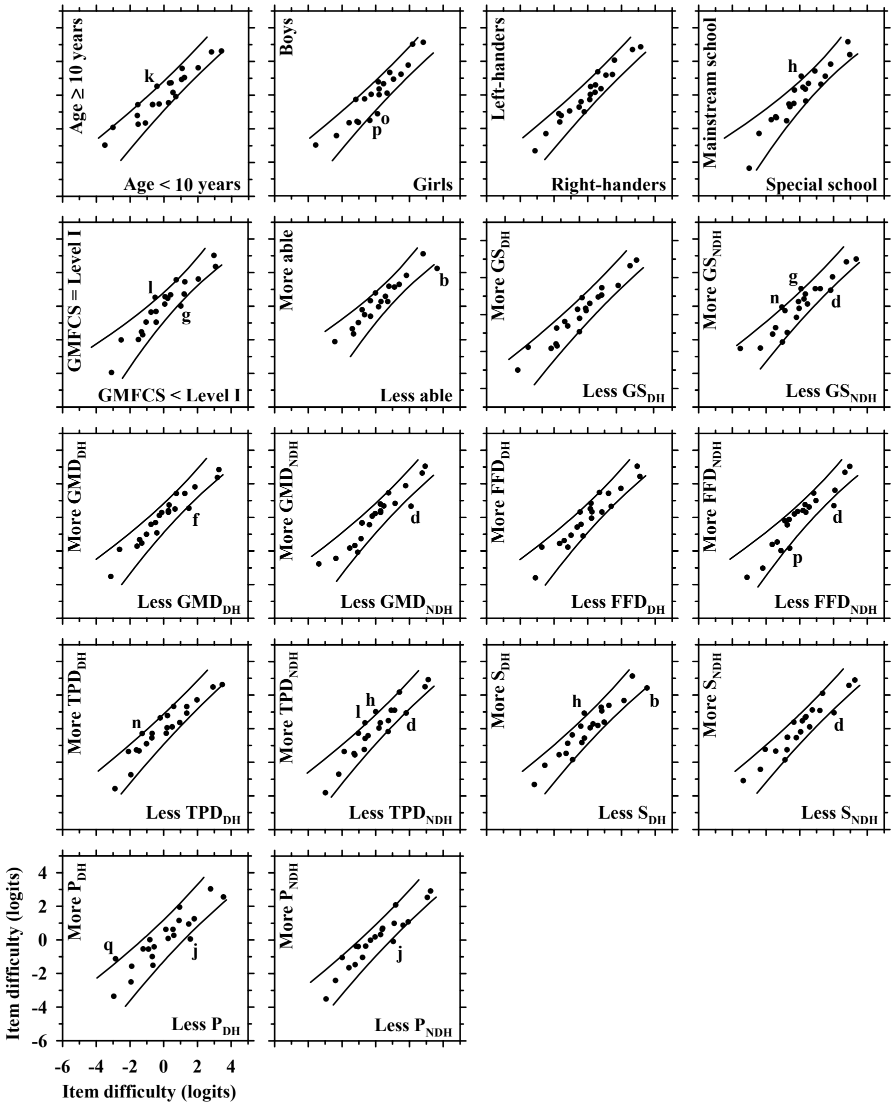

Figure 2. Differential item functioning plots of the 21 ABILHAND-Kids items across 18 criteria dividing the sample into two subgroups and the 95% confidence interval (solid lines) of the ideal invariance. More difficult items are plotted on the top/right part of the panels. Items (dots) lying within the 95% confidence interval have the same difficulty in both subgroups. Items lying outside the confidence interval are identified by their labels. The following abbreviations are used for the different criteria: DH: dominant hand; NDH: non-dominant hand; GMFCS: gross motor function classification system; GS: grip strength; GMD: gross manual dexterity; FFD: fine finger dexterity; TPD: tactile pressure detection; S: stereognosis; and P: proprioception.

Chapter 3
Hand impairments and their relationship with manual ability in children with cerebral palsy

Abstract

Objective: To study hand impairments and their relationship with manual ability in children with cerebral palsy (CP).

Methods: One hundred and one children with CP (58% boys; mean age, 10 years) were administered the Jamar dynamometer test, the Box and Block gross manual dexterity test, the Purdue Pegboard fine finger dexterity test, the Semmes-Weinstein tactile pressure detection test, the Manual Form Perception Test, and a proprioception test. Manual ability was also measured with the ABILHAND-Kids questionnaire. The relationship between hand impairments and manual ability was studied through a correlation matrix and a multiple linear regression analysis.

Results: On both hands, manual ability was significantly but moderately correlated with grip strength, gross manual dexterity, fine finger dexterity and stereognosis. Tactile pressure detection and proprioception were not related to manual ability. Multiple linear regression indicated that gross manual dexterity on the dominant hand and grip strength on the non-dominant hand were the best independent predictors of the CP children's manual ability. However, this combination of hand functions predicted only 58% of the variance in manual ability measures.

Conclusion: Manual ability cannot simply be inferred from hand impairments as it depends on complex interactions between functional, personal, and environmental factors. Consequently, manual ability should be measured per se.

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Introduction

Cerebral palsy (CP) is the most prevalent form of motor disability in children (Rosenbaum, 2003). Hand and upper limb dysfunctions constitute the main problems in about half the children with CP (Uvebrant, 1988) and are generally thought to be largely responsible for the difficulty experienced in daily activities (Fedrizzi et al., 2003). Based on this assumption, physiotherapy and medical treatments traditionally endeavour to reduce hand impairments (Koman et al., 2004). However, the relationship between hand impairments and a child's ability to execute manual activities is not straightforward and has not really been investigated empirically in children with CP (Ostensjo et al., 2003).

The International Classification of Functioning, Disability and Health (ICF) (World Health Organization, 2001) provides a conceptual framework for formalizing the impact of health condition on an individual's functioning. The ICF defines three dimensions of functioning: body functions and structures (body dimension), activity (individual dimension), and participation (social dimension). Problems in either dimension are respectively designated as impairments, activity limitations and participation restrictions. Impairments refer to anomaly, defect, loss or other significant deviation in body functions and structures (e.g., reduction in grip strength); activity limitations occur when the person has some difficulties in executing daily activities (e.g., dressing); and participation restrictions are the problems an individual may experience in the involvement in life situations (e.g., participation in school life). The impact of a pathology such as CP on an individual's health results from interactions between the three dimensions, that may also be influenced by personal (e.g., age, lifestyle, motivation) and environmental (e.g., architectural characteristics, legal system) contextual factors. Nevertheless, the nature and the strength of these interactions are not documented in the ICF and remain a question open to empirical testing.

Manual ability refers to the "capacity to manage daily activities requiring the use of hands and upper limbs, whatever the strategies involved" (Penta et al., 2001) and therefore relates to the activity dimension. While most hand impairments can be measured with physical units (e.g., grip strength can be measured in Newtons), manual ability is a behaviour concealed within a person or a child and cannot be directly measured (Penta et al., 2001). Nevertheless, such a behaviour can be inferred from a child's performance in manual activities as determined by questionnaires. In a previous study (Arnould et al., 2004), the ABILHAND-Kids questionnaire was developed as a measure of manual ability in children with CP. The questionnaire assesses the perception of a child's difficulty in performing manual activities and the score is transformed into a unidimensional, linear measure of manual ability via the Rasch measurement model (Rasch, 1960).

The objectives of this study were to measure the hand impairments of children with CP and investigate the relationship between hand impairments and manual ability as measured with the ABILHAND-Kids questionnaire.

Methods

Subjects

The study was authorized by the ethics committee of the Université catholique de Louvain, Faculty of Medicine in Brussels, Belgium. The definition adopted for selecting children with CP was "all non-progressive but often changing motor impairment syndromes secondary to lesions or anomalies of the brain arising in the early stages of its development" (Mutch, 1992). One hundred and one children with CP (59 boys, mean age ± SD: 10 ± 2 years; 42 girls, mean age ± SD: 10 ± 3 years) were recruited through seven centres specializing in CP and were assessed by the same examiner. They were older than 6 years old to make sure that their manipulative skills in activities of daily living were mature (Illingworth, 1975) and no children had a major intellectual deficit (IQ ≥ 60). The sample description is provided in Table 1.

Table 1. Sample description (n = 101)

Age* (years)	10 (6-15)
Gender	
Boy	59
Girl	42
Handedness	
Right	51
Left	49
Ambidextrous	1
School education	
Mainstream	41
Type 1: mild mental retardation	1
Type 4: physical handicap	52
Type 8: learning disabilities	6
Home	1
Type of CP	
Tetraplegia/paresis	31
Diplegia	20
Hemiplegia/paresis	
Right	25
Left	25

* Mean (range)

Hand impairment assessment

The children were tested individually in a quiet room and were instructed how to perform each test. Three motor and three sensory functions were assessed on both hands, starting with the dominant hand. Handedness was determined by writing hand preference and actually corresponds to the less affected hand in all children tested.

The motor functions included the grip strength, gross manual dexterity, and fine finger dexterity. Grip strength was measured with a Jamar dynamometer (Therapeutic Equipment Corporation, Clifton, New Jersey, USA) according to the procedure described by Mathiowetz et al. (1986b). The grip strength was determined as the average of the maximal force exerted on the dynamometer across three trials. Gross manual dexterity was measured with the Box and Block Test (Cromwell, 1960) according to the procedure of Mathiowetz et al. (1985). The gross manual dexterity was determined as the maximum number of blocks transported individually from one compartment of a box to the other within one minute. Fine finger dexterity was measured with the Purdue Pegboard Test (Lafayette Instrument Model 32020, Sagamore, USA) (Tiffin & Asher, 1948) according to the procedure described by Mathiowetz et al. (1986a). The fine finger dexterity was determined on three trials as the average number of pegs picked up from a cup and placed into the holes of a board within 30s.

The sensory functions included tactile pressure detection, stereognosis, and proprioception. Tactile pressure detection was measured at the tip of the index finger with a Semmes-Weinstein aesthesiometer (Lafayette Instrument Company, Loughborough, UK) (Bell-Krotoski, 1990) according to the procedure described by Bell-Krotoski (1990). The tactile pressure detection threshold was determined as the force required to bend the thinnest filament the blindfolded children could feel. Stereognosis was measured with the Manual Form Perception Test as modified by Cooper et al. (1995). The stereognosis was determined as the number of objects out of 10 correctly identified by touch by the blindfolded children. Proprioception was measured by passively moving the metacarpophalangeal joints of the thumb and the index finger according to the procedure of Cooper et al. (1995). The proprioception was determined as the number of joint movement directions the blindfolded children correctly identified out of ten trials (five for the thumb and five for the index finger).

