Predicting the development of communication skills by children with motor disorders

Dr Lindsay Pennington, Dr Mark Pearce, Professor Allan Colver,
Professor Helen McConachie, Newcastle University
Dr Mary Gibson, Newcastle upon Tyne Hospitals NHS Trust

Background

Children with motor disorders, such as cerebral palsy (CP) often have difficulties developing speech, language and gestural communication. Parents' first question, when referred to speech and language therapy (SLT), is "will my child speak?" But we have little evidence to help answer this question. This study will show which children develop intelligible speech and which may benefit from the early introduction of augmentative and alternative communication systems to ensure the development of independent, active communication, thereby facilitating participation in family, social and educational life.

Cohort studies have shown that speech disorders are associated with all types of CP. Difficulties are more often reported in children with dyskinetic than spastic types of CP (Bax *et al.*, 2006; Odding *et al.*, 2006). In children with spastic type CP difficulties are more frequently observed in children with wider distributions of motor impairment (Bax *et al.*, 2006; Odding *et al.*, 2006). However, associations between speech disorder and motor impairment distribution are not entirely linear nor are they clear cut.

Although current research estimates the prevalence of speech disorder in different types of CP it cannot be used to predict children's speech intelligibility. Severity of speech disorder is rated in only one study to date (Kennes *et al.*, 2002), several studies conflate speech, language and communication (Wiklund & Uvebrant, 1991; Lepage *et al.*, 1998; Bax *et al.*, 2006) and all are single observations of children in middle childhood.

To be able to predict children's speech development longitudinal studies are needed. The study proposed here is a cohort study which will follow children from two to five years of age. Pre-pilot studies have shown that separate assessment of children's functioning is needed, as: SLTs assess children's development according to need; similar batteries of assessments are not used for children with different disorders; and informal assessments are used, which cannot be replicated reliably. A recently completed pilot study has determined the measures and procedures to be used in the proposed cohort study, testing these for the youngest children to ensure commitment

from families for longitudinal follow-up and informing the sample size for the main study and the required duration of the research.

The study aims to show if characteristics of children with CP at 2.00-2.06 years (years.months), whose speech is giving cause for concern, can predict the children who at 5 years will have:

- intelligible speech that is appropriate for their developmental age; or
- disordered speech, which is nevertheless intelligible to familiar adults and strangers; or
- severely disordered speech which requires augmentative and alternative communication systems to ensure the development of independent, active communication.

METHODS

Participants: All children with non-progressive motor disorders (many of whom will already have had a diagnosis of CP) residing in North of England (North of England Children's Cerebral Palsy Survey (NECCPS) area) born between 1.11.2006 and 31.10.08, whose communication is giving cause for parental and/or clinician concern at age 2.0-2.06 years. As some children may only receive a diagnosis of cerebral palsy at three to four years of age we will include all children who have a non-progressive motor disorder at two years. Many of these may have received a diagnosis of CP. Some children will not have received a diagnosis. All children's difficulties will be referred to as motor disorders, unless parents use the term 'cerebral palsy'.

Children will be excluded only if they have a progressive motor disorder, if they have a diagnosis other than CP or if their parent/caregiver and a clinician with whom they have regular contact have no concerns about the child's speech or communication. prevalence of communication disorders in older children with CP, prevalence data from the NECCPS and data from the pilot study, it is estimated that 60 children will be eligible to join the study for each of two birth years, and that 50 will be wiling to participate. Including children from the pilot study will give an expected sample size of 127. With this sample size, and allowing for 27 children to be lost to follow-up, it is estimated that the study will have in excess of 90% power to detect at least a 0.8 standard deviation difference in speech production test (DEAP) scores at age 5 between the highest and lowest tertiles of DEAP scores at age 3. It should also be noted that using continuous measures of early performance will result in even greater statistical power.

