A COMPARISON OF OBJECTIVE, STANDARDISED PARENT-ADMINISTERED QUESTIONNAIRES TO THAT OF SUBJECTIVE SCREENING PRACTICES FOR THE EARLY DETECTION OF DEVELOPMENTAL DELAY IN AT-RISK INFANTS

Melinda Lee E Silva

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2 Declaration

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I, Melinda Lee E Silva declare that this research report is my own work. It is being submitted for the degree of Master of Science in Medicine (Child Health and Neurodevelopment) in the University of the Witwatersrand, Johannesburg. It has not been submitted before for any degree or examination at this or any other University.

Melinda Lee E Silva

28th day of January 2010

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3 Dedications

To Ricky and Daniel, for putting up with my absences, my distraction from your lives and for making sure I kept to my Christmas deadline.

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5 Abstract

Background

The early identification of developmental disorders facilitates early intervention, improving childhood outcomes. Parent-administered questionnaires have been recommended for this purpose. The PEDS COMBINED, which includes the Parents Evaluation of Developmental Status (PEDS) and PEDS Developmental Milestones (PEDS:DM), and the Ages and Stages Questionnaire (ASQ) are parent-administered questionnaires with good psychometric properties. It has not been determined if they identify the same infants at-risk for developmental delays however. It is also not know how South African paediatricians monitor childhood development.

Objective

To compare the ASQ, PEDS COMBINED and South African paediatricians' subjective assessment (PSA) of neonatal intensive care graduates at 6-months corrected age.

To identify trends in developmental screening practices, including the knowledge and use of parent-administered screening tools, in a diverse group of paediatricans working in Gauteng, South Africa.

Methods

Developmental screening and referral practices of paediatricians were ascertained by analysis of a short questionnaire sent to participating paediatricians.

Concordance between the questionnaires and PSA was determined using the kappa coefficient (κ) and Test of Symmetry ($\kappa \leq 0.4$ indicating poor agreement; $\kappa \geq 0.75$ indicating excellent agreement).

Results

Concordance between the ASQ and PEDS COMBINED was 90.7% ($\kappa = 0.82$, $\rho = 0.05$). The PSA showed poor concordance with both PEDS COMBINED and ASQ ($\kappa = 0.28$, $\rho = 0.03$ and $\kappa = 0.26$, $\rho = 0.01$ respectively). The ASQ and PEDS COMBINED identified 40% and 42% of the cohort with developmental concerns and the PSA identified 6%. Only 13% of paediatricians used specific guidelines for developmental assessment and none used, or could name any parent-administered questionnaire.

Conclusion

There is excellent agreement between the ASQ and PEDS COMBINED in identifying the same children with developmental concerns. Paediatricians identified significantly fewer infants and showed statistically significant poor agreement with both questionnaires. Most paediatricians in Gauteng, South Africa are not using standardized screening tools to detect developmental delay and have poor knowledge regarding these tools.

Key Words

Developmental screening; Developmental delay; PEDS; PEDS:DM; PEDS COMBINED ;ASQ; NICU infant; paediatrician; South Africa.

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9 List of Abbreviations

Abbreviation	Description
6 mnth CA date	Six month corrected age date
AAP	American Academy of Pediatrics
ABCD	Assuring Better Child Health and Development
ASQ	Ages and Stages Questionnaire
BSID-II	Bayley Scales of Infant and Toddler Development – II
Comm	Communication
EI	early intervention
ELBW	Extremely low-birth weight infants
FM	Fine Motor
FT	Full-term
GM	Gross Motor
GMFM	Gross Motor Functional Measure
HIV	Human immunodeficiency virus
HLBW	high low-birth-weight
ICF-CY	International Classification of Function – Child and Youth
IEP	Individualized Education Plan
IHDP	Infant Health and Development Program
IQ	Intelligent Quotient
LAMI	Low and Middle Income
LBW	Low-birth weight
LLBW	low low-birth-weight
LPT	Late pre-term
NGST	neuronal group selection theories
NICU	Neonatal Intensive Care Unit
PEDS	Parent's Evaluation of Developmental Status
PEDS:DM	Parent's Evaluation of Developmental Status:Developmental Milestones
PEDS COMBINED	PEDS + PEDS:DM
PS	Problem Solving

PSA	Paediatrician's subjective assessment
QoL	Quality of Life
ROP	Retinopathy of prematurity
SD	Standard Deviation
SE	Social-Emotional
SES	Socioeconomic status
TB	Tuberculosis
USA	United States of America
VLBW	Very low-birth-weight
VPT	Very preterm
κ	Kappa co-efficient

10 Literature Review

10.1 Outcomes of the At-Risk Infant

The immature nervous system is continuously influenced by an interaction between biology and the environment and as a result the developing infant is vulnerable to the negative impact of adverse experiences (1). These adverse influences can include social risk factors (such as poverty, lower parental education and maternal depression) as well as medical risk factors (such as premature birth, lung disease and post-natal infections). Infants who experience social or medical risk factors are called high-risk or at-risk infants as they are at greater risk for poor neurological, cognitive, social and behavioral outcomes than infants who do not experience these risk factors. Social and medical risk factors are individually responsible for adverse developmental outcomes, although they are interdependent with compounding effect. (1, 2, 3) This review concentrates on the medical risk factors experienced by infants requiring neonatal intensive care in the perinatal period that result in neurological impairment or dysfunction.

Alyward & Verhulst (4) describe a typical high-risk nursery as one containing primarily preterm infants as well as full-term infants who have experienced perinatal complications. The literature categorises infants in the neonatal intensive care unit (NICU) as either higher risk or lower risk (Table 1) depending on their gestational age, birth weight or primary morbidity. (5, 6)

Table 1 Classification of Risk in NICU infants

Classification	Infants born…	Description	Abbreviation
Lower risk	between 34 – 37 weeks gestation	Late pre-term	LPT
	after 37 weeks	Full-term	FT
	weighing 1500g - 2500g	Low-birth weight	LBW
Higher risk	before 34 weeks gestation	Very preterm	VPT
	weighing less than 1000g.	Extremely low-birth weight infants	ELBW
	weighing less than 1500g	Very low birth-weight	VLBW

10.1.1 Significant Neurological Impairments

The risk of serious neurological morbidities such as intraventricular haemorrhage and periventricular leucomalacia increases with decreasing gestational age and birth weight. They are a significant cause of cerebral palsy and other major neurodevelopmental impairments in the NICU infant. Chronic lung disease, infection and necrotizing enterocolitis are also known predictors of adverse neurological outcomes, along with male gender and exposure to postnatal steroids. (3, 7)

Retinopathy of prematurity and cortical visual impairment are noted to be the most common causes of severe visual impairment and blindness in the preterm infant, with amblyopia and refractive errors also being common. The auditory system in the preterm infant can be affected by ototoxic drugs and NICU infants are therefore also at risk for conductive hearing loss and auditory neuropathies (8).