Manual ability assessment

Manual ability was measured with the ABILHAND-Kids questionnaire (Arnould et al., 2004). This questionnaire measures the child's "capacity to manage daily activities requiring the use of hands and upper limbs, whatever the strategies involved" (Penta et al., 2001). Twenty-one mostly bimanual activities were rated by the children's parents on a three level-scale (0: impossible, 1: difficult, or 2: easy) by providing their child's perceived difficulty in performing each activity. The parents were instructed to give their responses when the activities were done without any technical or human help, irrespective of the hand(s) actually used to do the activity, and whatever the child's compensatory strategies. Activities not attempted in the last three months were not scored and were encoded as missing responses. The ordinal total score obtained on the ABILHAND-Kids questionnaire was subsequently transformed into an interval-level measure of manual ability according to the Rasch model (Rasch, 1960). The manual ability measures are expressed in logits, a constant measurement unit representing the probability of the successful achievement of manual activities, the origin being conventionally fixed at the average item difficulty. Manual ability measures of CP children were already reported in a previous study (Arnould et al., 2004) and are therefore not described here.

Statistical analysis

Descriptive statistics were used to determine the extent of hand impairments and manual ability in our sample of children with CP. The scores of grip strength, gross manual dexterity, and fine finger dexterity were converted into standardized scores (z-scores) according to normative data available in the literature (Mathiowetz et al., 1986a and 1986b) and norms established in our laboratory (for gross and fine manual dexterity). This procedure determines the extent to which a CP child deviates from normal given his or her age, gender, and handedness and allows all scores to be expressed on a common z-score scale. Grip strength, gross manual dexterity, and fine finger dexterity were considered as significantly impaired when the z-score was lower than -2. The scores of tactile pressure detection, stereognosis, and proprioception were not z-transformed since the normative data were not normally distributed despite various attempts of data normalization. The scores were therefore compared with those of age- and sex-matched healthy children using a Wilcoxon Signed Rank test. Tactile pressure detection, stereognosis, and proprioception were considered as significantly impaired when the score was worse than the fifth percentile of the distribution observed for the age- and sex-matched healthy children (i.e., 166mg for tactile pressure detection; 9 objects correctly identified for stereognosis; 7 joint movement directions correctly identified for proprioception).

The strength of the linear association between each variable was determined according to a Spearman's or Pearson's correlation coefficient, depending on the nature of the data.

A multiple linear regression was subsequently performed to identify which combination of hand impairments might best predict variations in manual ability measures. All hand impairments that were significantly related to manual ability in correlation analyses were included in a forward stepwise regression procedure to select the best independent predictors of manual ability. The adjusted coefficient of determination which considers the number of selected variables was used to avoid the overestimation of the real predictive capacity of the regression equation. All assumptions underlying the multiple linear regression analysis were verified, namely the linearity of the regression function, the normality and the constant variance of the errors, the absence of influential outliers, and the absence of multicollinearity. The alpha level of significance was fixed at 0.001 for all statistical tests to minimize type 1 errors.

Results

Hand impairments

The z-transformed score distributions of grip strength (GS), gross manual dexterity (GMD), and fine finger dexterity (FFD) computed on both hands in children with CP are presented in Figure 1. All scores on the dominant hand (DH) were significantly higher (Paired t-test or Wilcoxon Signed Rank test, $p < 0.001$) than on the non-dominant hand (NDH) indicating a systematically better performance on the DH. Among the motor hand functions, grip strength was the least impaired function and fine finger dexterity the most impaired function.

The median and the interquartile range (IQR) of tactile pressure detection, stereognosis, and proprioception are presented in Table 2. Although no significant difference was observed in CP children between the tactile pressure detection of both hands, stereognosis and proprioception were significantly higher in the DH than in the NDH confirming the better performance of the DH (Wilcoxon Signed Rank test, $p < 0.001$). For both hands, tactile pressure detection and stereognosis were significantly impaired in CP children as compared with age- and sex- matched healthy children ($p < 0.001$). Proprioception was not significantly impaired on the DH and just slightly impaired on the NDH ($p = 0.03$).

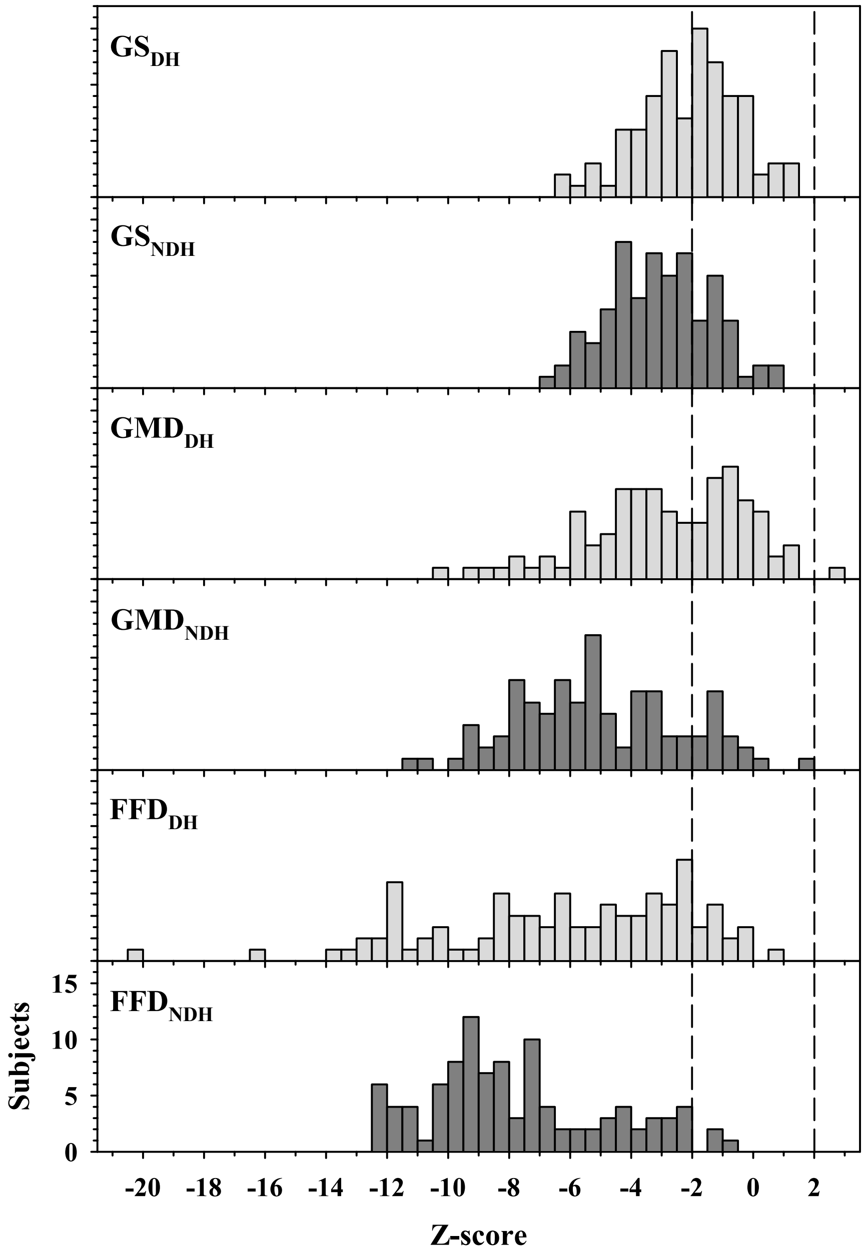

Figure 1. Z-score distributions of grip strength (GS), gross manual dexterity (GMD), and fine finger dexterity (FFD) computed in children with CP for both the dominant hand (DH, in pale grey) and the non-dominant hand (NDH, in dark grey). A z-score range between -2 and 2 was considered as not significantly different from normal.

Table 2. Tactile pressure detection, stereognosis, and proprioception impairments

	CP_{DH} Median (IQR)	CP_{NDH} Median (IQR)	H_{DH} Median (IQR)	H_{NDH} Median (IQR)	*P* value (CP_{DH}/H_{DH})	*P* value (CP_{NDH}/H_{NDH})
TPD (mg)	67.7 (27.5 - 166)	67.7 (27.5 - 408.2)	67.7 (27.5 - 67.7)	67.7 (27.5 - 166)	<0.001	<0.001
S (n)	10 (9 - 10)	9 (6.75 - 10)	10 (10 - 10)	10 (10 - 10)	<0.001	<0.001
P (n)	10 (10 - 10)	10 (9 - 10)	10 (10 - 10)	10 (10 - 10)	0.944	0.025

CP = children with cerebral palsy; H = age- and sex- matched healthy children; DH = dominant hand; NDH = non-dominant hand; IQR = interquartile range; TPD = tactile pressure detection; S = stereognosis; P = proprioception.

The prevalence of the six hand impairments is presented in Table 3 for each type of CP (tetraplegia: n = 31; diplegia: n = 20; hemiplegia: n = 50). On both hands, the prevalence of hand impairments in the total sample indicated that hand functions were from the most to the least impaired: fine finger dexterity, gross manual dexterity, grip strength, tactile pressure detection or stereognosis, and finally proprioception. The prevalence of impairments in gross manual dexterity was slightly higher than in grip strength as almost all children with tetraplegia had a significantly impaired gross manual dexterity. Apart from that, the same pattern of hand impairments was observed in the different types of CP. Overall, children with CP displayed more motor impairments than sensory impairments. Interestingly, more than 30% of the hemiparetic children presented an impairment of both grip strength and gross manual dexterity on the non-paretic DH. Moreover, 76% of these children presented an impairment of the fine finger dexterity on the same hand.

Table 3. Prevalence of hand impairments according to CP types

Impairments	Total sample (%) n = 101	Tetraplegia (%) n = 31	Diplegia (%) n = 20	Hemiplegia (%) n = 50
DH				
Fine finger dexterity	86	100	90	76
Gross manual dexterity	57	97	50	36
Grip strength	47	61	55	34
Tactile pressure detection	21	32	15	16
Stereognosis	20	39	10	12
Proprioception	4	6	0	4
NDH				
Fine finger dexterity	97	100	90	98
Gross manual dexterity	83	100	65	80
Grip strength	73	77	50	80
Stereognosis	38	42	15	44
Tactile pressure detection	33	32	20	38
Proprioception	15	13	0	20

Relationships among hand impairments

The correlation coefficients between each of the six hand impairments measured in children with CP are presented in Table 4. For both hands, gross manual dexterity and fine finger dexterity were the most related hand impairments. On the DH, grip strength, gross manual dexterity, fine finger dexterity, and stereognosis had a significant but moderate relationship with each other; tactile pressure detection and proprioception showed no significant association with any other hand impairments. Overall, a similar pattern of associations was observed on the NDH with, however, a higher correlation of stereognosis and proprioception with other hand impairments.

