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Examensarbete masternivå, 30 högskolepoäng
Vårterminen 2014

Missing data in physiotherapists' assessments of children with cerebral palsy

Mätbortfall vid fysioterapeuters bedömningar av barn med cerebral pares

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Abstract
Aims: The aim of this study was to investigate the extent and pattern of internal missing data in physiotherapy assessments of children with cerebral palsy (CP), regarding passive range of motion (PROM) and spasticity (MAS). *Methodology:* Data from 2.936 children were included. Extent and pattern of missing data of PROM and MAS were related to Gross Motor Function Classification System (GMFCS) level, CP subtype, age and gender. The material was divided into groups and the proportions of the different variables were compared. Chi squared statistics were used to assess differences in proportions. *Major findings:* The extent of missing data of PROM and MAS was small (6.2%). The frequency of missing data of MAS was larger than for PROM. The frequency of missing data varied according to GMFCS level and age. *Principal conclusion:* The extent of missing data in this study can be considered small and thus would not influence the generalizability of findings for PROM and MAS. It is however advisable to report the degree of missing data especially if data concern passive range of motion and spasticity from children GMFCS level I, GMFCS level III, children from age group 1 – 5 years and adolescents aged 13 – 20 years.

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Sammanfattning

Syfte: Studiens syfte var att undersöka omfattning och mönster av internt bortfall i sjukgymnastbedömningar av barn med cerebral pares (CP), gällande passiv ledrörlighet (PROM) och spasticitet (MAS).*Metod:* Data från 2.936 barn inkluderades. Omfattning och mönster av internt bortfall av PROM och MAS relaterades till Gross Motor Function Classification System (GMFCS) nivå, subtyp av CP, ålder och kön. Materialet delades upp i grupper och proportionerna av de olika variablerna jämfördes. Chi2-test användes för att bedöma proportionella skillnader. *Resultat:* Omfattningen av internt bortfall av mätvärden för PROM och MAS var liten (6.2%). Frekvensen av internt bortfall av mätvärden för MAS var högre än för PROM. Frekvensen av bortfall varierade beroende på GMFCS nivå och ålder. *Konklusion:* Omfattningen av internt bortfall kan anses vara liten i denna studie och torde inte påverka generaliserbarheten av resultaten för PROM och MAS. Dock bör graden av internt bortfall rapporteras vid studier, speciellt för resultat vad gäller passiv ledrörlighet och spasticitet för barn från GMFCS nivå I, GMFCS nivå III, barn 1-5 år och ungdomar 13-20 år.

What is known on this topic

- Children with CP have an increased risk of musculoskeletal problems, e.g. hip dislocation, scoliosis and muscle contractures.

- Surveillance programs have proven beneficial to prevent these secondary complications.

What this paper adds

- The amount of missing data regarding passive range of motion and spasticity in physiotherapy assessments in the National Quality Registry, CPUP, was small (6.2%), indicating that this data give valid information for children/adolescents with CP.

- The percentage of missing data for assessments of spasticity was higher than the percentage of missing data for measuring of passive range of motion, implying difficulties for physiotherapists to assess spasticity.

- The frequency of missing data varied according to GFMCS level and age.

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Background

Cerebral palsy

Cerebral palsy is an umbrella term covering a heterogenous group with permanent brain lesions which always encompasses motor disorders. The lesions affect the immature brain, during gestation or before 2 years of age. The disorders are permanent but not unchanging (Mutch, 1992; SCPE, 2000). The prevalence in the western world is 2 – 3 per 1000 live births (Himmelmann, 2006; Nordmark, 2001a; Westbom, 2007). The prevalence is higher the more pre-term the children are born and the lower the birth-weight is (Krägeloh-Mann, 2009; Himmelmann, 2006) . The motor disability varies greatly between individuals according to the lesion´s size and localization. The motor function ranges from independent walking to severe limitations of voluntary movements. The prevalence of comorbidities, such as epilepsy, visual and hearing impairments, communication disorders and learning disabilities increase with severity of gross motor function disability (Himmelmann, 2006; Nordmark, 2001b; Shevell, 2009).