In their review of extremely premature infants Robertson, Watt and Dinu (8) noted improved medical care had resulted in a decreasing incidence of these major neurological impairments over the past 30 years with the exception of intellectual impairment. The prevalence of cerebral palsy decreased from 77/1000 extremely preterm births in the 1990's to as low as 19/1000 births by the early 2000's. By the early 2000's blindness and hearing loss as a result of premature birth had also decreased to less than 1% of survivors.

Major neurological impairments are an important consideration in NICU infants but inline with their decreasing incidence, literature has started to look at more subtle outcomes of developmental delay in NICU infants such as learning disabilities, behavioral problems and quality of life (QoL). (9, 10, 11)

10.1.2 Developmental Delay

Normal development proceeds from basic skills to integrated functions involving higher cognitive process such as problem solving and planning. Initial skills are species specific and "prewired" into the neurological system resulting in easily identifiable milestones typically involved in early infant screening and assessment. These

milestones are divided into the domains of motor (gross and fine), language (expressive and receptive), personal-social (behavior, social, emotional) and adaptive (cognitive, problem solving) development. (12, 13)

When an infant or child does not reach a milestone in one or more of these domains at the expected age (within the range of normal variation) they are described as developmentally delayed (14). Developmental delay is most often determined by a percentage (e.g. 20% delay), a standard deviation (SD) below the mean in a reference group (normally 2SD) or a comparison of milestones to the 50th percentile of population groups (12). Developmental delay normally refers to milder delays resulting in coordination difficulties, academic difficulties, behavioural problems or social-emotional disturbances negatively affecting the quality of life of the children and their families. Health related QoL measures take into account the World Health Organizations' definition of health, which looks at an individual's physical, mental and social well-being, rather than merely the absence of disease (15).

10.1.2.1 The NICU infant

The NICU infant has been shown to be at a greater risk for developmental delay and poorer health related QoL than infants born full term, regardless of their gestational age, birth-weight or diagnosis (10, 15, 16). Compared to full-term controls, pre-school aged children who required neonatal intensive care needed more help for activities of daily living such as eating, bathing, dressing and toileting. Even in children with no co-morbidities that required NICU care, difficulty getting around was four times greater and problems with learning and memory were three times greater than children who did not require NICU care (15).

Lower-risk NICU infants are still significantly more likely to require early intervention or special education than infants born without medical risk (10, 17, 18). A large study looking at 7187 NICU infants who were lower-risk (due to a birth weight greater than 1500g) found that compared to full-term healthy infants, these infants were significantly more likely to have special health care needs (requiring medication, using more health services and

having an activity limitation). These infants were also significantly more likely to have a learning disability or Attention Disorder (6).

NICU infants who are lower-risk due to their full-term status also show more difficulties with learning, memory and motor development compared to children who did not require intensive care at birth (19). Full-term NICU infants have been shown to deviate from full-term healthy children in motor performance and social interactions by their first year; and speech and linguistic skills by 6 ½ years. In a 10-year follow-up study of NICU infants, although 94% of NICU graduates were able to attend mainstream schooling, more than 40% required some sort of educational support such as speech therapy, a special education teacher or a special teaching group outside of the regular classroom. Children who were very preterm (VPT) were more likely to be a year behind their expected grade, require educational support or attend special educational facilities than the children that were late preterm (LPT) or full-term NICU graduates. (16, 20)

Kalia et.al also found VPT infants required more intervention than LPT infants with 30% of LPT infants qualifying for enrollment in a physiotherapy, speech therapy or occupational therapy programme compared to 70% of VPT infants. After controlling for co-morbidities (which correlated inversely with gestational age) infants in either group were equally likely to qualify for intervention services however, indicating a significant risk for developmental difficulties in lower-risk NICU infants and highlighting the negative impact of co-morbidities (21).

NICU infants of all gestational ages and birth-weights are clearly a population group at risk for developmental difficulties. The morbidities experienced in the newborn period are the most significant cause of developmental difficulties requiring Early Intervention (EI), rather than the degree of prematurity, however VPT babies are likely to have more problems and experience worse outcomes (21). Accordingly, most of the literature on outcomes of the NICU infant is concerned with the higher-risk NICU infant, those born at less than 34 weeks or weighing less than 1500g. The literature on this specific population group is therefore discussed in the following section.

10.1.2.2 The Higher Risk NICU Infant

The “modern era” of neonatal intensive care beginning in the 1960’s has resulted in increasing survival of low birth weight (LBW) and extremely low birth weight (ELBW) infants with significant reductions in major morbidities. Outcomes of ELBW infants from the EPICure study show that although developmental disabilities were present in at least half of all survivors, only a quarter of these met the criteria for severe disability (22). Along with the World Health Organization’s International Classification of Function for Children and Youths (ICF-CY) and increasing interest in QoL, research on these high risk premature infants has shifted from identifying the disability towards identifying the more subtle developmental delays with their long-term implications for activity limitations and participation in the context of the individual and society. (23, 24, 25)

Studies following very low-birth-weight (VLBW) infants into adolescence show higher rates of neurodevelopmental disabilities compared to their full-term peers, despite normal intelligence. This results in poorer academic and social functioning and higher educational needs (26). Clinically, preterm infants show significantly lower reading, mathematics and spelling scores than their full-term peers (9). In addition studies following these infants into adulthood show similar trends of lower levels of academic achievement and lower salaries, increasing the burden of support on tax payers and society as a whole. (27, 28) These long-term outcomes of preterm infants are found cross-culturally and within many different countries (9).