Table 4. Correlation matrices

Dominant hand							
	MA	**GS**	**GMD**	**FFD**	**TPD**	**S**	**P**
Manual ability (MA)	1.00						
Grip strength (GS)	0.46*	1.00					
Gross manual dexterity (GMD)	0.67*	0.48*	1.00				
Fine finger dexterity (FFD)	0.57*	0.47*	0.84*	1.00			
Tactile pressure detection (TPD)	0.11	-0.02	-0.11	-0.20	1.00		
Stereognosis (S)	0.49*	0.40*	0.43*	0.50*	-0.11	1.00	
Proprioception (P)	0.15	0.15	0.16	0.12	-0.14	0.32	1.00

Non-dominant hand							
	MA	**GS**	**GMD**	**FFD**	**TPD**	**S**	**P**
Manual ability (MA)	1.00						
Grip strength (GS)	0.56*	1.00					
Gross manual dexterity (GMD)	0.66*	0.63*	1.00				
Fine finger dexterity (FFD)	0.45*	0.43*	0.79*	1.00			
Tactile pressure detection (TPD)	-0.13	-0.23	-0.30	-0.22	1.00		
Stereognosis (S)	0.48*	0.60*	0.60*	0.49*	-0.44*	1.00	
Proprioception (P)	0.26	0.38*	0.33*	0.30	-0.49*	0.55*	1.00

* Significant correlations ($p < 0.001$)

Relationship between hand impairments and manual ability

Table 4 also shows the correlation coefficients between hand impairments and manual ability. On both hands, manual ability was significantly but moderately correlated with grip strength, gross manual dexterity, fine finger dexterity, and stereognosis. No significant relationship was found between manual ability and tactile pressure detection nor proprioception on either hand although tactile pressure detection was impaired on both hands. Gross manual dexterity presented the highest correlation with manual ability on both hands, immediately followed by fine finger dexterity on the DH and grip strength on the NDH.

Grip strength, gross manual dexterity, fine finger dexterity, and stereognosis of both hands were entered into the forward stepwise regression procedure. The results of the multiple linear regression are presented in Table 5. The gross manual dexterity on the DH was the strongest predictor of manual ability accounting for 44% of the variance in manual ability measures. The grip strength on the NDH was the second best independent predictor of manual ability but accounted for only a further 14% of the variance. The addition of other hand functions hardly improved the prediction of manual ability measures (less than 5%). The regression equation obtained by the forward stepwise method was therefore the following: manual ability = 4.42 + 0.51 (gross manual dexterity on DH) + 0.56 (grip strength on NDH). The combination of gross manual dexterity on the DH and grip strength on the NDH predicted altogether 58% of the variance in manual ability measures.

Table 5. Multiple linear regression (forward stepwise method)

Hand impairments selected in the model	R	R^2	$R^2_{adjusted}$	Delta $R^2_{adjusted}$
GMD_{DH}	0.67	0.45	0.44	0.44
GMD_{DH} + GS_{NDH}	0.77	0.59	0.58	0.14

GMD_{DH} = gross manual dexterity on the dominant hand; GS_{NDH} = grip strength on the non-dominant hand.

The combined influence of gross manual dexterity of the DH and grip strength of the NDH on manual ability is illustrated in Figure 2. In our sample, children without impairment in these functions had the highest manual ability measures. Children with one significant impairment, either in gross manual dexterity on the DH (n = 9) or in grip strength on the NDH (n = 25), presented a slightly lower manual ability. The 49 children significantly impaired in both gross manual dexterity on the DH and grip strength on the NDH had the lowest manual ability measures, with a median measure of 0 logit equivalent to the average item difficulty of the ABILHAND-Kids questionnaire. A Kruskal-Wallis test also confirmed that cumulative impairments were significantly related ($p < 0.001$) to a decrease in manual ability.

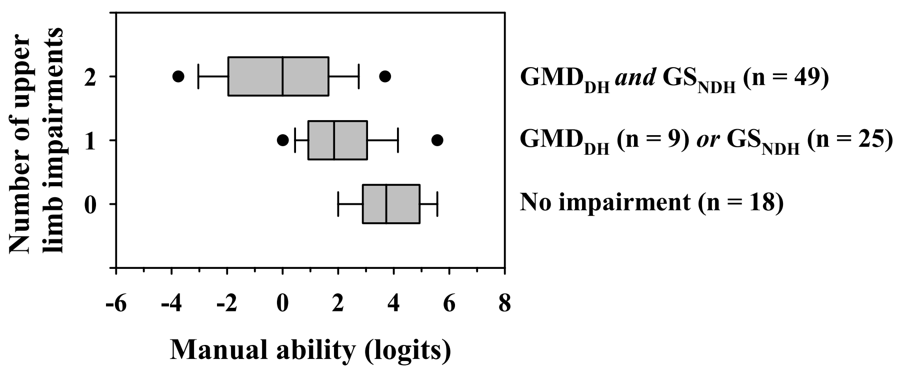

Figure 2. Box plots showing the manual ability measure distributions of children with CP according to the presence of a significant impairment (z-score lower than -2) of gross manual dexterity on the dominant hand (GMD_{DH}) and grip strength on the non-dominant hand (GS_{NDH}). Solid dots indicate the 5% and 95% outliers; vertical bars outside the box indicate the 10% and 90% limits; box indicates the 25% and 75% limits (i.e., the interquartile range); and the vertical line inside the box indicates the median of the distributions.

Discussion

Hand impairments and their relationship with manual ability in children with CP were investigated in the present study. Motor functions, especially fine finger dexterity, were more frequently impaired than sensory functions. Grip strength, gross manual dexterity, fine finger dexterity, and stereognosis of both hands were significantly related to manual ability. Alternatively, tactile pressure detection and proprioception did not play a significant role in the capacity of CP children to perform manual activities. Gross manual dexterity on the DH and grip strength on the NDH was the combination of hand impairments that best predicted the limitations perceived in manual ability, although the limitations cannot result from just hand impairments.

Hand impairments

All hand functions, except tactile pressure detection, were more severely affected on the NDH than on the DH confirming that children with CP have developed their handedness on the less affected side. This explains why half of the CP children in our sample are left-handers as compared to the 10% generally observed in healthy children (Illingworth, 1974). Although DH was less affected than NDH, both hands were significantly impaired in our sample of children with CP as compared with age- and sex-matched healthy children. Indeed, bilateral hand impairments were observed for all types of CP including children with hemiplegia. This finding is in agreement with previous studies (Cooper et al., 1995; Duqué et al., 2003) which also report that the non-paretic hand of hemiplegic children was significantly impaired, although to a lesser extent than the paretic hand.

Motor impairments were markedly more prevalent than sensory impairments, whatever the CP types taken into account. Eighty-nine percents of CP children displayed at least one motor impairment on the DH and 97% on the NDH. On the other hand, only 33% of CP children displayed at least one sensory impairment on the DH and 48% on the NDH. Among motor hand impairments, fine finger dexterity was the most prevalent. Several phylogenetic studies (Heffner & Masterton, 1983; Porter & Lemon, 1993) have highlighted the critical role of the corticospinal tract in fine finger dexterity. Lesion studies in animals and humans (Porter & Lemon, 1993; Lang & Schieber, 2004) have also demonstrated that the ability to perform highly skilled finger movements was severely affected by damage to the corticospinal tract such as observed in CP (Duqué et al., 2003). The finding that gross manual dexterity was less severely affected than fine finger dexterity is in agreement with previous monkey studies (Passingham et al., 1983; Rouiller et al., 1998) showing that an early lesion of the corticospinal tract irremediably disrupted the fine finger dexterity while a noteworthy recovery may be observed in gross manual dexterity.

The significant impairments observed on tactile pressure detection and stereognosis confirm previous reports generally made on smaller samples (Tizard et al., 1954; Tachdjian & Minear, 1958; Cooper et al., 1995; Gordon & Duff, 1999a; Krumlinde-Sundholm & Eliasson, 2002). Some studies (Tizard et al., 1954; Cooper et al., 1995; Krumlinde-Sundholm & Eliasson, 2002) have suggested that stereognosis was more commonly impaired in CP children than tactile pressure detection. This is not supported by our study in which approximately the same proportion of children displayed a significant impairment in tactile pressure detection and stereognosis. However, a severe tactile pressure detection impairment, namely a loss of protective sensation (threshold > 2.05g), was only observed in 6% of the children on the DH and in 15% on the NDH. In our sample of children with CP, proprioception was rarely affected, although previous studies (Tachdjian & Minear, 1958; van Heest et al., 1993) found a higher prevalence of proprioception impairment than we did. This difference might be explained by the fact that these studies did not compare CP scores with those of age-and sex-matched healthy children while, according to our observations, proprioception improves in some healthy children up to 9 years old.

Relationships among hand impairments

The correlation between gross manual and fine finger dexterity indicates that these hand functions were closely related but not identical. This is in accordance with previous studies (Fleishman, 1954; Fleishman & Ellison, 1962) in which fine finger and gross manual dexterity were identified as two different psychomotor abilities. In our study of CP children, both types of dexterity were moderately associated with grip strength. On the contrary, no significant relationship was found between dexterity and grip strength in healthy children (Haward & Griffin, 2002). However, a disease such as CP or stroke (Mercier & Bourbonnais, 2004) could affect in the same proportions both dexterity and grip strength, resulting in a correlation between these functions. Among all sensory impairments, only stereognosis was moderately related to motor impairments. Stereognosis is a very complex function that involves the cortical integration of primary sensory afferents which are decoded and compared with brain representations of familiar objects built up during the whole life of the individual (Thonnard et al., 1994; Krumlinde-Sundholm & Eliasson, 2002). A richer sensory information can be extracted and employed in object identification by using active in-hand manipulation rather than passive manipulation (i.e., objects are stationary put on the palm or are passively rotated over the surface of the skin) (Gibson, 1962). Consequently, stereognosis impairments observed in children with CP may result, at least partially, from in-hand manipulation deficits, hence possibly explaining the relationship observed with motor impairments. Proprioception was poorly related to motor impairments most likely because this hand function was rarely impaired in children with CP. Tactile pressure detection was slightly impaired in our sample of CP children which could support its weak relationship with motor impairments. Although tactile sensory information is generally considered as essential for motor functions (Moberg, 1962), this sensory function must be severely impaired before affecting the motor functions in a significant way (Thonnard et al., 1997 and 1999; Augurelle et al., 2003; Nowak et al., 2003).