Definitions

Throughout the years there have been several attempts to agree on a definition of CP. One of the earliest was the one used by Bax in 1964 *"a disorder of posture and movement due to a defect or lesion in the immature brain"* (Bax, 1964). In 1992 a group of researchers amended the definition *"an umbrella term covering a group of 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 most recent definition was presented by Rosenbaum et al in 2007 *"Cerebral palsy describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems"* (Rosenbaum, 2007).

Classifications

Cerebral palsy is classified into subtypes according to the dominating motor symptoms. The subtypes are spastic, dyskinetic, ataxic and unclassified. Children with spastic CP have increased tone and pathological reflexes. In cases of dyskinetic CP, involuntary, uncontrolled, recurring and stereotyped movements are present. Children with ataxic CP, present decreased muscular coordination, so that movements are performed with abnormal force, rhytm and accuracy. A group of children has mixed forms of CP, i.e. a combination of motor symptoms, for example spasticity and ataxia. If no motor symptom is clearly dominating, the children are registered as "unclassifiable" (Cans, 2006) The most common subtype is spastic followed by dyskinetic, ataxic and unclassifiable (Nordmark, 2001a; SCPE, 2002; Soo, 2006; Westbom, 2007). Interrater reliability for classification of subtype of CP between pediatricians is generally good, except for selection between spastic and dyskinetic subtypes (Sellier, 2012; Gainsborough, 2008) The distribution of the motor symptoms can further be described as unilateral (affecting one side of the body) or bilateral (affecting both sides of the body) (SCPE, 2000).

The subtype of cerebral palsy is often combined with a classification of the gross motor function. The Gross Motor Function Classification System (GMFCS) is an ordinal scale with five levels of motor performance. The different levels represent clinically meaningful distinctions in motor function. Level I contains the children that are most independent. They

can walk without restrictions, but have limitations in more advanced gross motor skills. Children at Level II walk without assistive devices, but have some limitations when they are walking outdoors and in the community. Children at Level III walk with assisitive devices, but have limitations when walking outdoors and in the community. Children at Level IV have self-mobility with limitations. They are transported or use power mobility outdoors and in the community. Children classified as Level V have the most severe motor disability. Their self-mobility is limited, even with the use of assistive technology. The classification is based on the usual motor performance of the child/adolescent rather than on motor capacity (Palisano, 1997; Rosenbaum, 2008). The GMFCS was initally developed for children from the age of 2 to 12 years. In 2007 Palisano et al developed GMFCS expanded and revised, GMFCS-E & R (Palisano, 2007). They added an age-band, 12 – 18 years and revised the age-band 6 – 12 years.The GMFCS-E & R covers five age-bands – before 2 years, 2 – 4 years, 4 – 6 years, 6 – 12 years and 12 – 18 years. The criteria of the different GMFCS levels take into account the change of motor ability with age. Thus a child on GMFCS Level II who at the age of 6 years walks independently without any assistive device, might at the age of 14 choose to use a wheelchair for longer transportations, but the child is still classified as Level II (Palisano, 2008). Palisano and co-workers agreed that it is useful to make a distinction between gross motor capacity and gross motor performance, i.e. what the child has the capacity to do and what the child actually does in their everyday life. The GMFCS-E & R takes into account that the gross motor performance is also influenced by the attitudinal environment and personal factors, such as interests and motivation. With this conclusion Palisano and co-workers (Palisano, 2008) found that GMFCS-E & R can coincide with the International Classification of Functioning, Disability and Health (ICF) (WHO, 2001). GMFCS-E & R has demonstrated good validity and inter-rater reliability (Palisano, 2008; Mutlu, 2011). The agreement between the GMFCS and GMFCS-E & R is excellent. The results imply that GMFCS-E & R can be used when comparing longitudinal data with GMFCS (Gudmundsson, 2013).