Study design: prospective cohort study

Measures

Independent measures

1. Corrected age: months
2. Birth weight: grams
3. Site, type and extent of neurological lesions: findings from MRI

4. Type and distribution of motor impairment: Surveillance of Cerebral Palsy in Europe classification
5. Gross motor skills: Gross Motor Function Classification System (GMFCS) (Palisano *et al.*, 1997) - a validated, reliable method of rating the independent mobility and gross motor function of children with CP, which uses a five point ordinal scale to classify children's current function.
6. Upper limb function: Manual Ability Classification System for Children with Cerebral Palsy (Eliasson *et al.*, 2006) - developed to rate function of children with motor disorders, has validity and reliability data.
7. Vision and hearing acuity
8. Language development: Pre-School Language Scales UK edition (PLS3) (Boucher & Lewis, 1997) - can be completed without adaptation by children who can grasp and release objects, easily adapted for children with more severely impaired upper limb function and used in previous research involving children with motor disorders (Pennington et al, in press). Raw scores will be used for all children, as the adaptation will nullify the use of standardised scores.
9. Cognition: Mullen Scales of Early Learning, (visual receptive scale) (Mullen, 1995) for children who do not show understanding of sentences containing two information carrying words on the PLS3. Raw scores will be used. Children understanding at least two words will be assessed using the Leiter-R Scales (Roid & Miller, 1997).
10. Oro-motor skills: Oral Speech Motor Control Protocol (Robbins & Klee, 1985)- standardised on children from 2years, for children who are able to imitate some oral movements.
11. Diagnostic speech assessment: DEAP (Dodd *et al.*, 2002) diagnoses type of speech disorder (e.g. motor speech disorder, phonological disorder).
12. Parent's perceptions of communication and speech intelligibility: Children's communication questionnaire (see attached)
13. Spoken vocabulary: MacArthur Communicative Development Inventory UK edition (MCDI) (Klee *et al.*, 1999), which asks parents to select from vocabulary lists the words their child can produce.

Dependent measures

1. Classification of children's communication according to the 4 point system used by the NECCPS (1= no problem, 2 = some delay/disorder but use spoken language, 3 = uses ACC or should have AAC system, 4 = no formal method of communication).
2. Communication Function Classification Scale. % point classification of children's ability to express their messages and respond to others' communication (Heidecker et al, 2008)

Procedures

A positive opinion from relevant ethics committees and R&D approval will be in place before children are recruited to the study.

Sampling
SLTs, physiotherapists and consultant paediatricians from all NHS trusts in the NECCPS area will be asked to identify all eligible children residing in their Trust. Strong collaborative networks have been developed with local therapists and NECCPS coordinators through previous research. Local clinicians support this application. The networks for sampling, referral and recruitment were developed as part of the pilot study and processes are working well.

Recruitment
Short information sheet will be given by the SLT or physiotherapist to children's parents/caregivers informing them about the aims of the research and giving a brief description of the study. If parents do not opt out at that stage, their telephone numbers will be passed to the research team. The team will telephone parents, at least 14 days after their receipt the introductory letter**,** explain the aims of the research and discuss what is involved. If families wish to join the project they will be visited at home on up to two occasions at the following ages: two years, three years and five years. In the first visit for the research parents will be asked to sign a Parent Information Sheet and Consent Form.

Data collection
In the home visits the following measures will be conducted: Mullen Visual Reception Scales/ Leiter-R, Pre-School Language Scales and Oral Motor Control Protocol. Parents will be asked to complete Children's Communication Questionnaire. Parents of children showing intentional communication and understanding of spoken language from a single word level (on the PLS3) will be asked to complete the MCDI UK. Children producing at least 20 recognisable words (as reported on the MCDI UK) will be assessed using the DEAP.

One of the research team will read the child's medical notes to obtain data on chronological age, birth weight, vision, hearing, type and distribution of motor impairment, and results of MRI scans (where available). The child's regular physiotherapist will classify the child's gross motor function and upper limb according to the GMFCS and MACS respectively.