Some studies of QoL and outcomes in teenagers and adults who were born VPT or VLBW have shown these infants achieve similar educational attainment, socioeconomic status and marital status compared to their full-term peers. Parents of these teenagers and adults often noted significantly poorer performance in global health, behaviour and physical functioning however; with more cognitive, mobility and self-care limitations. It is therefore important when looking at the QoL and long-term outcomes of VPT or VLBW adults to also look at the therapy they received in their early years, as well as their specific schooling requirements or schooling patterns. This may assist in a better understanding of the interventions needed to achieve more positive outcomes for these higher risk infants. (11, 29, 30)

Reuner, Hassenpflug and Pietz (10) attributed the positive outcomes seen in their LBW population at 17 years of age to the fact that their study group was from a tertiary neonatal centre where preterm infants received an intensive follow-up and early intervention (EI) programme. Even though outcomes were positive, the LBW children in their study tended to start school later than their full-term peers, with a trend towards lower school graduation. Therapeutic intervention such as occupational therapy, speech therapy and physiotherapy was required by 72% of the LBW children, compared to 51% of their full term peers and LBW children often required two or even three different interventions. Similarly, in an outcome study of 12-year-old children born prematurely, 76% with a diagnosed brain injury and 35% without a diagnosed brain injury at birth needed an Individualized Education Plan (IEP) compared to 10% of children born at full-term (26). These studies show how the pathway to more positive outcomes in preterm children may differ significantly from their full-term peers, with an increased need for therapeutic intervention and delayed school enrollment placing a greater burden on preterm children and their families.

A recent meta-analysis by Aarnoudse-Moens, Weisglas-Kuperus and van Goudoever (9) in August 2009 suggest that the disadvantage in academic achievement, behaviour and neurocognitive function persists into young adulthood (22.3 years of age) for very preterm and very low birth weight (VLBW) infants and highlights the need to focus on early intervention strategies to achieve more positive outcomes.

10.2 Early Intervention

Early Intervention is described as a multidisciplinary approach to enhance emerging abilities and prevent delays in children from birth to 5 years of age. It includes the remediation of existing or emerging disabilities and the prevention of functional deterioration. Adaptive parenting and overall family function is an important aspect of EI, as is promoting the child's health and wellbeing. EI occurs through individualized developmental, educational and therapeutic services using techniques derived from the disciplines of physiotherapy, occupational therapy, developmental psychology and education (31).

There is considerable empirical evidence that various types of EI promote child development, reduce gaps in school readiness, and improve outcomes in later life. Learning and skill formation build on previous learning, therefore school readiness is critical for academic achievement. A child's health status and academic achievement are co-dependent outcomes of EI. Less education is associated with a shorter life span, earlier onset of chronic disease or disability and increased risk of poor health related behaviors such as smoking, teenage pregnancies and crime; whilst poor health may be associated with lower academic achievement. (32, 33, 34)

Neural plasticity and the neuronal group selection theories (NGST) can be used to motivate for intervention to be started as early as possible. Neural plasticity refers to change in function of existing neurons and synapses in the brain and is most active in the first 8 months of life. NGST is the reprogramming and reorganizing of neural tissue and their networks into functional units through variable experiences and repetition. Before 3 years of age, NGST suggests that intervention through variable experiences could increase the primary repertoire of neuronal networks and enhance selectivity. In the older child, networks are already established and intervention will be less effective in changing them. Neural plasticity and neuronal group selection can result in both negative and positive adaptive behavior depending on the experience of the infant. EI is therefore important not only in remediating difficulties, but in preventing deviant development. (35)

The Dynamic Systems Theory of development looks at development in relation to the interaction of multiple subsystems within the child and the environment. Systems within the child such as muscle strength and motivation interact and are changed by systems within the environment such as experience and the demand of a task (35, 36). The Dynamic Systems Theory supports EI by acknowledging that small, critical changes in one subsystem (such improving trunk control) can result in large changes in developmental abilities (such as improved ability to play) (37).

In the context of neural plasticity, NGST and the Dynamic Systems Approach, EI should be most effective when started as early as possible. This is supported by studies that have shown improved effect of intervention started earlier, with some interventions aiming to start before 6 months of age. (38, 39, 47)

When intervention is begun early it is more effective than remediation in addressing developmental delay, as the rate of learning for remediation must exceed the rate of learning of those children who are performing on age-level. Children who are delayed must not only "catch-up" to their peers, but also "keep-up" with them as they learn new learn skills. This highlights the importance of EI in ensuring children start school with developmentally appropriate skills (32).

The development of competence is highly related to the development of emotional regulation, social skills and intelligence. Lack of competence creates negative experiences with far reaching impact for those children who are not assisted in developing skills for which they have difficulty. The provision of high-quality EI with appropriately skilled staff can produce short-term gains in individual development as well as long-term human capital gains. Investing in effective services that promote child development well before the start of school and facilitate full access to early intervention is therefore optimal (1, 40).

10.2.1 Effectiveness of Early Intervention

The effectiveness of intervention generally is supported by studies which look at specific goals in children such as an occupational therapy programme to improve upper limb function (41), speech therapy to address language delay (42) or gross motor programmes to improve physical activity and proficiency (43).

The effectiveness of EI in at-risk infants is supported by a substantial amount of literature. Cochrane reviews and meta-analyses have looked at intervention programmes for prematurely born infants that begin before the first 12 months of life (44, 45). The literature shows a significantly positive effect for this type of early intervention, particularly when the parent-child relationship is emphasized. Interventions addressing both the

parent-child interaction, as well as the child's specific developmental issues were more effective than programmes that addressed only one of these components. A higher level of intensity or duration of early intervention programmes also resulted in more sustained positive effects and better outcomes. Other positive factors contributing to the success of early intervention were the early start of the programme, professionally trained staff and adequate staff-child ratios. (1, 32)

One of the largest and most comprehensive EI studies cited in the systematic reviews on the long-term benefits of EI is the Infant Health and Development Program (IHDP). Since so many studies examining the effect of EI for premature infants from infancy to 18 years of age are based on this programme (45), it is described in the following section in more detail.

10.2.1.1 IHDP

IHDP was a multi-centre EI programme for low birth weight, premature infants, with long-term follow up. The Promising Practices Network (46) described the IHDP study as using rigorous standards, including a randomized experimental design and longitudinal follow-up. McCormick, Brooks-Gunn and Buka (33) published the 18 year outcomes of this study and highlighted the fact that whereas most studies that document the educational and social benefits of early intervention programmes focus on lower socioeconomic groups, this study looked at a group of low birth weight infants who were otherwise heterogeneous for socioeconomic and health status. The intervention programme consisted of 3 parts: home visits by a professional to implement a general developmental programme and target specific difficulties identified; a centre-based education programme for infants from 12 months to 36 months (5 days/week for 4 hours each day) and a bimonthly parent support group. It is important to note that infants in the control group who did not receive these services did receive standard care including referral to community services when indicated.