The group of patients with the diagnosis CP is heterogenuos, therefore it can be hard to compare different studies of patients with CP. To describe which patients are included in studies of cerebral palsy, several authors have stated the importance of using the classification of subtype of CP as well as the classification of gross motor function, GMFCS (Himmelmann, 2006; Hägglund, 2007; Shevell, 2009).

Musculoskeletal problems

Children with CP have motor problems, expressed as impaired selective muscle control, muscular weakness, impaired postural control and coordination (Bache, 2003; Dodd, 2010). Furthermore, individuals with CP may develop musculoskeletal problems, such as muscle/tendon contractures, bony torsion, hip displacement and scoliosis. These problems may occur throughout life and possible causes are muscle spasticity, muscle growth and ageing (Rosenbaum, 2007; Hilberink, 2007; Morrell, 2002). Spasticity is one symptom of CP as a sequelae of the central brain lesion. The definition of spasticity that is most often used is *"a velocity-dependent increase in tonic stretch reflexes with exaggerated tendon jerks, resulting from hyperexcitability of the stretch reflex"* (Lance, 1980) Children with CP have more limited range of motion in the lower extremity compared to a control group of typically developed children (Kilgour, 2005). Passive range of motion in the lower extremities decline with age (Nordmark, 2009; Hägglund, 2011; Hilberink, 2007).

Hip displacement can occur at an early age (Dobson, 2002; Hägglund G, 2007; Morton, 2006; Scrutton, 2001). The risk for hip displacement or hip dislocation increases with a higher GMFCS level. Individuals with GMFCS level V have the highest risk of hip dislocation. The subtype of CP also effects the risk of hip dislocation. The highest risk for children with bilateral, spastic CP and lowest risk for children with unilateral, spastic CP or ataxic CP (Hägglund, 2007; Soo, 2006).

Surveillance program

From the hypothesis that complications as hip displacement or hip dislocation can be prevented, surveillance programs for children with CP were started. They have proven beneficial (Hägglund, 2005; NICE, 2012; Elkamil, 2011; Dobson, 2002; Morton, 2006). Initially it may be hard to be certain of the diagnosis cerebral palsy. Some motor disabilities may be transient anomalies in preterm babies or they may develop into a progressive condition. Milder cases of CP, such as unilateral spastic or ataxic CP may not be obvious initially (Cans, 2006). Therefore it is important that the registers follow the individual throughout the years. The definite diagnosis CP should not be set until the age of 4 – 5 years (Mutch, 1992; SCPE, 2000; Zarrinkalam, 2010).

Although CP is the most common motor disability for children, it is still rare; 2 – 3 per 1000 live births. Since the diagnosis of CP is a heterogenous condition, large populations are needed for sufficient numbers of participants to make research possible, particularly when studying children from different subgroups of CP (SCPE, 2000). To meet these issues, a network of CP surveys and registers was formed across Europe – Surveillance of Cerebral Palsy in Europe, SCPE. This network can help to monitor trends in the CP rate, provide a framework for collaborative research and give information to form common guidelines for healthcare programs for CP (SCPE, 2000).

CPUP – national quality register

CPUP is a national follow up program for people with cerebral palsy in Sweden. It was initiated in the southern parts of Sweden in 1994 (counties of Skåne and Blekinge). It was formed in a collaboration between the Habilitation centres of the counties and the Department of Orthopaedics at Lund University Hospital. The aim was to prevent hip dislocation, scoliosis and severe contractures in children with CP by early detection and intervention (Hägglund, 2005). Further aims are to enhance collaboration between different professionals and to increase knowledge about CP (www.cpup.se). The CPUP has proven beneficial in reducing the incidence of hip dislocation. The incidence in Sweden, where CPUP was used, was <1% compared to 15% in Norway (before CPUP was introduced there)(Elkamil, 2011).