Relationship between hand impairments and manual ability

On both hands, motor functions and stereognosis were moderately related to manual ability while tactile pressure detection and proprioception showed no significant association with manual ability. Similar results were found in chronic stroke adults (Penta et al., 2001). Another study (Krumlinde-Sundholm & Eliasson, 2002) also reported that motor functions of hemiparetic children were more vital than sensory functions for the use of the paretic hand in bimanual performance. In our study, gross manual and fine finger dexterity presented the highest correlations with manual ability on the DH while it was gross manual dexterity and grip strength on the NDH. Several studies also found a significant relationship between manual ability and either grip strength or dexterity in CP children with hemiplegia (Brown et al., 1987) and stroke adults (Mercier & Bourbonnais, 2004) but also in other pathologies such as motor delays (Case-Smith, 1996), rheumatoid arthritis (van Lankveld et al., 1998; Adams et al., 2004), and work hand injuries (Gloss & Wardle, 1981). Dexterity is an important function in the achievement of manual activities probably because it reflects the integration of several other hand functions such as mobility, strength, muscle tonus, or sensitivity (Thonnard et al., 1994). Grip strength is also an important function needed to successfully complete manual activities though this type of prehension concerns a minority of daily hand activities (Adams et al., 2004). However, several studies have demonstrated that grip strength was a good indicator of general muscle strength (van Heuvelen et al., 2000; Mercier & Bourbonnais, 2004) and could therefore reflect the strength of various types of prehension.

The multiple linear regression integrating the functions of both hands showed that gross manual dexterity on the DH and grip strength on the NDH were the combination of hand functions which best predicted the variations observed in manual ability measures. This finding suggests that the achievement of manual activities requires the combination of a dexterous DH to manipulate the objects and a strong NDH to stabilize the objects. Such asymmetrical functions of the hands are observed in most manual activities. The NDH classically plays a postural role in holding an object steady, thereby ensuring a spatial reference frame into which the DH can execute a manipulative action (Guiard, 1987). Holding a bottle with the NDH and unscrewing the cap with the DH is a typical example. It can be assumed that the degree of success of such manual activities will depend on the integrity of manipulative functions of the DH and postural functions of the NDH. However, the combination of impairments in gross manual dexterity on the DH and in grip strength on the NDH predicted only 58% of the variance in manual ability measures indicating that CP children with similar hand impairments may vary considerably in their manual ability. Children with CP may compensate their hand impairments by learning adapted strategies (McCuaig & Frank, 1991; Penta et al., 2001). Several bimanual activities (e.g., taking off a T-shirt) may be broken down into several unimanual sequences involving just the less affected hand (Arnould et al., 2004).

Other bimanual activities requiring manipulation with one hand and stabilisation with the other may be achieved by transferring the postural role of the more affected hand to another body structure. For instance, a bottle may be gripped between a child's thighs while the less affected hand unscrews the bottle cap. The success of compensatory strategies will depend on the integrity of both the dominant and the non-dominant hand but also on children's motivation, adaptability, emotional control, cognitive skills, familial and social environment. Parents of children with CP may also adapt their habits to facilitate some activities (e.g., by not overtightening a bottle) or, on the contrary, may inhibit some activities to prevent risk or save time (Arnould et al., 2004). In addition, assisting devices may be adopted to complete the most difficult bimanual activities. So, it is the interaction of many functional, personal, and environmental factors which contributes to the achievement of manual activities.

The present study has shown that, in children with CP, manual ability cannot simply be inferred from hand impairments but should be measured per se with specific and appropriate scales such as ABILHAND-Kids (Arnould et al., 2004). However, statements regarding the causality of the relationship cannot be made since this study was cross-sectional in nature.

Conclusion and perspectives

Hand functioning has evolved through the species to reach its highest level in the human being. The superiority of the human hand results from some modifications in its anatomical structures and biomechanical properties but above all from noteworthy changes in the central nervous system (CNS) controlling the hand movements (Phillips, 1971; Heffner & Masterton, 1983; Porter & Lemon, 1993; Flanagan & Johansson, 2002). The hand functioning of the humans may therefore be disrupted by various brain lesions. Cerebral palsy (CP) which results from an injury in the developing CNS is the most common cause of physical disability in children (Rosenbaum, 2003). It commonly affects the brain structures responsible of highly skilled hand movements (Uvebrant, 1988; Yokochi et al., 1992). Consequently, the sensorimotor functions of the hand (e.g., grip strength, dexterity, sensitivity) are frequently impaired and constitute the main problem in many children with CP (Uvebrant, 1988).

Although one fundamental goal of rehabilitation is to improve the children's ability to manage the daily activities necessary for autonomous living (Ottenbacher et al., 1999; Case-Smith, 2001; Ketelaar et al., 2001; Ostensjo et al., 2003), most of the conventional treatments are focused on the hand impairments (Case-Smith, 1995 and 1996; Ketelaar, 2001; Ostensjo et al., 2003; Rosenbaum, 2003). For instance, many interventions aim at lengthening spastic muscles, improving the strength of weakened muscles, increasing the range of motion at restricted joints, or improving movement coordination (Goldstein, 2004). This traditional approach is based on the assumption that when hand functions improve, the capacity to manage manual activities (i.e., manual ability) improves in parallel (Case-Smith, 1995 and 1996; Ostensjo et al., 2003). This assumption is not strictly supported by the International Classification of Functioning, Disability and Health (ICF) which conceptualizes the manual ability as the result of a bidirectional interaction between hand functions and several other contextual factors (World Health Organization, 2001). According to the ICF, the improvement in hand functions may lead to a higher manual ability but not necessarily in a predictable straightforward relationship.

The purpose of the present work was to validate empirically the conceptual framework proposed by the ICF for hand functioning in children with CP. Several hand functions including gross manual and fine finger dexterity were measured in children with CP. Appraising the degree of hand impairments requires normative data to differentiate the real dysfunctions of CP children from the normal difficulties according to their age, sex, or handedness. As there is no normative data for gross manual and fine finger dexterity, a first experiment was conducted to establish these norms. A second experiment was carried out to develop through the Rasch model a measure of manual ability in children with CP since such a measure was not yet available. Finally, a third experiment was performed to investigate the relationship between the hand impairments and the manual ability of children with CP.

In the first experiment, we have investigated, from childhood to early adulthood, the developmental changes occurring with age as well as the influence of handedness and sex on fine finger and gross manual dexterity (Chapter 1). Normative data were also established for both types of dexterity. The results have shown that fine finger dexterity significantly increased with age until 10 years old and gross manual dexterity until 17-18 years old; the dominant hand presented a higher dexterity than the non-dominant hand; and girls had a higher fine finger dexterity than boys on their dominant hand. Therefore, the measurement of dexterity impairments in children with CP should take age, handedness, and sex into account to determine properly the extent to which the child's dexterity deviates from normal. Several hypotheses related to the neurobiological correlates of dexterity were suggested to explain the results of the study. The development of fine finger dexterity until 10 years old seems to tally with the maturation of the corticospinal tract, confirming the critical role of this structure in the capacity to move the hand and the fingers with skill. The longer development of gross manual dexterity has been suggested to reflect the maturation of other neuromotor systems than the corticospinal tract as well as the development of more efficient strategies. The higher dexterity of the dominant hand seems to be consistent with the asymmetrical structure of the motor cortex in the cerebral hemispheres. As compared with the non-dominant hemisphere, the dominant hemisphere shows a larger and more connected motor cortex that results in a more effective experience-based reorganization in the brain. Finally, it has been suggested that girls had a higher fine finger dexterity than boys on their dominant hand as the girls' dominant hemisphere presented a more connected motor cortex. However, all of these hypotheses remain at a theoretical level and need to be tested empirically. It would be interesting to use brain exploring techniques such as magnetic resonance imagery (MRI) or transcranial magnetic stimulation (TMS) in combination with the measure of dexterity in healthy children to better understand the cerebral structures involved in the dexterity.

In the second experiment, we have developed and validated a measure of manual ability in children with CP (Chapter 2). An item questionnaire, ABILHAND-Kids, was developed from existing scales. Experts' advice was used to explore the most representative inventory of children's manual activities. The questionnaire addresses the perception of a child's difficulty in performing manual activities. For each activity, CP children and their parents were asked to provide their responses on a three level-scale (0: impossible, 1: difficult, or 2: easy). Children's and parents' responses were analysed separately with the Rasch measurement model which converts finite ordinal total scores into continuous interval measures on a unidimensional scale. The Rasch model was successfully used to select items presenting an ordered rating scale, sharing the same response format, and fitting a unidimensional scale and to compare children's and parents' perceptions. Rasch analysis has shown that children had a dichotomous perception of their manual ability; the activities were perceived as either "impossible" or "easy" with very rare intermediate responses. The polytomous parents' perception appeared to be a more accurate source of information about manual ability than the dichotomous children's perception, leading to a wider range of measurement and a higher reliability. The ABILHAND-Kids questionnaire was therefore exclusively built from the parents' perceptions. The 21 items retained for the ABILHAND-Kids scale showed an invariant difficulty hierarchy over time and across various demographic and clinical subgroups of CP children, supporting the clinical application of ABILHAND-Kids as a measure of manual ability in children with CP.

In the third experiment, we have provided a clinical picture of hand impairments in children with CP and investigated their relationship with manual ability as measured with the ABILHAND-Kids questionnaire (Chapter 3). Motor impairments, markedly more prevalent than sensory impairments, showed a moderate association with manual ability. Gross manual dexterity on the dominant hand and grip strength on the non-dominant hand were the combination of hand functions which best predicted the manual ability of children with CP. This finding suggests that the achievement of manual activities requires the combination of a dexterous dominant hand to manipulate the objects and a strong non-dominant hand to stabilize the objects. However, this combination of hand functions predicts only 58% of the variance in manual ability measures indicating that CP children with similar hand impairments may vary considerably in their manual ability. Many factors, other than the hand impairments investigated in the study, might influence the child's ability to achieve manual activities (e.g., compensatory techniques, cognitive impairments, environmental and personal factors). Consequently, manual ability cannot simply be inferred from the underlying hand impairments.