In the total intervention group, there were no statistically significant differences in outcome at age 18 years compared to the control group, however, when the intervention group was stratified into infants with a birth

weight of < 2000g i.e. low low-birth-weight (LLBW) and those with a birth weight of >2000g i.e. high low-birth-weight (HLBW), statistically significant differences were found in the HLBW group who received intervention compared to the HLBW group who did not receive intervention. Higher IQ scores, better maths and reading scores and fewer risky behaviours were found in the intervention group compared to the controls. There was also a trend towards fewer of the intervention group being classified as needing special education (17% vs 24%). (47) This highlights not only the benefits of EI, but also the importance of identifying and defining specific variables when assessing outcomes of early intervention programmes

Stratifying the groups based on the amount and intensity of therapy services therefore may have resulted in statistically different outcomes as it did when groups were stratified by birth-weight. In looking at the poor effect of EI for the LLBW group, the authors did not account for the fact that most of the infants in the intervention and control groups were likely to have received EI as standard care (10, 16). Kalia, Visintainer and Brumberg (21) reported 70% of VPT, compared to 30% of LPT infants required enrollment in speech, physiotherapy or occupational therapy. The percentage of infants from the IHDP study receiving EI in the LLBW groups may have been similar. This could therefore have accounted for similar outcomes in the intervention and control groups, as both groups received standard follow-up care.

The influence of intensity of intervention on outcomes can be seen in the IHDP study where infants who attended >400 hours of the EI programme (out of the 500 hours in total) scored higher on IQ tests and did not show an attenuation of EI benefits with age (33). Reuner et.al. also noted that intensive follow-up and intervention received by the infants in their follow-up study of low birth-weight infants may have been the reason for the low prevalence of cognitive deficits found in their cohort (10).

A four-point difference in favour of the intervention group on the Peabody Picture Vocabulary test (PPV-III) a measure of scholastic aptitude - remained stable from 8 to 18 years showing the potential of early intervention to lower the percentage of children who do poorly at school. The authors (33) stated that although true long-term

economic benefits could only be identified when the subjects reached 27 – 40 years of age, the current phase of the IHDP study supported the benefits of early intervention for long-term outcomes.

Fiscella and Kitzman (32) concluded their report on the disparities of academic achievement and health by recommending that closing gaps in children's academic achievements will require the funding of early child and family intervention programs. In 2008 the cost of EI for pre-term infants was reported to be $611 million a year and special education $1.1 billion a year, with the annual economic burden being $26.2 billion (30). It is therefore essential that the benefits of EI be scientifically documented and services for this at-risk population group optimized to ensure best possible outcomes. Infants and families that participated in the IHDP programme have shown quantifiable benefits thus offering excellent scientific evidence for the effectiveness of EI.

Regarding the IHDP,

> *"It is very significant that early intervention can still make a measurable difference in such a diverse group of children after 18 years"* (48)

10.2.2 Issues in Early Intervention

The practice of developmental-behavioral medicine and the implementation of early intervention are encapsulated in the statement from the collaborative scientific report 'From Neurons to Neighborhoods',

> "*the course of development can be altered in early childhood by effective interventions that change the balance between risk and protection, thereby shifting the odds in favor of more adaptive outcomes*." (1)

EI is expensive and labour intensive however. Not all infants need it, but those that do need it earlier rather than later. It is therefore important not only to identify which interventions are truly effective in improving the long-term outcomes of this vulnerable population, but which infants would in fact benefit from the different interventions. (10, 45, 49)

Reviews and meta-analyses on the effectiveness of EI find the diversity of assessment tools, length of follow-ups, as well as the heterogeneity of treatments (time of initiation, duration, intensity and type of professional leading the programme) limit the ability to compare results between studies effectively. (44, 45, 48)

There are also a number of important issues to consider when reviewing literature on EI. Firstly careful selection of cohorts and outcome measures are essential when assessing the benefit or effect of EI. Secondly, an understanding not only of the great variability of normal development, but of the interplay and interdependency between the different developmental domains is critical, particularly in the younger infant. Thirdly, the effect of intervening variables on these developmental domains such as socioeconomic status (SES) and medical risk factors cannot be ignored as they strongly influence infant outcomes.

10.2.2.1 Intervening variables

Intervening variables confound research into the long-term outcomes of EI. The developing child remains vulnerable throughout the early years and even into adulthood so that early benefits can be negated by negative experiences or enhanced by positive ones (1).

The effect of experiences can be shown in long-term follow-up studies where it has been found that the positive effects seen in children who received EI attenuates with age. There is less of an advantage in the older child and young adult who received EI compared to those who didn't. This decreasing advantage may be explained by the fact that in many of the long-term outcome studies, although "control" group children did not receive specific EI treatment they received therapy or assistance in line with normal protocols. This potentially influenced the long-term outcomes of the control group and may have been a reason for their "catching-up" with the intervention group (10). On the other hand variables such as disadvantaged economic backgrounds can negatively influence development. For example, performance by school-age in the domain of fine motor

development has been shown to be particularly influenced lower SES and lower birth weight, resulting in worse outcomes for these children (50).

10.2.2.2 Selection of Cohorts

EI can mean intervention that occurs early in life or intervention that occurs early in the expression of a morbidity. Most studies looking at the benefits of early intervention focus on infants at risk for developmental difficulties. At these early stages, developmental difficulties are not readily apparent and intervention services are therefore often targeted at children who may not need or benefit from them. These EI programmes can be classified predominantly as "prevention programmes" since specific problems that require "treatment" are not yet identifiable. This results in studies not assessing the true benefit of early intervention in the infant and child with actual developmental delay (44). On the other hand, once a developmental delay or disability is recognized it would be unethical to withhold treatment, making it difficult to assess the true benefit of intervention and the effects of intervention applied earlier rather than later in an identified condition.

10.2.2.3 Assessment Times and Tools

Spittle, Orton & Doyle (44) questioned how the reliability and validity of the assessment tools at different ages influenced results. For example, two of the studies assessed in their review which had shown no difference between the treatment and control group at one year, showed a significant difference in favor of the treatment group at two years.

The possibility of selecting the incorrect tools to detect change was pointed out by Rosenbaum when describing their study of gross motor development in pre-school children with Down Syndrome. In this study, although the Bayley Scales of Infant and Toddler Development – II (BSID-II) was able to discriminate the motor function of children based on age, it was unable to pick up change over a 6-month period. In the same population group however, the GMFM (Gross Motor Function Measure) was able to show graded changes over the same time

period, illustrating the limitations of well-developed and validated measures when they are used for purposes other than what they were designed for (51).