In 2005 CPUP was accepted as a National Quality Register by The National Board of Health and Welfare. From the year of 2007 all counties of Sweden are participating in the CPUP programme (Arner, 2007). Since 2009 CPUP for adults has been introduced in some regions of Sweden. In Norway CPUP is a national healthcare program since 2009. The program has also been initiated in Denmark, Iceland, Scotland and New South Wales, Australia (www.cpup.se).

The CPUP program includes a standardized follow up with assessment of fine and gross motor function, spasticity, joint range of motion, radiographics of hips and spine, clinical findings and treatment (www.cpup.se). As soon as there are signs of cerebral palsy the parents

of the child are invited to take part of CPUP as a follow up program. Due to the changing clinical picture in young children with motor disorders, the CP diagnosis is not set by the neuropediatrician until the age of four (SCPE, 2000). If the assessment, by the age of four, shows that the child does not fulfill the criteria for CP, the child´s data are deleted from the CPUP register (www.cpup.se).

Quality registers

In the Swedish health and medical services, several national quality registers exist.
A national quality register contains individual-based data concerning patient problems or diagnoses, medical interventions and outcomes after treatment. The aim of the quality registers is to constitute a knowledge system that can be used by all levels of healthcare providers for continuous learning and quality improvement. The information from the registers are also used for general planning and management of healthcare services.
From 2007 the Swedish Association of Local Authorities and Regions (SALAR) are primary responsible for the continuing operation, development and financing of the registers.
The registers are being developed and managed by representatives of the professional groups that use them (SKL, 2007).

Missing data

Missing data can be a problem in clinical research. The extent and pattern of missing data must be analyzed to be able to draw correct conclusions from the results (El-Masri, 2005; Kneipp, 2001; Newgard, 2006). The aims of the quality registers are to work for continuous learning and quality improvement, therefore data from quality registers must be as complete as possible to enhance good research conditions (Landsting, 2010).

Statistical literature and articles describe methods to handle missing data in multi-variable research. The first step is to analyze the extent and pattern of missing data (Little, 2002; Johansson, 2013; El-Masri, 2005; Haukoos, 2007). Cohen and Cohen suggest that up to 10% of missing data on a variable is not large (Cohen J., 2003). If the extent is large, different methods of handling the missingness are present. One method is to exclude all cases that have missing data on some variable and make a "complete-case analysis". Another method is to use imputation. You replace each missing value with a value that is approximate to the missing value , for example the mean of values from the complete assessments (Kneipp, 2001; Haukoos, 2007). One disadvantage of "complete-case analysis" is that it excludes participants that vary in a systematic way from those with missing values (Kneipp, 2001). Analysis of the pattern of missing data can give us important information (Little, 2002). Despite these different techniques, the most effective way to handle missing data is to prevent it from occurring (El-Masri, 2005).

Aim of study

The aim of this study was to investigate the extent and pattern of internal missing data in physiotherapy assessments in the National Quality Register CPUP, regarding passive range of motion and spasticity of lower extremity

Materials and methods

Study design

The study was a retrospective register analysis of extent and pattern of internal missing data.

Inclusion
All children/adolescents registered in the CPUP register in Sweden from January 2009 to December 2012. The most recent physiotherapist assessment during the period was analysed. During this period data from 2.936 children were registered. 57% boys and 42% girls. 35% were age 1 – 5 years, 50% were age 6 – 12 years and 15% were 13 – 20 years. The distribution between the different GMFCS levels was - GMFCS I 44%, GMFCS II 15%, GMFCS III 9%, GMFCS IV 14% and GMFCS V 16%. The distribution of subtype was – spastic 76%, dyskinetic 11%, ataxic 4% and unclassified 7% (Table 1 in article)

Procedure
Data from both legs were analysed. Thus 5.872 values from each individual could be analysed from each muscle and joint angle that was assessed. In the CPUP physiotherapist assessment the passive range of motion (PROM) is measured for nine different joint angles/leg. The spasticity is assessed according to modified Ashworth scale (MAS), for six different muscle groups/leg.

