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Pilot feasibility study of a digital technology approach to the systematic electronic capture of parent-reported data on cognitive and language development in children aged 2 years

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ABSTRACT

Background The assessment of language and cognition in children at risk of impaired neurodevelopment following neonatal care is a UK standard of care but there is no national, systematic approach for obtaining these data. To overcome these challenges, we developed and evaluated a digital version of a validated parent questionnaire to assess cognitive and language development at age 2 years, the Parent Report of Children's Abilities-Revised (PARCA-R).

Methods We involved clinicians and parents of babies born very preterm who received care in north-west London neonatal units. We developed a digital version of the PARCA-R questionnaire using standard software. Following informed consent, parents received automated notifications and an invitation to complete the questionnaire on a mobile phone, tablet or computer when their child approached the appropriate age window. Parents could save and print a copy of the results. We evaluated ease of use, parent acceptability, consent for data sharing through integration into a research database and making results available to the clinical team.

Results Clinical staff approached the parents of 41 infants; 38 completed the e-registration form and 30 signed the e-consent. The digital version of the PARCA-R was completed by the parents of 21 of 23 children who reached the appropriate age window. Clinicians and parents found the system easy to use. Only one parent declined permission to integrate data into the National Neonatal Research Database for approved secondary purposes.

Discussion This electronic data collection system and associated automated processes enabled efficient systematic capture of data on language and cognitive development in high-risk children, suitable for national delivery at scale.

WHAT IS ALREADY KNOWN ON THIS TOPIC

⇒ Though a UK standard of care for at-risk infants admitted for neonatal care, the assessment of language and cognitive development at age 2 years is not systematically undertaken, nor are results available for secondary purposes.

WHAT THIS STUDY ADDS

⇒ We show that a digital approach for obtaining consent, sending automated notifications and capturing parent-reported data on a child's cognitive and language development at the age of 2 years is acceptable to clinical teams and parents, and feasible for national delivery at scale. Parents also found acceptable the incorporation of results into a national database for approved secondary purposes.

HOW THIS STUDY MIGHT AFFECT RESEARCH, PRACTICE OR POLICY

⇒ The development of this digital approach to data collection provides an efficient solution to capture national data at scale and reduce the costs of obtaining data on cognitive and language development at the age of 2 years for clinical care, randomised trials, health services research and policy development.

INTRODUCTION

The assessment of cognitive and language development in children born very preterm and/or at risk of impaired neurodevelopment following admission to neonatal care is a UK standard of care. The National Institute for Health and Care Excellence (NICE) recommends that babies born below 30 weeks' gestation, or at 30–36 weeks' gestation with additional risk factors for developmental problems and disorders, should be followed up to 2 years of age.[1–3] These data are also essential for follow-up evaluations of babies who participate in obstetric and neonatal studies, for which neurodevelopmental impairment at 2 years of age is frequently a key outcome.[4] In addition, neurodevelopmental

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outcomes comprise components of the core outcomes set for neonatal medicine, which clinicians, parents and researchers consider all clinical trials should record.[5]

In the UK, there is currently no national, systematic approach for assessing and recording the results of assessments of cognitive and language development in children born very preterm and other infants who are at risk of impaired neurodevelopment. Among many challenges are the need to keep in touch with families, the inconvenience of having to attend clinic appointments, the requirement for appropriately trained staff and the costs of follow-up assessments.

The present situation is that many children do not receive these assessments, and some receive separate assessments for clinical and research purposes. Additionally, many neonatal studies lack this important information because of the difficulty in securing funding for long-term follow-up. The lack of a uniform system also results in wide variation in the types of developmental assessments undertaken, making it difficult to harmonise and interpret data at a population level. When funding for research evaluations is available, children and their families often experience the burden of multiple separate assessments for clinical care and research, as there are no systematic processes for sharing data obtained as part of clinical practice for research and vice versa.

The Parent Report of Children's Abilities-Revised (PARCA-R) is a standardised, norm-referenced, parent-completed questionnaire that can be used to assess cognitive and language development at 23–27 months of age and identify children with developmental delay.[6 7] It is recommended by NICE as a clinical tool to assess the development of children born preterm[1–3] and by The International Consortium for Health Outcomes Measurement for assessing development at 2 years of age for all children who were born preterm or hospitalised in the neonatal period.[8] It is also widely used as an outcome measure in observational studies and clinical trials.[9–15]

For follow-up to be effective, parent participation must be high, and the data require secure storage and well-governanced processes for access. Here, we describe the development and refinement of a systematic approach to developmental follow-up using a digital version of the parent-completed PARCA-R and processes to incorporate the results into a mature well established, UK Health Research Authority approved national asset, the National Neonatal Research Database (NNRD)[16 17] so that they can be used for approved secondary purposes with parent agreement.