10.2.2.4 Developmental Domains and Outcomes

Understanding the relationship between early developmental delays and long term outcomes is also vital to the effective analysis of EI programmes. Some studies have shown a poor correlation between early difficulties and later outcomes in a specific domain. As an example there may be little significant difference in motor outcomes in the older child who had motor delays in infancy compared to the normal population. (52, 53) Piek, Dawson, Smith and Gasson (50) assessed developmental trajectories in infants across several developmental domains using parent-administered questionnaires. They found that although the infant's gross and fine motor performance was not predictive of school-age performance in gross and fine motor domains, there was a significant relationship between early gross motor performance and school-aged academic performance.

Other studies have supported this link between gross motor performance in the infant and cognitive outcomes in later years (54). In their cohort of low-risk infants (mean gestational age = 37 weeks), Piek et.al. (50) found motor development was significantly predictive of school age cognitive performance, specifically working memory and processing speed, regardless of gestational age or socioeconomic status (SES). In contrast there was no correlation between the infant's cognitive function (problem-solving and personal-social skills) and later intelligence tests at school-age.

10.2.2.5 Variations in normal development

A unique aspect of the study by Piek et.al. (50) was the ability to assess development at several time points (each child underwent 11 separate assessments by their fourth year), creating an early developmental trajectory, rather than a once-off assessment of development for each child. This accounted for the increasing evidence of intra-individual and inter-individual variation of infant development as shown in a study of toddlers who were all assessed as normal at 18 months. Prior to their 18-month assessment 31% had fallen below the

10th percentile in at least one developmental assessment (55). Typical development is not linear and does not occur at a consistent rate. This makes it difficult to distinguish normal from delayed and transient disorders from persistent impairments (1) and highlights the need for the assessment of development to include multiple domains and be repeated at multiple time points (55, 56).

To assess a group of infants for research or intervention purposes at frequent intervals is prohibitive in terms of costs, time and the level of expertise required, as experienced clinicians are essential in performing developmental assessments of acceptable quality (12, 57). Piek et.al (50) clearly demonstrated that using parent-administered screening tools can be effective in research and assessment and therefore may be used to overcome some of these barriers. Use of parent-administered screening tools in screening for developmental delay has been frequently recommended for use in identifying children with possible disordered development for both clinical and research purposes. (57, 58)

10.3 Screening for Developmental Delay

Early identification of developmental delay is critical for the effective implementation of early intervention (EI). The earlier intervention is begun, the more effective it can be in remediating or even preventing the negative sequelae which result from developmental difficulties. (39, 47, 59)

The early detection of developmental delay is not straight forward and has been decribed as trying to measure a moving target, with a wide variance in the timing of skills emerging, or the possiblity of skills being latent (not yet measurable), delayed, deficient, or disordered at any one point in time (49). The wide variability in normal development makes subjective assessments unreliable and studies have shown improvements in accuracy and a decrease in missed early diagnosis with the use of standardised tools. (47, 60, 61)

In a 2006 policy statement on identifying infants and young children with developmental disorders, the American Academy of Pediatrics (AAP) stated that the early identification of developmental disorders is the responsibility

of all paediatric health care professionals (58). The AAP recommend using surveillance and screening processes to identify those children who would benefit from an assessment or evaluation. Surveillance is described as the process of identifying children who may be at risk of developmental difficulties through a continuous and longitudinal relationship with the family and child, whilst screening is described as the use of validated tools to identify and define that risk. An assessment or evaluation is described as a complex process aimed at identifying a specific developmental disorder and involves the use of tools that are expensive, time intensive and require a high level of expertise and experience from the administrator.

A screening tool does not result in either a diagnosis or a treatment plan but rather identifies areas in which a child's development differs from the same-age norms. Screening tools are quick to administer and can often be completed by the parent or caregiver. The AAP recommends developmental screening using high quality standardised tools as part of the usual practice of well-baby care for all infants and children regardless of their risk status and even if they appear to be developing normally. Tools should be used at regular intervals or whenever there are any concerns raised by the parent, health professionals or others involved in the care of the child. They recommend repeated and regular screening to account for the dynamic nature of development and the inherent limits of screening tools. They caution that waiting until a child misses a major milestone before initiating evaluation processes will deprive the family of the benefits of early identification and early intervention. Failure to detect difficulties early results in missed opportunities to address problems before they become more limiting and costly to treat. Problems which may have been easily remediated with EI may require much more extensive intervention later in life, with the possibility of less optimal outcomes. (58, 62, 63, 64)

One argument against the use of ongoing developmental screening is presented by Johnson & Marlow (65). The authors argue that in premature infants the initial screening stage is redundant because abnormalities shown by premature infants within the first two years of life are variable and transient, whilst some disabilities such as cerebral palsy only become apparent around two years of age. They state that because prematurity is such a 'clear medical risk factor' for developmental delay, it is preferable for all children born prematurely to rather have a hands-on, in-depth developmental assessment at 2 years of age. The problem with this approach

is three-fold. Firstly, many children are lost to follow-up and may miss the 2-year assessment, only to re-emerge at school age when EI is too late (17, 62). Secondly, enrollment of infants in EI programmes before the end of the first year is possible and has been shown in many studies to be a realistic goal for optimizing intervention (10, 32, 45, 47, 66). Waiting 2 years before identifying children with difficulties is not conducive to EI which aims to start as early as possible in order to maximize the benefits of neural plasticity and neuronal network selection (35). Thirdly, most children with severe or clinically evident delays such as cerebral palsy are identified earlier, but more subtle impairments are difficult to identify, highlighting the need for ongoing and regular screening (2, 67, 68). Even though prematurity is a higher risk for developmental delay, developmental problems in premature babies are not always identified earlier than in full term babies, but earlier recognition of problems is associated with earlier referral to rehabilitation services. The use of early systematic developmental screening to aid early identification and early referral is therefore recommended (17, 69).

The value of these recommendations was recently shown in 2007 by van Agt, et al. (38) in a cluster-randomized control trial of 5406 children. In this study children were followed up at eight years of age to assess differences in educational needs and academic function between those who had undergone systematic developmental screening and those who had had normal paediatric care. More than fifty physicians were randomly assigned to control or intervention groups. Physicians in the intervention group were trained in EI screening methods for language delays, whilst physicians in the control group provided normal care which involved physician's observations and parent questioning, without standardised methods of assessment or clear-cut referral criteria. Children were seen by either the 'control group' or 'intervention group' physician before 2 years of age and followed up at 8 years of age with a variety of assessments. Low and high SES status between the groups was similar as was the number of children lost to follow-up. The study found that significantly more children in the intervention group received treatment and support before the age of 5 years than in the control group (ρ = 0.024). By 8 years of age the number of children in either group receiving therapy was no longer statistically significant however the difference in the number of children with special education requirements and linguistic difficulties was statistically significant between the two groups. The intervention group showed a 30% reduction in the number of children attending special education and a 33% reduction in children with spelling difficulties,

emphasizing the positive benefits of starting intervention earlier. This study clearly shows how implementing standardised screening can improve the timing of early referrals to enhance the benefit of EI in children with developmental difficulties.