Passive range of motion (PROM)
In CPUP, measurements of PROM are done with a goniometer in a stated and standardized way, described in a manual accompanying the recording form (for manual see www.cpup.se) Validity of measurements of range of motion with goniometer shows that it is a reasonable and simple method for clinical measurements on healthy subjects (Chapleau, 2011). The concurrent validity with other methods, such as radiographic measurements and digital inclinometer has shown that those methods should be used when a higher level of precision is required (Roach, 2013). The reliability of measuring PROM with a goniometer has been tested. Interrater reliability varies from low to high, intrarater reliability is higher, and varies from high to excellent (Herrero, 2011; Mutlu, 2007). McDowell and co-workers stated that results from goniometer measurements should be used with caution for clinical decision making. Especially for joints that are effected by biarticular muscles such as the hamstrings muscles (McDowell, 2000). Decreased PROM can be a sign of loss of motor function (Pinero, 2012).

Modified Ashworth scale (MAS)
In CPUP, assessment of spasticity is done according to modified Ashworth scale. The MAS is a 6 grade, ordinal scale, 0, 1, +1, 2, 3 or 4. “0” signifies normal tonus and “4” total rigidity. The lower or upper limb is moved through the range of movement with the velocity of 180◦/s. The movement goes from the position of relative muscle shortening to a position of relative muscle lengthening. Degree of resistance and under which part of the range of movement the resistance can be felt, is estimated (Bohannon, 1987).

Reliability and validity have been tested for MAS. The interrater reliability varies from poor to good in different studies (Clopton, 2005; Fosang, 2003; Mutlu, 2008). It seems to differ depending on which muscles are tested. Intrarater reliability varies from moderate to good (Clopton, 2005). Test-retest variability varies from poor to good (Fosang, 2003; Mutlu, 2008).

Statistical analysis
All data were treated confidentially and subject identification was coded before analyses.

Descriptive statistics were used for the proportions of the variables GMFCS level, CP subtype, age and gender in the study group, and for the frequencies of missing data for the measurements of passive range of motion (PROM) and assessments of spasticity (MAS).

Chi squared statistics were used for assessing differences in proportions between groups. This statistical analysis can be used to compare proportions between groups when the data is nominal and/or ordinal (Ejlertsson, 2003). When the amount of individuals is too small ($n \leq 5$), Fisher´s exact test is considered appropriate to use. In the present study, Fisher´s exact test was used when comparing the proportions of the different CP-subtypes in the group with $\leq 1/3$ of values missing for PROM and MAS respectively, with the group that were missing $>1/3$ of the values (Table 5). P-values ≤ 0.05 were considered significant. SPSS version 18.0 (SPSS Inc., Chicago, IL, USA) was used for the statistical analyses.

Ethical considerations

As soon as there are signs of cerebral palsy for a child, the parents get information about CPUP and they get an invitation for the child to participate in the follow up program.
The parents get information, both written and oral, and they are informed that their decision to participate or not, does not affect the child´s continued treatment at the habilitation center. The data of the register is used for the local habilitation center to plan future treatment for the child. The data may be used for research purpose, but then unidentified. The parents, or later the adolescent himself, can at any time decide to withdraw their participation in CPUP. The data of earlier assessments is then deleted from the register.
Each region has to apply for the persons who can have access to the register to both read and add new data about the child. The authorization is given from the person who is responsible for the quality register in Lund, Sweden. These authorizations are given for a limited time. It has to be renewed on a regular basis. Each region also has a coordinator who is responsible for follow up of the persons who get authorization.

One of the aims for CPUP is to perform research about individuals with CP. Each authorized person have access to data from the region where they work. To get authorization for access to data from the whole register, the researcher must make an application to a publication committee at CPUP, with a description of the intended future research. The researcher has to describe the research questions and how the proposed studies can bring new knowledge about CP. If the application is accepted the reseracher gets access to data from the whole register for a certain time-period.