The assessment of hand impairments was focused on the measurement of three motor (i.e., grip strength, gross manual dexterity, fine finger dexterity) and three sensory (i.e., tactile pressure detection, stereognosis, proprioception) functions which were thought to be relevant with regard to the hand functioning of children with CP. While it is relatively clear that the grip strength and the sensory functions belong to the body functions, the place of the dexterity in the ICF framework may appear less obvious. People may be tempted to classify the dexterity at the individual level (i.e., activities dimension) rather than at the body level (i.e., body functions dimension) for several reasons. First, dexterity is sometimes used to estimate the performance of the hands and upper limbs in the activities of daily living (Desrosiers et al., 1991). Second, some studies (Williams & Hadler, 1981; Williams et al., 1982; Ostwald et al., 1989; Falconer et al., 1991) have shown a good correlation between dexterity and the independence in activities of daily living. Third, several instruments aiming at measuring manual ability (Jebsen et al., 1969; MacBain, 1970; Potvin et al., 1972; Smith, 1973; Taylor et al., 1973; Wilson et al., 1984; Desrosiers et al., 1995; Kopp et al., 1997) evaluate the time of execution of manual activities. Using the time of execution gives the impression that the results allow quantitative comparisons and statistical operations as it is the case in dexterity tests. However, for the purpose of measuring manual ability, the time recorded with such instruments is essentially ordinal and therefore fails to make quantitative comparisons (Fisher, 1992). A child who achieves a given activity twice as fast as another is not necessarily twice as able; it means solely that the child is twice as fast (Merbitz et al., 1989; Wright & Linacre, 1989). Unfortunately, this has contributed to the confusion between dexterity and manual ability. Dexterity is defined as "a manual skill that requires rapid coordination of fine and gross voluntary movements" (Poirier, 1987) while manual ability is defined as "the capacity to use the hands and upper limbs in managing manual activities of daily life" (Penta et al., 2001). The two concepts make a clear distinction between a purposeless functioning and a functioning that has a purpose at the individual level (McDougall & Miller, 2003). Dexterity refers to the physiological functions of the hand and the CNS which make it possible to execute rapid and coordinated hand movements without any specific purpose. By contrast, manual ability refers to the use of a combination of hand functions (e.g., dexterity, grip strength, sensory functions) for the purpose of executing activities that are generally considered as essential for the individual's everyday life. Moreover, the tasks classically used in the dexterity tests (e.g., manipulating pegs or blocks) are too artificial in nature and require too limited movement patterns (e.g., a standardized grasping is used) to reproduce the meaningful situations encountered in the daily life (Neistadt, 1994; Desrosiers et al., 1995; Chen et al., 2005). Consequently, it seems more judicious to classify the dexterity in the body functions rather than in the activities dimension of the ICF.

Other hand functions than those measured in this work may also be impaired in CP children and may play a role in the achievement of manual activities. For instance, one study (Brown et al., 1987) has shown that the distal spasticity of hemiplegic children was related to the performance observed in various manual tasks when the spasticity was severe. However, this relationship was markedly smaller in children with moderate and mild spasticity whose performance in manual tasks widely varied while they had the same degree of hypertonus. Another hand function which seems interesting to explore in CP children is the tactile spatial resolution (i.e., perception of spatial features of objects and surfaces). Several studies have shown that tactile spatial resolution was frequently affected in CP children and at least more often impaired than tactile pressure detection (Wislon & Wilson, 1967b; Gordon & Duff, 1999a; Krumlinde-Sundholm & Eliasson, 2002). Unlike tactile pressure detection which largely reflects the integrity of peripheral nerve fibres (Weinstein, 1993), tactile spatial resolution involves the cortical integration of peripheral impulses (Gordon & Duff, 1999a). Tactile spatial resolution seems therefore more appropriate for the detection of cortical lesions such as those observed in CP than tactile pressure detection (Krumlinde-Sundholm & Eliasson, 2002). However, the role of tactile spatial resolution in executing motor functions and its influence on manual ability remain to be empirically tested in children with CP.

Although the motor impairment syndromes of CP children are non-progressive (Mutch, 1992), hand functioning may change over time (Kuban & Leviton, 1994). Several treatments have been developed to improve children's manual ability but their effectiveness was hardly ever established probably due to the lack of appropriate instruments (Pagliano et al., 2001). ABILHAND-Kids was therefore developed using the Rasch model to obtain a linear and unidimensional scale measuring the ability of the children to use their hands in daily activities. ABILHAND-Kids appears to be precise enough to discriminate children's manual abilities and, presumably, to capture even subtle manual ability changes over time that may result for instance from surgery, rehabilitation, or biomedical treatment. However, a longitudinal study is required to verify empirically the responsiveness of ABILHAND-Kids as well as to investigate its predictive validity.

ABILHAND-Kids was calibrated from the responses of the children's parents as the parents provided a more precise and reliable source of information about their child's manual ability than the children themselves. Furthermore, the use of the parents’ perceptions on behalf of the children's makes it possible to measure the manual ability of severely impaired children as well as those with mental, psychological, attentional or communicative disorders. However, it remains to determine whether parents provide more valid information about their child's manual ability than the children themselves. One way to investigate this issue would be to compare the children's performance observed during the achievement of the 21 ABILHAND-Kids activities[1] with the perceptions of both CP children and their parents about the difficulty experienced by the child in performing the 21 activities. Perceptions which will best match the observed performance can be assumed to be the most valid indicators of the child's manual ability.

ABILHAND-Kids was calibrated in CP children older than 6 years old to focus on subjects with mature manipulative skills in activities of daily life (Illingworth, 1975). This eligibility criterion was chosen to develop a manual ability scale sensitive to the pathological disruption of manual ability rather than to a maturation of manual ability. This was confirmed by the significant relationships observed between ABILHAND-Kids measures and some indexes related to the severity of CP (i.e., school education, type of CP, Gross Motor Function Classification System) while no relationship was found with the age (Arnould et al., 2004). However, these results do not preclude a potential relationship with age in younger children with immature manual ability. Several developmental studies have shown that the daily living skills develop during the early childhood (Gesell et al. 1940; Gesell & Ilg, 1943; Gesell & Amatruda, 1965; Coley, 1978; Brigance, 1978; Henderson, 1995). Although these studies provide information on the ages at which children typically master manipulative skills in daily activities, they are merely based on qualitative observations. As a complement, the ABILHAND-Kids questionnaire could be calibrated in healthy children younger than 6 years old. This calibration of ABILHAND-Kids should provide the hierarchy of item difficulties characterizing the ontogenesis of manual ability. The disruption in manual ability development due to CP could then be appraised by submitting the calibrated questionnaire to young disabled children.

[1] The observational criteria for assessing the children’s performance during the achievement of the 21 ABILHAND-Kids activities could, for instance, include the observed difficulty, the degree of external assistance, the time required to do the activities, or the precision/quality of the performance.

Although the motor impairment syndromes persist into adulthood, CP is predominantly viewed as a paediatric disorder (Maruishi et al., 2001). As a consequence, there is little information about the hand functioning of adults with CP and that despite the increasing number of CP adults resulting from the medical and technical advances (Bottos et al., 2001). In this work, we have solely studied the hand functioning of CP children aged between 6 and 15 years old. ABILHAND-Kids therefore includes manual activities relevant for children but not necessarily for adults. Indeed, several typically adult activities (e.g., “Hammering a nail”, “Threading a needle”, “Peeling potatoes”) were removed following the experts' suggestions during the questionnaire development process (Arnould et al., 2004). Nevertheless, such activities may be informative if we want to measure the manual ability of adults with CP. The ABILHAND questionnaire (Penta et al., 2001) includes typically adult activities and was developed using the Rasch model in chronic stroke adults. Actually, ABILHAND is the counterpart of ABILHAND-Kids but for adults. Although the manual ability measures obtained in ABILHAND and ABILHAND-Kids cannot be compared as such, their comparison would be possible if a common metric delineating manual ability is constructed (Smith, 1992). The Rasch model can be used to locate ABILHAND and ABILHAND-Kids items onto a common metric even if they are administered to completely separate groups (McHorney & Cohen, 2000), provided that all items fit a single unidimensional construct. In practice, combining items from two different scales onto a single variable just entails that some common items are shared by both scales and that the items of both scales fit a unidimensional construct (Wright & Stone, 1979; Ingebo, 1997). The common items allow the two otherwise different scales to be connected together by adjusting their local origins provided that the difficulty hierarchy of the common items is invariant across both scales (Smith, 1992; McHorney & Cohen, 2000; Wolfe, 2000). ABILHAND and ABILHAND-Kids share 11 common items that can therefore be used to link both scales. Overall, the common items constitute the more difficult items of ABILHAND-Kids while they represent the less difficult items of ABILHAND indicating that ABILHAND-Kids tends to be less challenging than ABILHAND. The co-calibration of both scales should therefore extend the measurement range of each separate scale allowing the manual ability of a CP subject to be followed from childhood to adulthood.

The original version of ABILHAND-Kids was developed in French. However, other versions are currently available in English and Dutch. The latter were established through independent forward and backward translations from the original French version in a attempt to make new versions a replica of the original. However, linguistic equivalence does not guarantee metric equivalence (Tesio, 2003). Different cultural traditions may result in subtle differences in the degree of difficulty of the activities. For instance, the activity "eating" can be much more difficult in Asia than in Europe as chopsticks are used. It is therefore essential to assess the equivalence of ABILHAND-Kids item calibrations across different cultures. This can be easily performed with the Rasch model through differential item functioning tests (Wright & Stone, 1979; Smith, 1992). The difficulty hierarchy of ABILHAND-Kids activities should remain the same across the cultures to obtain a cross-cultural measure of manual ability in children with CP; just like one meter of fabric represents the same length of fabric throughout the world. Consequently, activities showing a differential functioning across the cultures should be either deleted or split into several items that are specific to each culture (Tennant et al., 2004).

Conceptually, ABILHAND-Kids has been developed to measure the extent to which children are able to use their hands in executing manual activities, whatever their underlying health condition. A sample of CP children was chosen to calibrate the questionnaire as CP covers a wide range of hand disability and represents the most frequent diagnosis of children who are receiving physical therapy (Tieman et al., 2004). We hope that in the future the ABILHAND-Kids questionnaire will be investigated in other pediatric disorders with the view of building a generic measure of children's manual ability. However, the way a disorder leads to a limitation in children's manual ability might be specific to the disorder itself. It is therefore crucial to verify how closely any given activity of ABILHAND-Kids represents an invariant challenge across diagnosis (Wright & Stone, 1979). For the purpose of building a generic children's manual ability measure, activities identified as diagnosis-specific should be either removed from the questionnaire or calibrated at a diagnosis specific difficulty (Tennant et al., 2004).

The investigation of the relationship between hand impairments and manual ability has shown that sensory impairments, markedly less prevalent than motor impairments, seem not to play a significant role in the capacity of CP children to perform manual activities. In our opinion, the sensory functions of CP children measured in this work are not enough impaired to affect the achievement of manual activities in a significant way (Thonnard et al., 1999; Nowak et al., 2003). However, it remains to be verified whether other sensory functions are more severely affected in children with CP (e.g., tactile spatial resolution) and whether they significantly affect manual ability.