Rydz, Shevell, Majnemer and Oskoui (13) justified screening for developmental delay within the guidelines from the World Health Organization on "Principles of Screening for Disease". These include showing that developmental disability carries an appreciable burden to the individual, the family and society; that there is suitable testing available and acceptable treatment for patients with the condition; and the costs of case-finding and treatment is balanced by the benefits to the individual and society.

An added benefit of implementing standardized developmental screening was found in a study that showed parents whose children received a developmental assessment were more likely to be aware of issues related to development. This included knowledge of developmental milestones such as first steps, first words or toilet training as well as the impact of the family and community on development. Examples of this include better knowledge about nutrition and health, discipline, smoking and substance abuse, parental support and spousal support. The parents were more likely to report being satisfied with the medical care their child received and their children were more likely to have access to developmental services (70).

10.3.1 Properties of Screening Tools

The AAP recommends that screening tools should address the different developmental domains including gross motor, fine motor, language and social-emotional development, as well as adaptive skills. Screening tools should be reliable and valid with sensitivity and specificity of 70% - 80%. The AAP recommendations note that it is acceptable for screening tool values to be lower than other medical screening tests because of the challenges inherent in measuring child development (58). Screening tools must be brief with proven predictive value. They must have been standardised within the last 10 years and validated against other high quality diagnostic tests

with proven reliability including test-retest, inter-rater and internal consistency. They must also included clear criteria for passing or failing and guidance on what to do with the results. (49, 71).

Screening tests are considered successful when they accurately identify the presence or absence of disease in a population group. Formal terminology around this concept has been summarized in Table 2 (13).

Table 2 Testing Terminology

Terminology	Definition	Referral for treatment
True-positive	Test correctly identifies someone with the disease	Appropriate
False-positive	Tests incorrectly identifies someone with the disease	Over-referral
True-negative	Test correctly identifies someone without the disease	Appropriate
False-negative	Tests incorrectly identifies someone without the disease	Under-referral
Sensitivity	The proportion of people with the disease whom the test will correctly identify	High sensitivity → appropriate Low sensitivity → under-referral
Specificity	The proportion of people without the disease whom the test will correctly identify	High specificity → appropriate Low Specificity → over-referral
Reliability	The stability of the screening tool when it is repeated under identical conditions. Variance in score results is due to true variance in those tested. Includes: interrater reliability, intrarater reliability, test-re-test reliability, internal consistency	
Validity	The extent to which an assessment measures what it is actually supposed to measure. Includes: content validity, construct validity, criterion validity (concurrent and predictive)	

A test that has not been compared to "true" diagnostic states should not be labeled with sensitivity or specificity claims. Aylward (12) therefore cautions against the use of sensitivity and specificity terminology, arguing that there is no real "gold standard" in developmental evaluation and suggests the terms 'copositivity' and 'conegativity' may be more accurate. Copositivity refers to the extent to which two tests agree that a patient has a condition, whilst conegativity is the extent to which the test agree on dismissing those patients without a condition. They are tests of reliability, not validity, because both tests could be wrong, but still correlate highly. Developmental tests also need to be constantly updated to keep up with new knowledge and to accommodate the Flynn effect, making newer tests (including the BSID-II) less well validated. The Flynn effect is the rise of the

average IQ test scores over generations (IQ gains over time). It is an effect seen in most parts of the world, although at varying rates. 'Copositivity' and 'conegativity' are therefore more accurate, but not used frequently in the literature however and sensitivity and specificity are still the most frequently used terminology. To account for the fact that there is no true standard reference or 'gold standard' in developmental testing, screening tests should therefore also be compared to a battery of tests that take functional outcomes into consideration such as school performance, in-grade retention and enrollment in special services.

Although limitations of screening tools can lead to misclassification of a child's developmental status (false positives and false negatives), the goal of developmental screening is to detect children who need further evaluation, not to diagnose developmental delays or disorders. Children with false positive screens will be identified as age-appropriate in the next stage evaluation, avoiding over-burdening intervention services, whilst ensuring that most children who do require early intervention are in fact identified early (49).

Further, there is evidence that children with a false positive screen (those who fail screening but are not found to have a clinically significant delay on further assessment) are at greater risk for scoring in the lower ranges of normal than those who pass initial screening, provided the screening test used has good psychometric properties. Glasocoe (72) found that 70% of children with false-positive screening results scored below the 25th percentile (the cut-off used for placement in remedial reading and mathematics programmes) on one or more diagnostic measures of adaptive behavior, intelligence or academic achievement. Therefore, although this group of children do not need special education, they are a high-risk group for under achievement and poor school performance and benefit from closer monitoring and early stimulation programmes. This indicates that over-referrals are not really a negative characteristic of developmental screening, but rather provide a beneficial service to highlight a group of children who may benefit substantially from further educational support. This is especially important in light of the fact that those children with more subtle problems are more likely to have a significantly positive response to treatment, but are less likely than those with more serious conditions to be identified early (73).

10.3.2 Paediatricians and Developmental Screening

The AAP document (58) states that although undetected or untreated disordered development in early childhood can contribute to early school failure and attendant social and emotional problems, detection of developmental delay before school age is significantly lower than the prevalence of these delays. Only about 30% of children who would benefit from early intervention are detected before school age (74).

Paediatricians play a crucial role in identifying infants with developmental delay. They have an accepted role of authority and are in regular contact with the child from birth. Children are more likely to see a paediatrician than any other professional before school age and parents have been shown to take their recommendations seriously. Paediatricians are ideally situated to implement developmental screening and identify developmental delay in time for effective referrals to early intervention services. (13, 68)

In the 2006 report, the AAP identified developmental screening as an area of paediatric care that is critical to the well-being of children and their families and the responsibility of all pediatric health care professionals. They found very few paediatricians were using effective means to screen their patients however (58). Most physicians in the USA were committed to the early diagnosis of developmental delay. Barriers of time and poor reimbursement as well as under-reliance on parent-administered questionnaires were identified as areas to be addressed (75). This is not unique to the USA and similar rates of under detection with poor reliance on parent-administered questionnaires were reported in other first world countries including England, Australia, Canada and Ireland. (64, 76,77)