Data from all measures will be entered onto an Access database with a unique identifier assigned to each record to allow patient information details to be stored separately from the dataset to be used in the analysis.

Children will be visited at two, three and five years of age. Prior to these visits the local teams will be contacted by the researchers to ensure that researchers are aware of relevant issues; for example, ill health of children, parents, house moves, child death. If parents request the results of the assessments they will be given these verbally be the researcher conducting the data collection.

Once a year the families participating in the study will be sent a newsletter which will update them on the progress of the research, giving the numbers of children who are participating and any groups results currently available from the study. At the end of the study each family will receive a written summary of the results from the study. Individual children's results will not be supplied within reports.

Analysis

Continuous outcome measures at age 5 years such as DEAP will be analysed using linear regression, with suitable transformations if appropriate. Outcomes such as 5 point severity rating scales will be analysed using logistic regression. All regression models will be assessed for goodness of fit and to ensure adherence to statistical assumptions. As speech tests are only standardized from 3 years of age regression using speech test data will undertaken using data from children at 3 and 5 years. For other developmental domains (e.g. receptive and expressive language, cognition), regression analysis will include data from children at 2 and 5 years.

References

Bax M, Tydeman C & Flodmark O (2006) Clinical and MRI Correlates of Cerebral Palsy: The European Cerebral Palsy Study. *JAMA* **296**, 1602-1608.

Boucher J & Lewis V (1997) *Preschool Language Scales-3 (UK)*. London: The Psychological Corporation.

Dodd B, Hua Z, Crosbie S, Holm A & Ozanne A (2002) *Diagnostic Evaluation of Articulation and Phonolgy*. London: The Psychological Corporation.

Eliasson A, Krumlinde-Sundholm L, Roblad B, Beckung E, Arner M, Ohrvall A & Rosenbaum P (2006) The Manual Classification System (MACS) for children with cerebral palsy: scale development and evidence of validity and reliability. *Developmental Medicine & Child Neurology* **48**, 549-554.

Hidecker, M. J. C., Paneth, N, Rosenbaum, P., Kent, R. D., Lillie, J., Johnson, B., & Chester, K. (2008, March). Developing the Communication Function Classification System (CFCS). Poster session presented at a Cerebral Palsy Research conference, Ann Arbor, Michigan.

Kennes J, Rosenbaum P, Hanna SE, Walter S, Russell D, Raina P, Bartlett D & Galuppi B (2002) Health-status of school aged children with cerebral palsy: information from a population-based sample. *Developmental Medicine & Child Neurology* **44**, 240-247.

Klee T, Marr C, Robertson E & Harrison C (1999) *The MacArthur Communicative Development Inventory: Toddler. British English Adaptation*. Newcastle: Newcastle University.

Lepage Cl, Noreau L, Bernard P-M & Fougeyrollas P (1998) Profile of hndicap situations inchildren with cerebral palsy. *Scandinavian Journal of Rehabilitation Medicine* **30**, 263-272.

Mullen EM (1995) *Mullen Scales of Early Learning*. Circle Pines, MN: American Guidance Service, Inc.

Odding E, Roebroeck M & Stam H (2006) The epidemiology of cerebral palsy: Incidence, impairments and risk factors. *Disability & Rehabilitation* **28**, 183-191.

Palisano RJ, Rosenbaum P, Walter S, Russell D, Wood E & Galuppi B (1997) Development and reliability of a system to classify gross motor function in children with cerebral palsy. *Developmental Medicine & Child Neurology* **39**, 214-223.

Robbins J & Klee T (1985) Clinical assessment of oropharyngeal motor development in young children. *Journal of Speech & Hearing Disorders* **52**, 271-277.

Roid GH & Miller LJ (1997) *Leiter-R*. Wood Dale, IL: Stoelting.

Wiklund LM & Uvebrant P (1991) Hemiplegic cerebral palsy: correlation between CT morphology and clinical findings. *Developmental Medicine & Child Neurology* **33**, 512-523.