Gross manual dexterity on the dominant hand and grip strength on the non-dominant hand are the combination of hand functions which best predict the manual ability of children with CP. This finding emphasizes that manual activities typically require the cooperation of both hands which tend to be specialized for different functions (Guiard, 1987; MacNeilage, 1990; Wiesendanger et al., 1996). For instance, when we remove the lid from a jar or we button up a shirt, the non-dominant hand holds the object in a stable position while the dominant hand acts upon it. Hence, the non-dominant hand plays a postural role in stabilizing the grasped object and at the same time provides a spatial reference frame into which the dominant hand manipulates the object (MacNeilage, 1990). However, saying that the non-dominant hand offers stability does not mean that the hand is immobile. On the contrary, the non-dominant hand ensures a plastic stabilization and therefore produces steady states that are subject to frequent alterations (Guiard, 1987). For instance, in sewing, the non-dominant hand periodically re-positions the fabric so that the position and the orientation of the fabric always remain appropriate to the action of the dominant hand. It is therefore fallacious to differentiate the roles of the hands in terms of a stationary non-dominant hand and a mobile dominant hand as mobility is undeniably required on both sides. This is consistent with our findings that gross manual dexterity presents the highest correlation with manual ability on both hands, followed by fine finger dexterity on the dominant hand and grip strength on the non-dominant hand. In other words, the achievement of manual activities requires: 1) a highly dexterous dominant hand to perform both fine and gross manipulations, and 2) a strong and an enough dexterous non-dominant hand to stabilize the objects and to provide a spatial reference frame which is always appropriate to the action of the dominant hand.

The combination of the hand functions, however, predicts only 58% of the variance in the manual ability measures of children with CP. This finding supports the theoretical standpoint of the ICF that hand impairments and manual ability are not straightforwardly related since other contextual factors interfere[2] (World Health Organization, 2001). The ICF therefore appears to be a suitable framework useful for describing the impact of CP on child's hand functioning and for organizing this information in a structured and meaningful way. We encourage the health care providers to use the ICF framework during the rehabilitation process to identify the problems and needs of the child, determine the factors of the child and the environment that are related to the problems, define realistic therapeutic goals, plan and implement the most appropriate interventions, and assess the effectiveness of the interventions (Steiner et al., 2002; Stucki et al., 2002). We are convinced that the application of the ICF in clinical practice will lead to a more rational and efficient approach to the hand rehabilitation management of children with CP.

The finding that hand impairments only partially predict the manual ability measures in children with CP has several clinical implications. The therapist cannot assume that the reduction of hand impairments will necessarily result in a higher manual ability. Consequently, interventions focused just on the reduction of hand impairments may be questionable, especially as it is more important for the child to manage daily activities to be autonomous than to have “normal” hand functions (Ketelaar et al., 2001). A comprehensive intervention should always endeavor to

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[2] The ICF suggests that both personal and environmental contextual factors may facilitate or hinder the achievement of manual activities and could therefore explain, in our study, the residual variance observed in manual ability measures. However, such contextual factors and their influence on manual ability have not been investigated in the present work. Our study simply shows that other factors than just hand impairments may contribute to the achievement of manual activities, and thus that manual ability cannot simply be inferred from hand impairments. Consequently, future researches are required to identify which factors really influence the achievement of manual activities.

improve manual ability by training the child to perform the daily activities that are limited (Bekkering et al., 2001). The therapist should teach the child to optimize the use of his or her existing hand functions in the management of meaningful activities. The most effective way to manage a specific activity is to practice regularly that activity (Mathiowetz, 1993). Teaching adapted strategies should also be an important part of the hand rehabilitation as they can help the child compensating for the hand impairments (McCuaig & Frank, 1991; Wade, 1997; Penta et al., 2001). They are particularly useful when the reduction of some hand impairments is hardly or not possible (Nakayama et al., 1994; Case-Smith, 2001). For instance, a child with hemiparesis may learn a one-handed buttoning technique to compensate his or her paretic hand. The therapist must so far as possible enable the child to have an active role in finding adaptive strategies for the achievement of daily activities (Ketelaar et al., 2001). Indeed, teaching the child to find self-initiated solutions is important as the child will continually be confronted with new challenges (Ketelaar et al., 2001). Several contextual factors of the person (e.g., cognitive status, motivation, temperament) and the environment (e.g., health services, parental expectations, financial support) may facilitate or hinder the achievement of manual activities and thus should be considered in the rehabilitation process (Case-Smith, 1995; Bartlett & Palisano, 2000 and 2002). The therapist should attempt to identify the contextual factors that are crucial for the child's manual ability (Steiner et al., 2002) and to find what can be change within these contextual factors to facilitate the achievement of daily activities (Law, 1993). However, some contextual factors are hardly modifiable (e.g., the attitudes of the society, the parents' incomes, the community facilities) (Stucki et al., 2003). When planning the rehabilitation intervention, it is thus necessary to address the contextual factors that are easily modifiable and that have the greatest potential to improve the child's manual ability (Stucki et al., 2003). In future researches, it would be interesting to identify such contextual factors and to examine the strength of their relationship with manual ability.

The present work stresses the importance to address the impact of CP on child's hand functioning beyond the body level. Manual ability is not simply the integration of hand functions in daily activities and must therefore be treated and measured per se. It does not mean that interventions intended to reduce hand impairments are useless. They may be important, especially for preventing secondary impairments such as contractures or deformities (Mathiowetz, 1993; Nakayama et al., 1994; Case-Smith, 2001). However, improving child's manual ability is of great importance and should be a major goal in the hand rehabilitation of children with CP.

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Appendices

I. Item reduction process of ABILHAND-Kids

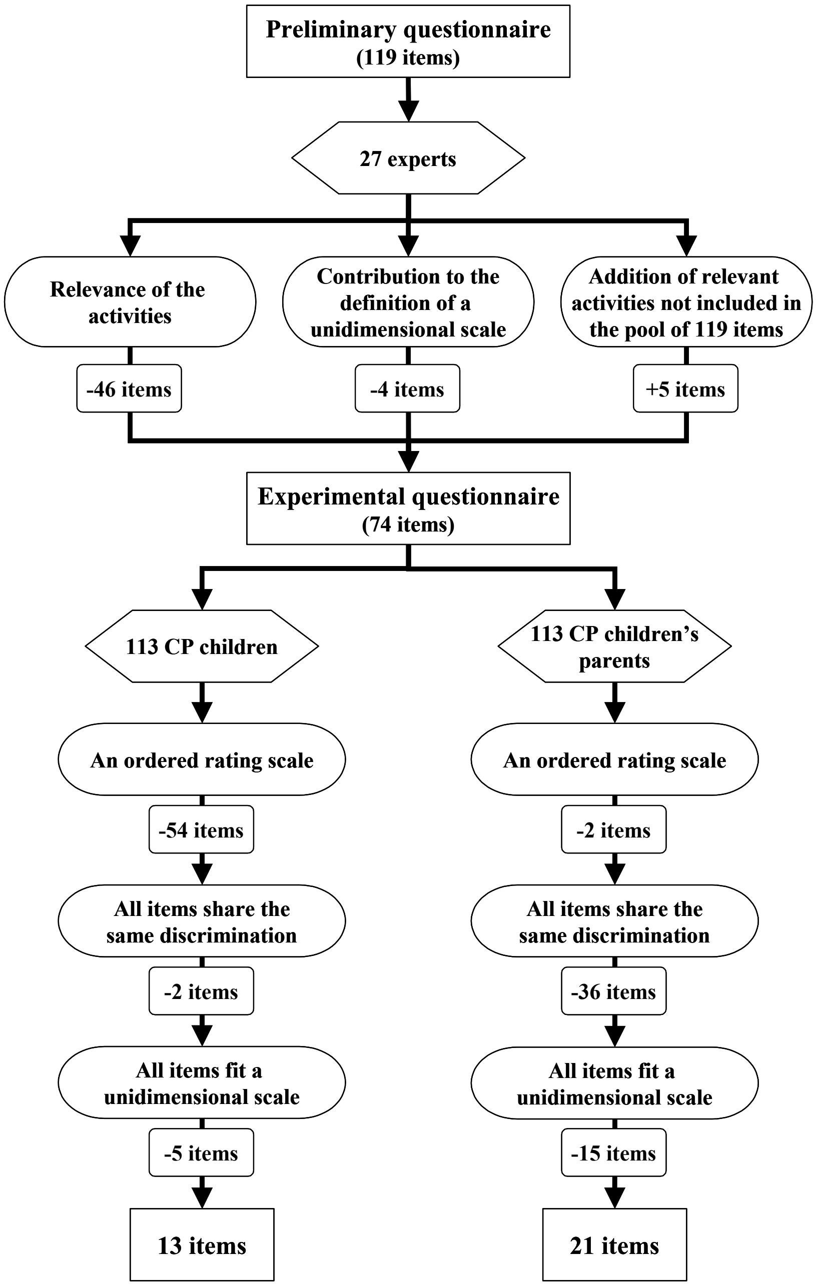
II. Preliminary version of ABILHAND-Kids

III. Experimental version of ABIHAND-Kids

IV. ABILHAND-Kids questionnaire

V. ABILHAND questionnaire

Appendix I.
Item reduction process of ABILHAND-Kids



Appendix II.
Preliminary version of ABILHAND-Kids

Items of the preliminary questionnaire*	Origin**	Reason(s) for item deletion***
Eating		
Eating with the fingers	ASK, PEDI	Too imprecise (depends on the nature of the food)
Using a fork	ASK, Denver, PEDI	
Drinking soup out of a bowl with a spoon	PEDI	Irrelevant (activity generally performed without spoon or with spoon out of a plate) Too imprecise (depends on the degree of cleanliness requested)
Cutting meat	ABILHAND	
Cutting pasta	PEDI (modified)	Irrelevant (rarely performed by children)
Clearing the table	Devised item	Too imprecise (depends on what is clearing from the table) Involvement of lower limbs Requirement of sequential organization (disrupted by praxic problems)
Lifting an open cup with two hands	Denver, PEDI (modified)	Too imprecise (depends on how the bowl is filled) Sometimes it is more easy with one hand, sometimes with two hands Preference: Drinking a glass of water
Lifting a cup with one hand	Denver, PEDI	Too imprecise (depends on how the bowl is filled) Preference: Drinking a glass of water
Drinking a glass of water	ABILHAND	
Picking up a can	ABILHAND	
Put a glass of water on the table	ABILHAND	
Unscrewing a bottle cap	Devised item	
Filling a glass with water	ASK (modified)	
Taking the cap off a bottle	ABILHAND	Irrelevant (typically adult activity)
Opening a jar of jam	ABILHAND (modified)	
Opening a bread box	Devised item	
Opening a packet of biscuits	Devised item	
Unwrapping a chocolate bar	ABILHAND	
Opening a bag of chips	ABILHAND	
Eating a sandwich	ABILHAND	
Unwrapping a sweet	Devised item	
Spreading butter on a slice of bread	ABILHAND	
Shelling hazel nuts	ABILHAND	Irrelevant (uncommon activity)
Peeling one banana	Devised item	
Eating an ice-cream cone	Devised item	

* The 74 items of the preliminary questionnaire which were selected for the experimental questionnaire are in bold type.
** ASK = Activities Scale for Kids; Denver = Denver Developmental Screening Test, versions I and II; KB = Klein-Bell Activities of Daily Living Scale; PEDI = Pediatric Evaluation of Disability Inventory.
*** Items were deleted from the experts' responses.