In a literature review on developmental screening it was reported that most physicians use developmental milestone lists and informal checklists rather than standardized screening tools (73). Using milestones and checklists are an ineffective means of detecting developmental delay. They often fail to include tasks that are strong predictors of delays and disabilities or school success. Many are based on studies that are outdated or based on limited sample sizes with a lack of population diversity. Most milestone checklists estimate the 50th percentile of population groups and do not effectively represent the large variability of normal development. It is

more useful to present information on the "typical range" of skill acquisition, or preferably, the mean age and standard deviation. This is especially true when the information is based on recent, large, diverse cohorts and linked to clinical problem solving strategies based on scientific research, as can be found in more modern and standardized screening tools. (61, 71)

Barriers to the use of screening tools for paediatricians include insufficient time to implement them and insufficient reimbursement for time spent using them; Unfamiliarity with screening tools, inadequate office structures or under-skilled support staff also impede their use. The lack of available services to refer children identified with risks, or a lack of knowledge around these resources is also a reason cited for not using screening tools (75, 78). Financial barriers to developmental screening were recognized in an economic analysis of developmental detection methods in 1997. This report concluded that it may not be in the best financial interests of physicians to perform developmental screening as although it is in the best financial interest of society to identify and treat children with delays, the financial burden is borne primarily by the paediatrician (79).

In a postal survey of paediatricians and general practitioners in America by Sices, Feudtner, McLaughlin and Drotar (75), only about half of physicians reported using a validated screening instrument and less than 15% used parent-completed questionnaires. Validated parent questionnaires address many of the barriers cited by paediatricians for not using screening tools. They are cost-effective, simple to use and reduce the amount of provider time needed for developmental screening (78).

The success of implementing developmental screening in physicians' offices using parent-administered questionnaires was documented in North Carolina. In 1999 only 2.6% of children between 0 – 3 years of age in North Carolina were receiving early intervention services despite the fact that an estimated 8% - 13% would have qualified and benefited from EI. The ABCD programme (Assuring Better Child Health and Development) was implemented statewide in physician practices to promote screening of all children using parent-administered questionnaires. The number of well-child visits incorporating developmental screens increased from 15% in the year 2000 to 80% by the year 2008. As a result, between years 2004 and 2008, referrals to EI

programmes quadrupled, resulting in fewer North Carolina children entering school with unrecognized or untreated developmental problems (62).

10.3.3 Parents and Developmental Screening

A 2005 study (69) found that physicians were more likely to be the first to identify concerns in prematurely born infants, but parents were more likely to be the first to identify concerns in full-term infants. Neither the severity of the delay or the SES of the parents was associated with who first identified concerns in the infant's development. Infants who were initially identified by physicians were likely to receive EI earlier than those first identified by their parents however. This implies that parent's concerns were not given adequate recognition, an assumption supported in a study conducted by Sices et.al (75). In their survey less than 15% of 800 paediatricians and family physicians agreed that parental concern was a good substitute for developmental screening. In another study parental concern was also not associated with any increased probability of referral to diagnostic or intervention services (80).

The ability of parents from differing economic and social backgrounds to accurately identify developmental difficulties in their infants has been established in the literature (69, 81), and has supported the development of screening tools using parent-administered questionnaires to elicit parental concerns and quantify their observations.

10.4 Parent-Administered Questionnaires

When systematically elicited, parental concerns can identify children with developmental delays with a specificity and sensitivity approaching those of physician administered screening tests (81, 82, 83, 84). Using parent-completed questionnaires involves parents in the process of child health supervision. This makes them partners early on in the process and consequently enhances early intervention, which is most effective when the parent-child relationship is emphasized. (1, 32, 47)

Using parent-completed questionnaires also offers substantial monetary and time-savings over other screening methods (68). The use of validated and standardised parent-administered questionnaires was recommended by Rosenbaum, Missiuna, Knox and Echeverria (57) as recently as 2009. The authors described parent questionnaires as an appropriate and cost effective method for identifying children who require more detailed assessments in large-scale epidemiological studies. According to Ronsenbaum et al., parent-administered questionnaires are able to address both quantitative aspects of development, through questions about specific milestone acquisitions, and qualitative aspects, which can be elicited through unambiguous open-ended questions.

Although parent-reported screening tests have many advantages, problems may arise when they are used in populations with limited literacy. Caregivers may respond randomly to questions, omit answers or not be able to use the questionnaires. These problems are easily overcome by offering assistance with filling out the questionnaire or performing the screen as an oral interview (13).

Two of the most frequently cited and recommended parent-administered questionnaires used for developmental screening purposes are the Ages and Stages Questionnaire (ASQ) and the Parents Evaluation of Developmental Status (PEDS) (56, 58, 62, 85, 86).

10.4.1 ASQ

The ASQ is a child monitoring system designed to be completed by the infant or child's primary caregiver in their home or in a clinic or community setting. It is primarily a milestone based checklist that has been standardized and overall has high levels of validity (0.86 to 0.91), reliability (interrater > 0.85, test–retest > 0.90), sensitivity (71.88%) and specificity (85.84%). (76, 87, 88)

The ASQ has been used for developmental monitoring in epidemiological studies (89, 90) and follow-up of premature infants and paediatric patients with various medical conditions or developmental risks (91, 92, 93, 94,

95, 96, 97, 98). It has also been used in state-wide child development projects such as the Assuring Better Child Health and Development (ABCD) project (99). The ASQ has also been shown to be effective in identifying developmental information that can predict specific school-age outcomes (50).

The ASQ is validated and is widely used in preventative health care in the USA and Canada. It has been used in studies from both first world and developing countries, including South Africa (76, 88, 89). It has been translated into at least 6 other languages, with additional validation studies being done in Norway, Korea and Holland where it has shown good psychometric properties (100).

The effectiveness and the cost of the ASQ in paediatric clinical practices was examined in a large study, consisting of 1428 parents or legal gaurdians and their infants aged 12, 24 or 30 months (17, 101). In this study parents or caregivers completed the ASQ when they arrived for their well-baby visit. Paediatricians were blinded as to the result of the questionnaire and followed usual assessment and referral procedures. It was found that the training required to implement the ASQ in the office setting took less than 30 minutes. It took less than 30 seconds for office staff to give instructions to the person completing the form and approximatley 4 minutes for the office staff to score the ASQ, enter results in the medical records and complete recommended referrals. The cost of the implementing the screen was less than $2.50 per patient in 2005.