METHODS

Aims and setting

The aim of this service improvement study was to develop and test a systematic approach to administering the PARCA-R questionnaire, obtaining parent consent and responses, storing the results in the NNRD and making them available to parents and the child's clinical team. We involved all west-London National Health Service neonatal units (Chelsea and Westminster; West Middlesex; Queen Charlotte's and Chelsea; St Mary's; Northwick Park; Hillingdon).

Study design

We developed the study in collaboration with parents; the British Association of Perinatal Medicine; British Association of Neonatal Neurodevelopmental Follow-Up; Bliss, the national charity for babies born preterm or sick; and clinicians from north-west London neonatal units. We held three focus groups to engage and involve parents (five women and one man) and obtain their views during the design of the study.[18]

Patient recruitment, sites and duration

We recruited parents of babies born less than 30^{+0} weeks of gestation, either at the time of neonatal unit discharge, or after discharge and prior to the 2-year follow-up outpatient visit. The study had a total duration of 1 year with a recruitment period of 6 months.

Technology, governance and storage

The Imperial Clinical Trials Unit Clinical Data Systems team, in collaboration with neonatal clinicians, employed standard software (OpenClinica V.4) to develop a digital technology application to enable parents to complete the PARCA-R electronically. Prototype e-forms were refined in-house taking into consideration feedback from clinicians and parents. The OpenClinica application included electronic consent forms completed by parents providing their permission to receive automated reminders and use personal identifiers to link PARCA-R data into the NNRD. In addition to facilitating electronic data capture, the OpenClinica V.4 software can generate reports, study metrics and functions in accordance with Standard Operating Processes. The OpenClinica V.4 platform is compliant with all necessary regulations (21 Code of Federal Regulations Part 11; Good Clinical Practice; General Data Protection Requirement; Health Insurance Portability and Insurance Act Annex 11) and is an all in one, cloud-hosted system, applying zero data loss architecture for failure resilience and recovery to the very last transaction with a 4-week backup data retention period. Personal identifiers were collected to facilitate completion of the questionnaire and reporting of results to parents (figure 1). These were stored securely and separately from all other study data. Data were encrypted at rest and in transit to ensure the highest level of data protection.

Study processes

At the time of discharge from the neonatal unit, or when approaching the 2-year follow-up clinic visit, a member of the neonatal clinical team approached parents to invite their participation (figure 1). The clinician explained the purpose of the study, provided written information (online supplemental file 1), sought e-consent to participate (online supplemental file 2), registered parent

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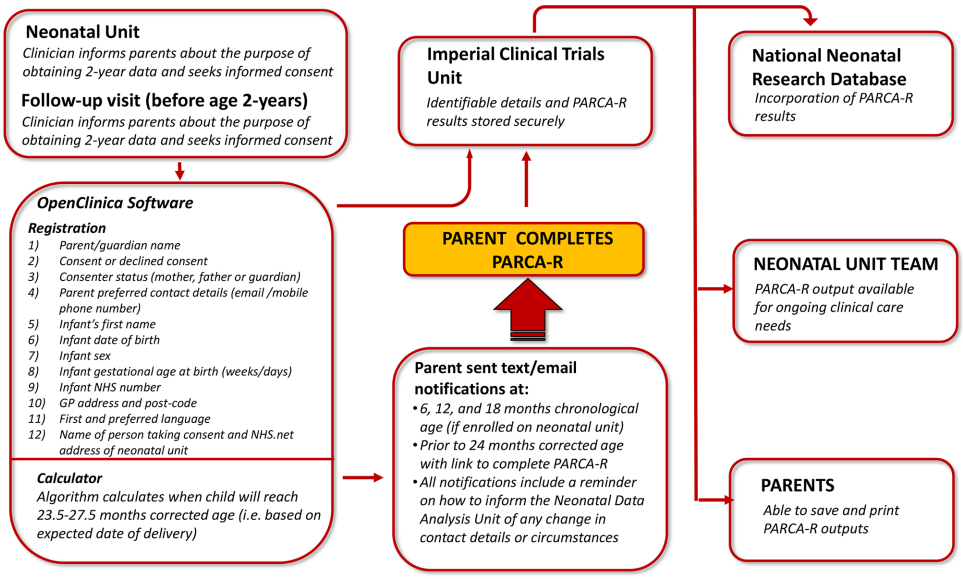