Items of the preliminary questionnaire*	Origin**	Reason(s) for item deletion***
Grooming		
Washing one's hands with soap	ABILHAND, Denver, PEDI	
Drying one's hands	Denver, PEDI	
Washing one's face	ABILHAND, PEDI	
Washing the upper body	ASK, PEDI	
Drying the upper body after a bath or a shower	PEDI	
Washing one's hair	Devised item	Irrelevant (rarely performed by young children) Involvement of lower limbs Requirement of balance
Combing one's hair	ABILHAND, ASK, KB, PEDI	
Putting a hairband	Devised item	Irrelevant (mainly performed by girls)
Putting a hair rubber band	Devised item	Irrelevant (mainly performed by girls) Requirement of sequential organization (disrupted by praxic problems)
Opening the cap of a toothpaste tube	PEDI	
Squeezing toothpaste onto a toothbrush	ABILHAND, ASK, KB	
Brushing one's teeth	ABILHAND, ASK, KB, PEDI	
Cutting one's nails	ABILHAND, KB	
Filing one's nails	ABILHAND	Irrelevant (uncommon activity)
Blowing one's nose	ABILHAND, KB, PEDI	
Wiping oneself thoroughly after toileting	ASK, KB, PEDI	Involvement of lower limbs Requirement of balance
Flushing the toilet	ASK, KB	
Turning on a tap	ASK, KB	
Turning off a tap	Devised item	Too imprecise (depends on the type of tap) Similar to "Turning on a tap" except that it does not require a controlled use of strength

* The 74 items of the preliminary questionnaire which were selected for the experimental questionnaire are in bold type.

** ASK = Activities Scale for Kids; Denver = Denver Developmental Screening Test, versions I and II; KB = Klein-Bell Activities of Daily Living Scale; PEDI = Pediatric Evaluation of Disability Inventory.

*** Items were deleted from the experts' responses.

Items of the preliminary questionnaire*	Origin**	Reason(s) for item deletion***
Dressing		
Putting on a pair of underpants/knickers	Devised item	Involvement of lower limbs Too imprecise (sitting or standing position?) Requirement of sequential organization (disrupted by praxic problems)
Taking off a pair of underpants/knickers	Devised item	Involvement of lower limbs Too imprecise (sitting or standing position?)
Buttoning up trousers	ABILHAND	
Zipping up trousers	ABILHAND, ASK	
Putting on a T-shirt	ASK, Denver, PEDI	
Taking off a T-shirt	PEDI	
Buttoning up a shirt/sweater	ABILHAND, ASK, Denver	
Zipping up a jacket	ABILHAND, ASK	
Fastening the snap of a jacket	ABILHAND	
Putting a shirt inside trousers	KB	Irrelevant (children have rarely a shirt or leave it outside trousers to follow fashion)
Putting on socks	ASK, PEDI	Involvement of lower limbs Requirement of balance
Removing socks	PEDI	Involvement of lower limbs Requirement of balance
Putting on shoes without laces	PEDI	
Putting on lace-up shoes	PEDI	
Taking off shoes	Devised item	Irrelevant (most children take off their shoes without their hands) Disrupted by foot deformities
Putting on a wristwatch	Devised item	Misfit
Putting on gloves	Devised item	Uncommon activity (just in winter) (mittens are more common than gloves for young children)
Putting on a hat	KB (modified)	
Putting on one's glasses	KB	Irrelevant (exclusively for children with glasses)

* The 74 items of the preliminary questionnaire which were selected for the experimental questionnaire are in bold type.

** ASK = Activities Scale for Kids; Denver = Denver Developmental Screening Test, versions I and II; KB = Klein-Bell Activities of Daily Living Scale; PEDI = Pediatric Evaluation of Disability Inventory.

*** Items were deleted from the experts' responses.

Items of the preliminary questionnaire*	Origin**	Reason(s) for item deletion***
Environment		
Ringing a door bell	Devised item	
Opening a door	Devised item	
Closing a door	Devised item	Irrelevant (most children slam the door) Preference: Opening a door
Turning a key in a keyhole	Devised item	
Opening a window	Devised item	Too imprecise (depends on the type of window's opening) Irrelevant (rarely performed by young children as it is dangerous) Preference: Opening a door/fridge door
Opening a cupboard	Devised item	Too imprecise (depends on the type of cupboard) Preference: Opening a door/fridge door
Opening a drawer	KB	Too imprecise (depends on the type of drawer: size, handle, …)
Switching on a bedside lamp	ABILHAND (modified)	
Turning on a radio	ABILHAND	Too imprecise (depends on the type of radio: push button or knob?)
Turning on a television set	ABILHAND	
Opening the fridge door	Devised item	
Throwing something in the bin	Devised item	Too imprecise (depends on whether there is a lid) (depends on the type of lid's opening: with hands or foot?)
Communication		
Turning over the pages of a comic book	ABILHAND (modified)	
Handling a 4 colour ballpoint pen with one hand	ABILHAND	Irrelevant (uncommon activity)
Handwritting	ABILHAND	Too imprecise (handwritting a word or a sentence?) Disrupted by praxic, visual, instrumental problems
Opening a mail	ABILHAND	
Putting a letter into an envelope	Devised item	Misfit
Using a standard computer keyboard	Devised item	Too imprecise (one or several keys at a time?) (just press a button or using a keyboard to make particular things?)
Inserting a diskette into a drive	ABILHAND	
Using a keypad phone	ABILHAND, KB	

* The 74 items of the preliminary questionnaire which were selected for the experimental questionnaire are in bold type.
** ASK = Activities Scale for Kids; Denver = Denver Developmental Screening Test, versions I and II; KB = Klein-Bell Activities of Daily Living Scale; PEDI = Pediatric Evaluation of Disability Inventory.
*** Items were deleted from the experts' responses.

Items of the preliminary questionnaire*	Origin**	Reason(s) for item deletion***
Do-it-yourself		
Drawing	ABILHAND	Too impecise (doodleing or drawing inside lines?) (drawing with a pencil or a pastel?) Disrupted by praxic problems
Colouring	Devised item	
Painting	Devised item	
Drawing a line with a slat	Devised item	
Sharpening a pencil	ABILHAND	
Handling a rubber	Devised item	
Cutting a sheet with scissors by following a line	Devised item	
Handling a stapler	ABILHAND	Misfit
Handling a punch	Devised item	
Handling an adhesive tape	Devised item	
Sticking a stamp on an envelope	Devised item	Too imprecise (lickable stamp or self-adhesive stamp?) Disrupted by salivation problems
Threading a needle	ABILHAND	Irrelevant (typically adult activity) Preference: Threading pearls
Using a screwdriver	ABILHAND	Irrelevant (typically adult activity)
Hammering a nail	ABILHAND	Irrelevant (typically adult activity)
Leisure and play		
Throwing a ball	Denver	
Catching a ball	Denver	
Rolling a small toy car	Devised item	Irrelevant (mainly performed by boys)
Threading pearls	Devised item	Misfit
Piling up some Lego blocks	Denver (modified)	
Throwing a dice	Devised item	
Holding cards in one's hands	Devised item	Too imprecise (depends on the number of cards)
Dealing cards	Devised item	
Handling a joystick	Devised item	

* The 74 items of the preliminary questionnaire which were selected for the experimental questionnaire are in bold type.
** ASK = Activities Scale for Kids; Denver = Denver Developmental Screening Test, versions I and II; KB = Klein-Bell Activities of Daily Living Scale; PEDI = Pediatric Evaluation of Disability Inventory.
*** Items were deleted from the experts' responses.

Items of the preliminary questionnaire*	Origin**	Reason(s) for item deletion***
Miscellaneous		
Making a pellet of paper	Devised item	Irrelevant (no functional interest)
Opening a pencil box	Devised item	Too imprecise (pencil box with zip or snap?) Preference: Zipping up trousers; Fastening the snap of a jacket
Opening a coin purse	Devised item	Too imprecise (coin purse with zip or snap?) Preference: Zipping up trousers; Fastening the snap of a jacket
Grasping a coin on a table	ABILHAND	
Taking a coin out of a pocket	ABILHAND	
Holding the hand of somebody	Devised item	Irrelevant (no functional interest)
Putting on a backpack/schoolbag	ASK	
Carrying a grocery bag	Devised item	Too imprecise (depends on the grocery bag: weight, size, …)
Opening a gift	Devised item	Too imprecise (tearing up or opening thoroughly?)
Scratching behind the head	Devised item	Irrelevant (no functional interest)
Drying up a plate	Devised item	Irrelevant (typically adult activity)
Opening a cardoor	KB	
Closing a cardoor	KB	
New items added following the experts' suggestions		
Putting a coin into a piggy bank		
Putting a key into a keyhole		
Hanging up a jacket		
Fastening one's seat belt		
Rolling up a sleeve of a sweater		

* The 74 items of the preliminary questionnaire which were selected for the experimental questionnaire are in bold type.
** ASK = Activities Scale for Kids; Denver = Denver Developmental Screening Test, versions I and II; KB = Klein-Bell Activities of Daily Living Scale; PEDI = Pediatric Evaluation of Disability Inventory.
*** Items were deleted from the experts' responses.

Appendix III.
Experimental version of ABILHAND-Kids

Items of the experimental questionnaire*	Reason for item deletion**	
	Parents	Children
Eating		
Using a fork	Misfit	Disordered thresholds
Cutting meat	Discrimination > the average	
Drinking a glass of water	Discrimination < the average	Disordered thresholds
Picking up a can	Disordered thresholds	Disordered thresholds
Put a glass of water on the table	Discrimination < the average	Disordered thresholds
Unscrewing a bottle cap		Disordered thresholds
Filling a glass with water		Disordered thresholds
Opening a jar of jam		Disordered thresholds
Opening a bread box		
Opening a packet of biscuits	Discrimination > the average	
Unwrapping a chocolate bar		Disordered thresholds
Opening a bag of chips		Disordered thresholds
Eating a sandwich	Misfit	Misfit
Unwrapping a sweet	Discrimination > the average	Disordered thresholds
Spreading butter on a slice of bread	Discrimination > the average	
Peeling one banana	Discrimination > the average	Disordered thresholds
Eating an ice-cream cone	Discrimination < the average	
Grooming		
Washing one's hands with soap	Discrimination > the average	Disordered thresholds
Drying one's hands	Discrimination > the average	
Washing one's face	Misfit	Disordered thresholds
Washing the upper body		Disordered thresholds
Drying the upper body after a bath or a shower	Discrimination > the average	Disordered thresholds
Combing one's hair	Discrimination > the average	Disordered thresholds
Opening the cap of a toothpaste tube		Disordered thresholds
Squeezing toothpaste onto a toothbrush		Disordered thresholds
Brushing one's teeth	Misfit	Disordered thresholds
Cutting one's nails	Misfit	Disordered thresholds
Blowing one's nose	Misfit	Disordered thresholds
Flushing the toilet	Discrimination < the average	Disordered thresholds
Turning on a tap	Discrimination < the average	Disordered thresholds
Dressing		
Buttoning up trousers		Disordered thresholds
Zipping up trousers		Disordered thresholds
Putting on a T-shirt	Discrimination < the average	Disordered thresholds
Taking off a T-shirt		
Buttoning up a shirt/sweater		
Zipping up a jacket		Disordered thresholds
Fastening the snap of a jacket		
Putting on shoes without laces	Discrimination < the average	Disordered thresholds
Putting on lace-up shoes	Discrimination < the average	Disordered thresholds
Putting on a hat		Disordered thresholds

* The 21 items of the final verison of the ABILHAND-Kids questionnaire are in bold type.
** Items were deleted from the parents' and children's responses.