Studies have shown that the CPUP follow up program can prevent hip dislocation and severe contractures for children with cerebral palsy (Hägglund,2005; Elkamil, 2011).
Therefore the gain for each individual, participating in the program, can make an important difference during the lifespan for an individual with cerebral palsy.

In this study, data registered from the physiotherapist assessments in the CPUP register was analyzed. The children, or families did not have to do any extra assessments or take time to fill in any surveys.

Results

6.2% of the total number of possible values were missing in the physiotherapy assessments, regarding measurements of passive range of motion (PROM) and assessments of spasticity (MAS). There was a significant difference of the percentage of missing values of PROM (6.0% of total possible values) versus the percentage of missing values of MAS (6.6% of the total possible values) ($p<0.0001$). The highest percentage of missing data of PROM occurred for Ely´s test, 12.7%. (Table 2 in the article) For MAS the highest percentage of missing data occurred for assessments of the hip flexors and hip extensors, 7.0% for both muscle groups. (Table 3 in the article)

Comparison was done between the group of children/adolescents that had complete assessments and the group of children/adolescents that had some missing data in their physiotherapy assessments. Children GMFCS V ($p=0.048$) and children from age group 1 – 5 years ($p<0.0001$) were significantly more common in the group that had missing data (Table 4 in the article).

Further analyses of the group of children with missing data of PROM and/or MAS were done by dividing them into two groups that were either missing $\leq 1/3$ of the values or $>1/3$ of the values. The comparison showed that children GMFCS level I and children from age group 1 – 5 years were more common in the group of children/adolescents that were missing $>1/3$ of the values of PROM. A similar comparison of missing data of MAS showed that children from GMFCS level III and adolescents from age group 13 – 20 years were more common in the group that were missing $>1/3$ of the values of MAS (Table 5 in the article).

Discussion

This study was a retrospective register analysis of data from the National Quality Register, CPUP. The extent and pattern of internal missing data in physiotherapy assessments, regarding passive range of motion and spasticity were analysed.
One strength of register-based research is that data already exists, which makes data collection faster and also gives the possibility to do follow-up studies over time. The registers make it possible to include all persons in a defined population and this enables research with large sample sizes. In the future an interesting progress would be if researchers, to a much larger extent, could get access to use data from registers from different countries in studies (Olsen, 2011; Thygesen, 2013). Limitations of register-based research are that data collection is not done by the researcher, the researcher is limited to use the variables that are included in the registers and it can be hard for the researcher to control the quality of the the data (Thygesen, 2013).

In the present study, one strength is that a well-defined population of children and adolescents with CP in Sweden was included. This enables a large study-group and an extensive amount of data. Furthermore the assessments of the participants of the study had already taken place so the children/adolescents in the study did not have to spend any extra time for collection of data.

Limitations of the study were that we did not investigate why data was missing. Different explanations can be plausible; it can depend on the physiotherapist, the child/adolescents or even on the caregivers that accompany the child. It was only the extent and pattern of missing data that were analyzed, the possible relations between the variables PROM and MAS were not investigated.

The extent of missing data were relatively small in the present study, as well as in the earlier studies based on data from CPUP that have described their extent of missing data (Rodby-Bosquet, 2010a; Rodby-Bosquet, 2010b; Rodby-Bosquet, 2012). This indicates that data from the National Quality Register CPUP can be considered to provide valid information about children with cerebral palsy.

Acknowledgements

I would like to express my gratitude to:
Eva Brogren Carlberg, my supervisor for invaluable support, guidance and your positive encouragement always given when needed the most

Lars Söderström, for unestimable help with statistical analysis

County council of Jämtland, department of research and development, for financial support

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Appendix Manuscript for European Journal of Physiotherapy

Title page not for review

Missing data in physiotherapists´ assessments of children with cerebral palsy

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Web-adress till artikel i European Journal of Physiotherapy:
doi:10.3109/21679169.2015.1009162