Figure 1 Data flows for families enrolled on the neonatal unit or after neonatal unit discharge. PARCA-R, Parent Report of Children's Abilities-Revised. NHS: National Health Service

contact details and completed an e-registration form (online supplemental file 3). Parents who agreed to participate received automated notifications when their child reached 6, 12 and 18 months chronological age. These reminded the parent to inform the Neonatal Data Analysis Unit at Imperial College London where the NNRD is maintained, of any change in contact details or circumstances. When approaching 24-months corrected age (online supplemental file 4), parents were sent an invitation to complete the PARCA-R electronically on a tablet, laptop, desktop computer or mobile phone when their child reached the age of 23.5–27.5 months age corrected for prematurity. On completion of the PARCA-R questionnaire, parents were able to save and print a copy of their child's results (online supplemental file 5). During the registration process, clinicians offered parents a choice to receive notifications by email, by text or both. We randomly allocated parents who selected both methods to receive notifications by either email or email plus text.

Access to personal identifiers was only available to an authorised member of the research team. We used personal identifiers and contact details with parent consent, only for the following purposes: (1) confirm eligibility, (2) contact parents, (3) personalise communications, (4) calculate the child's age, (5) link the PARCA-R score to the infant's NNRD record and (6) send without-obligation notifications about studies and links to information that might be of interest to the parents or importance to the child.

The PARCA-R results were stored pseudonymised with the child's record in the NNRD. In addition, we sent a copy by Secure File Transfer Protocol to a specific NHS.net address so that each neonatal unit participating in this study could incorporate the results into the child's clinical record.

We undertook descriptive analysis to examine consent, uptake and the proportion of missing data for all participants who opened a PARCA-R questionnaire.

RESULTS

The clinical neonatal teams of north-west London NHS Trusts approached the parents of 41 children: 10 in London North West University Healthcare NHS Trust; 6 in Imperial College Healthcare NHS Trust; 8 in Chelsea & Westminster Hospital NHS Foundation Trust and 17 in Hillingdon Hospitals NHS Foundation Trust. For unknown reasons, 3 out of the 41 parents did not complete the registration form. Out of the 38 participants who completed the registration form, 16 children were girls and 22 were boys. Their gestational ages at birth ranged from 24 to 29 weeks. Twenty-three parents selected English as their first language, 14 selected other languages (Romanian, Turkish, Gujarati, Italian, Arabic, Mandarin, Albanian, Polish and Punjabi) and one was unsure. Of the 38 parents who completed the registration form, 30 completed and 1 started but did not complete the e-agreement (figure 2).

Out of the 41 parents who were invited to participate in the study, 34 were happy to be contacted by text or email and 7 only by text. Of the 34 that agreed to be contacted either way, 19 were randomly allocated to receive notifications by email only and 15 by text and email. Of the ones receiving notifications by email only, 13 (68%) signed the e-consent compared with 11 (73%) in the text plus email group. This difference became more marked when comparing questionnaire completion with 10 (53%) vs 11 (73%) in each group, respectively.

By the end of the study, 30 out of the 38 (79%) participants who signed the registration form went on to sign the e-consent. Of these, only one participant did not agree to