Items of the experimental questionnaire*	Reason for item deletion**	
	Parents	Children
Environment		
Ringing a door bell	Discrimination < the average	Disordered thresholds
Opening a door	Misfit	Disordered thresholds
Turning a key in a keyhole	Misfit	
Switching on a bedside lamp		Disordered thresholds
Turning on a television set	Discrimination < the average	Disordered thresholds
Opening the fridge door	Misfit	Disordered thresholds
Communication		
Turning over the pages of a comic book	Misfit	Disordered thresholds
Opening a mail	Discrimination > the average	Disordered thresholds
Inserting a diskette into a drive	Discrimination < the average	Disordered thresholds
Using a keypad phone	Discrimination < the average	Disordered thresholds
Do-it-yourself		
Colouring	Discrimination > the average	Misfit
Painting	Discrimination > the average	Disordered thresholds
Drawing a line with a slat	Discrimination > the average	
Sharpening a pencil		Disordered thresholds
Handling a rubber	Discrimination > the average	Disordered thresholds
Cutting a sheet with scissors by following a line	Discrimination > the average	Discrimination > the average
Handling a punch	Misfit	Disordered thresholds
Handling an adhesive tape	Discrimination > the average	
Leisure and play		
Throwing a ball	Discrimination > the average	Disordered thresholds
Catching a ball	Discrimination > the average	Discrimination > the average
Piling up some Lego blocks	Discrimination > the average	Disordered thresholds
Throwing a dice	Discrimination < the average	Disordered thresholds
Dealing cards	Misfit	Misfit
Handling a joystick	Misfit	Misfit
Miscellaneous		
Grasping a coin on a table	Discrimination > the average	Misfit
Taking a coin out of a pocket		Disordered thresholds
Putting on a backpack/schoolbag		Disordered thresholds
Opening a cardoor	Discrimination < the average	Disordered thresholds
Closing a cardoor	Disordered thresholds	Disordered thresholds
New items added following the experts' suggestions		
Putting a coin into a piggy bank	Discrimination < the average	Disordered thresholds
Putting a key into a keyhole	Misfit	
Hanging up a jacket	Misfit	Disordered thresholds
Fastening one's seat belt	Discrimination < the average	Disordered thresholds
Rolling up a sleeve of a sweater		Disordered thresholds

* The 21 items of the final verison of the ABILHAND-Kids questionnaire are in bold type.
** Items were deleted from the parents' and children's responses.

Appendix IV. *ABILHAND-Kids questionnaire*

Instructions for the ABILHAND-Kids questionnaire

The ABILHAND-Kids questionnaire

The ABILHAND-Kids questionnaire was developed as a measure of manual ability in a sample of children with Cerebral Palsy (*Neurology 2004; 63:1045-52*). It explores the most representative inventory of manual activities. Some items derived from the ABILHAND questionnaire, a manual ability scale developed for adult patients (*Arch Phys Med Rehabil 1998; 79:1038-42*) (*Stroke 2001; 32:1627-34*). Other items were selected from existing scales or were devised to extend the range of activities. The parents reported a finer perception of their children's manual ability than the children themselves, leading to a wider range of measurement, a higher reliability (R = 0.94) and a good reproducibility over time (R = 0.91). ABILHAND-Kids was therefore exclusively built on the parents' perceptions. The 21 items of ABILHAND-Kids defined a valid and reliable manual ability scale. ABILHAND-Kids was originally developed using the Rasch measurement model. It allows to convert ordinal scores into linear measures located on a unidimensional scale.

Procedures

Parents are asked to fill in the questionnaire by estimating their child's ease or difficulty in performing each activity, when the activities are done:

- Without other technical or human help (even if the child actually uses help in daily life);
- Irrespective of the limb(s) actually used to do the activity;
- Whatever the strategy used (any compensation is allowed).

Parents are asked to provide their perceived child's difficulty on a three-level scale: "Impossible", "Difficult", or "Easy". Activities not attempted in the last 3 months are not scored and are entered as missing responses (tick the question mark). For any activity the four potential answers are:

- ***Impossible***: the child is unable to perform the activity without using any other help;
- ***Difficult***: the child is able to perform the activity without any help but experiences some difficulty;
- ***Easy***: the child is able to perform the activity without any help and experiences no difficulty;
- ***Question mark***: the parents cannot estimate the difficulty of the activity for their child because he/she has never done the activity. However, if the activity was never attempted because it is impossible, then it must be scored as "Impossible" rather than "Question mark".

The instructions are given to the parents only at the beginning of the test. Five items are used for training in order to help the parents in feeling each level of the rating scale and in using the whole amplitude of the response scale.

Activities order

The activities of the ABILHAND-Kids questionnaire are presented in a random order to avoid any systematic effect. Ten different random orders of presentation are used. The examiner must select the next one of the 10 orders for each new assessment, no matter which child is tested.

Website

The ten orders of the ABILHAND-Kids questionnaire can be downloaded from www.abilhand.org (accessed January 04, 2006) in English, French, and Dutch. The website also allows raw total scores to the ABILHAND-Kids questionnaire to be converted into linear measures of manual ability, according to the Rasch model.

ABILHAND-Kids - Manual Ability Measure
English version (order 1)

Patient ______________________________ **Date** _____________

	How **DIFFICULT** are the following activities?	**Impossible**	**Difficult**	**Easy**	**?**
1.	Opening a jar of jam				
2.	Putting on a backpack/schoolbag				
3.	Opening the cap of a toothpaste tube				
4.	Unwrapping a chocolate bar				
5.	Washing the upper-body				
6.	Rolling-up a sleeve of a sweater				
7.	Sharpening a pencil				
8.	Taking off a T-shirt				
9.	Squeezing toothpaste onto a toothbrush				
10.	Opening a bread box				
11.	Unscrewing a bottle cap				
12.	Zipping-up trousers				
13.	Buttoning up a shirt/sweater				
14.	Filling a glass with water				
15.	Switching on a bedside lamp				
16.	Putting on a hat				
17.	Fastening the snap of a jacket				
18.	Buttoning up trousers				
19.	Opening a bag of chips				
20.	Zipping-up a jacket				
21.	Taking a coin out of a pocket				

Appendix V.
ABILHAND questionnaire

Instructions for the ABILHAND questionnaire

The ABILHAND questionnaire

The ABILHAND questionnaire was developed as a measure of manual ability as perceived by the patient. It explores the most representative inventory of manual activities. Some items were selected from existing scales; others were devised to extend the range of activities. The first application of the questionnaire in a sample of rheumatoid arthritis patients (*Arch Phys Med Rehabil 1998; 79: 1038-42*) showed that the items defined a valid manual ability scale. A second application of the questionnaire in a larger sample of chronic stroke patients showed that the unimanual activities (usually realized with one hand) were too easy for the patients. So, a subset of 23 bimanual activities (usually realized with two hands) has been retained and calibrated for chronic stroke patients (*Stroke 2001; 32: 1627-34*). ABILHAND was originally developed using the Rasch measurement model. It allows to convert ordinal scores into linear measures located on a unidimensional scale.

Procedures

The ABILHAND questionnaire is administered on an interview basis (patients do not realize the activities). Patients are asked to estimate the ease or difficulty in performing each activity, when the activities are done:

- Without other technical or human help (even if the patient actually uses help in daily life);
- Irrespective of the limb(s) actually used to do the activity;
- Whatever the strategy used (any compensation is allowed).

During the evaluation, a 3-level response scale is presented to the patients. Patients are asked to rate their perception on the response scale as either "Impossible", "Difficult" or "Easy". Activities not attempted in the last 3 months are not scored and are entered as missing responses (tick the question mark). For any activity the four potential answers are:

- ***Impossible***: the patient is unable to perform the activity without using any other help;
- ***Difficult***: the patient is able to perform the activity without any help but experiences some difficulty;
- ***Easy***: the patient is able to perform the activity without any help and experiences no difficulty;
- ***Question mark***: the patient cannot estimate the difficulty of the activity because he/she has never done the activity. Note that when a patient has never attempted the activity, the rater needs to make sure why it is so. If an activity was never attempted because it is impossible, then it must be scored as "Impossible" rather than "Question mark".

The instructions are given to the patient only at the beginning of the test. Five items are used for training in order to help the patient in feeling each level of the rating scale and in using the whole amplitude of the response scale. The subsequent activities are neither preceded nor followed by any instruction. The examiner can repeat the instructions whenever the patient shows some hesitation in answering.

Activities order

The activities of the ABILHAND questionnaire are presented in a random order to avoid any systematic effect. Ten different random orders of presentation are used. The examiner must select the next one of the 10 orders for each new assessment, no matter which patient is tested.

Website

The ten orders of the ABILHAND questionnaire can be downloaded from www.abilhand.org (accessed January 04, 2006) in English, French, Dutch, Italian, and Swedish. The website also allows raw total scores to the ABILHAND questionnaire to be converted into linear measures of manual ability, according to the Rasch model.

ABILHAND - Manual Ability Measure
English version (order 1)

Patient ______________________________ **Date** ______________

	How **DIFFICULT** are the following activities?	**Impossible**	**Difficult**	**Easy**	**?**
1.	Pulling up the zipper of trousers				
2.	Peeling onions				
3.	Sharpening a pencil				
4.	Taking the cap off a bottle				
5.	Filing one's nails				
6.	Peeling potatoes with a knife				
7.	Buttoning up trousers				
8.	Opening a screw-topped jar				
9.	Cutting one's nails				
10.	Tearing open a pack of chips				
11.	Unwrapping a chocolate bar				
12.	Hammering a nail				
13.	Spreading butter on a slice of bread				
14.	Washing one's hands				
15.	Buttoning up a shirt				
16.	Threading a needle				
17.	Cutting meat				
18.	Wrapping up gifts				
19.	Fastening the zipper of a jacket				
20.	Fastening a snap (jacket, bag, ...)				
21.	Shelling hazel nuts				
22.	Opening mail				
23.	Squeezing toothpaste on a toothbrush				