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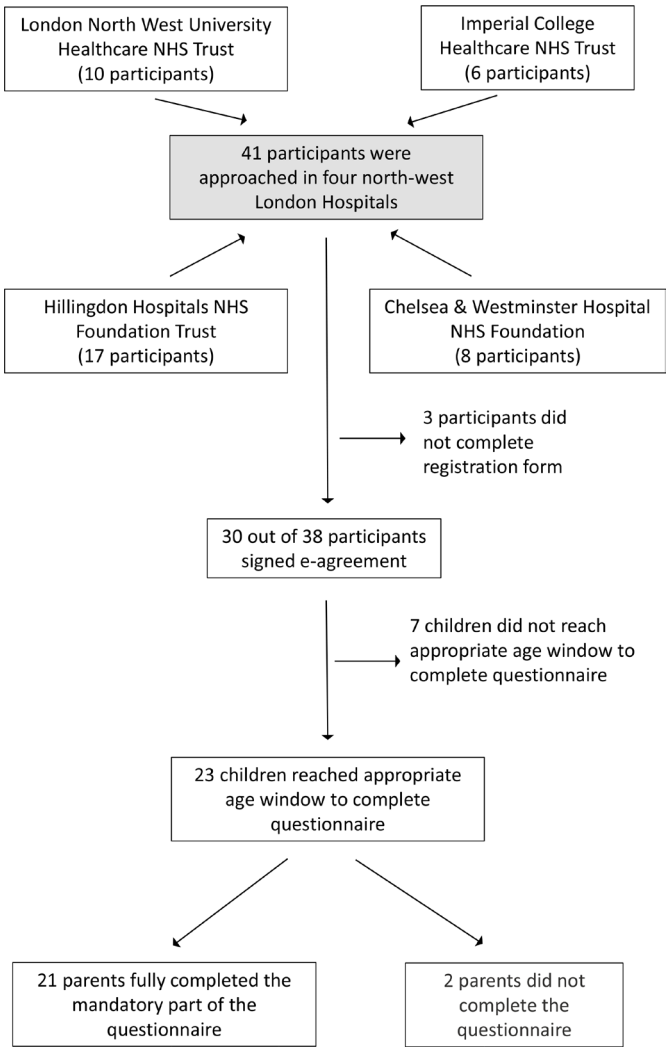

Figure 2 Numbers of participants.NHS: National Health Service

have their baby's data shared with the clinical team and to be integrated into the NNRD and only two did not agree to be contacted about future studies. Twenty-three of the 30 children reached the appropriate age window for completion of the PARCA-R questionnaire. For these, 21 parents (91%) completed it fully and correctly. Two (9%) were completed incorrectly, one because of erroneous enrolment after the child's second birthday and one because the questionnaire was completed early, before the child was aged 23.5 months, due to a mistake by the clinician when entering the schedule of events.

This digital version of the PARCA-R questionnaire was completed well and it was possible to calculate standardised scores for all 21 children.

Following parent and clinician feedback, we refined the electronic processes. We included a link to the parent information leaflet within the e-agreement form so that parents could access this at any time. We limited the number of characters per notification to reduce the cost of text notifications. We modified standard OpenClinica terminology by replacing the word 'subject' with 'participant' and referred to 'your child', instead of 'this child'. We resolved technical issues with the display of the PARCA-R scores using iOS in iPhones and a randomisation glitch. We introduced multifactor authentication to access the questionnaire. We modified the NHS number field within the registration form from numeric to text. Finally, we made programming changes to remove the capacity for a clinician to modify the schedule of events inadvertently.

DISCUSSION

We successfully developed, piloted and refined an electronic system to obtain parent-reported data on cognitive and language development at 2 years of age in children born very preterm. The system was easy to use and acceptable to clinicians and parents. Parents were able to access information about the process and its purpose at any time and were able to complete the questionnaire on a choice of devices. They could choose if they wished to give permission for the questionnaire results to be incorporated into the NNRD, to be made available for other approved uses. They were also able to choose if they wished to receive future information that might be relevant to their child. Importantly, the results of the PARCA-R questionnaire were available to the clinical teams for use in routine developmental follow-up care and parents were able to save and print out a copy of their child's results.

We were able to provide parents with notifications by email and text. Despite a slight increase in cost, this is a helpful option as some parents had no access to email. Overall, completion was higher in the group receiving dual notifications, rather than email alone. A current limitation is that while the PARCA-R questionnaire has been translated into 23 different languages (https://le.ac.uk/parca-r/translations), only a small proportion of those translations have been validated to ensure the appropriate selection of words in a culturally appropriate content. This is an important consideration for future development, as 40% of the participants in our study did not consider English their primary language.

The study period overlapped with the COVID-19 pandemic and the alterations to service provision may have affected uptake. We are unable to say whether this was to enhance or decrease uptake.

CONCLUSIONS

The success of this study suggests that electronic collection of parent-reported data on cognitive and language development for children born very preterm is feasible and suitable for national scale-up. It could improve the clinical care of individual children as well as the efficiency of clinical trials and other studies and reduce the costs of obtaining data on cognitive and language development at the age of 2 years. The incorporation of the results into the NNRD, thereby making these data available to other researchers for approved purposes would mean sparing children and their families the burden of repeated

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assessments. It would also enable systematic exploration of outcomes in complete populations of children. Finally, use of a single measure at 2 years of age would obviate the need to harmonise data from multiple developmental tests.

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