A follow-up study in 2009 examined the long-term effects of implementing the ASQ (17, 101). After implementing the screen, referral rates to EI services increased by 224% with the greatest increase being in the 12 month old age group. The number of children who actually became eligible for EI services more than doubled (from 42 – 89 patients) in the first year of the study. Without the results from the ASQ, 37 of these children would have missed EI referals and 52.9% of preterm children would not have had early referrals. Rather than comparing the questionnaire against a gold standard, refferals from the ASQ were determined as appropriate if the child met “real-practice” eligibility criteria used for EI services or qualified for Early Childhood Special Education as legislated by the Oregon Department of Education. Developmental difficulties in 38% of 12 month-olds and 23% of 24 month-olds were not identified by the paediatricians, whist a ‘wait and see” approach

resulted in missed EI referrals of a further 13% of 12-month-olds and 16% of 24-month-olds. Although paediatricians' appraisals were almost always accurate in the children they identified with developmental difficulties, they had more difficulty identifying younger infants, lower-risk preterm infants and those with more subtle delays. Implementing the ASQ was found to be feasable and possible to administer at low cost without impeding office flow. It was strongly recommended for use as a tool for the early detection of developmental delay.

10.4.2 PEDS / PEDS:DM

The PEDS is a very simple tool consisting of only 10 questions which are designed to systematically elicit parental concerns. The PEDS has been peer reviewed, validated and standardized. It is an appropriate method for use in children from birth to 8 years of age. (102, 103) Its application has recently been expanded by the introduction of the PEDS:DM (Developmental Milestones). This is a brief milestone-style checklist from birth to 7 years 11 months, and can be administered either through the screening/surveillance version, or as an assessment-level version for use with programmes serving children at risk (such as NICU follow-up and EI services). This version provides both age-equivalent scores, and determines percentage of delay . The PEDS:DM has also been standardized, has high levels of validity and reliability and overall excellent sensitivity (83%) and specificity (84%). (71) It is recommended that the PEDS and PEDS:DM be used together for optimal developmental screening. When used together, the screening test is refererd to as PEDS COMBINED. PEDS COMBINED results in more easily triangulating parents concerns and child's performance, negating the need for second stage screening and facilitating rapid referral to EI services when indicated (104).

The PEDS has been used in prevalence studies of developmental disorders (105). It has also been used to assess outcomes of parenting and social variables on development (2, 106). It has been validated in the USA and trialed in Australia and India (107, 108). It is recommended for use in state-wide child development projects like the Assuring Better Child Health and Development (ABCD) project (99).

In their 2009 study on practical implementation of the PEDS in an office setting, Schonwald, Horan and Huntington (85) reported that practitioners found the PEDS easy to use and because it provided an organized structure for discussing parents concerns, it actually saved time during the visit. Several providers noted an improved ability to identify developmental concerns following use of the PEDS. The PEDS was given to parents in the waiting room whilst they were waiting for their appointment. It was noted that parents consistently had enough time to complete the form. They found that it was helpful in reminding them of questions they had wanted to ask the doctor and in reminding them about the importance of specific aspects of their child's development. Following implementation of the PEDS in the office setting, significantly more parents reported that they had talked with the provider about developmental concerns and significantly more parents reported receiving answers to their concerns (ρ = 0.04), despite the providers impressions that they were addressing parents' concern's at every visit pre-implementation of PEDS. These findings, along with increased identification of developmental concerns, have been corroborated by other research. (109, 110, 111)

10.4.3 Comparison of the ASQ and PEDS COMBINED

Although both the PEDS and ASQ have acceptable psychometric properties and are recommended for screening similar population groups, a recently published paper has questioned if these two questionnaires are able to concur on which children require additional developmental assessments. No single pattern of discordance emerged, but ratings in the language and communication domain seemed to differ most often between the screens (112). It is important to note that the PEDS COMBINED separates language development into expressive and receptive language components, whilst the ASQ combines these developmental components into one domain. Sensitivity and specificity of the PEDS COMBINED and ASQ are summarized in Table 3.

Table 3 Sensitivity and Specificity of ASQ and PEDS COMBINED

		Overall	5-7 months	8 months	10 months
ASQ	**Sensitivity**	70%-90%	Not available	51.02%	Not available
	Specificity	76%-91%	Not available	83.2%	Not available
PEDS combined	**Sensitivity**	70%-90%	86%	76% (8 – 10 months)	
	Specificity	70%-93%	77%	83% (8 – 10 months)	

In comparing the questionnaires, the PEDS is predominantly designed to elicit parental concerns which are validated according to age norms, whilst the ASQ is predominantly a standardised developmental check-list. Parental concerns are also elicited, but not statistically validated (86, 88, 103, 113).

Both the ASQ and PEDS:DM are based on developmental milestones. Milestones were chosen according to validated, objective measures for the PEDS:DM, whereas ASQ measures were chosen more subjectively and then validated. For the ASQ, items that were thought to be easy for parents to observe or ellicit at home were selected from from a variety of different sources such as milestone checklists, textbooks and standardized developmental tests. The ASQ was then validated as a tool using these items (88). For the PEDS:DM data was drawn from standardization and validation studies of the Brigance Inventory of Early Development-II (IED-II) and the Brigance Comprehensive Inventory of Basic Skills-Revised (CIBS_R). Binary logistic regression analysis was used to determine which items were the most predictive of performance and development difficulties for each age in each domain. If more than one item was available, the final item was chosen based on properties such as how quickly it could be administered, how easily it could be explained and how likely it was for parents to be able to observe the skill. The PEDS:DM was then validated using these items. (71)

The PEDS:DM has a readability two grades lower than the ASQ, making it quicker to read, It contains only one item per domain per age level (from 0 – 8 years), whilst the ASQ contains 6 items per domain per age level (from 4 – 60 months). The ASQ consists of several age-appropriate forms and the correct form must be

completed at a specific month of age, whereas the PEDS:DM assessment level version is one form that can be completed at any age between 0 – 8 years. PEDS is designed to be completed in the office waiting room, whilst the ASQ is better suited to be completed in the child's home setting and then mailed back or brought in for the next well-child visit. (71, 88, 113)

In comparing the ASQ and the PEDS; Sices, Stancin, Kirchner and Bauchner (112) concluded that the poor concordance between the tools did not negate the importance of screening to prevent under-detection of developmental delays, but highlighted the need for further research to compare the performance of these tests in the clinical setting.

10.5 Low and Middle Income Countries

South Africa falls under the classification of Low and Middle Income (LAMI) countries (114). The public health care sector reportedly serves 80-85% of the South African population although it accounts for only 39% of the total health care expenditure (115, 116). It is therefore important to orientate developmental screening and EI in South Africa within the context of LAMI countries.

The causes of developmental disability in children from developing countries are often similar to those from developed countries, however additional risk factors amplify adverse outcomes. These may include poverty and poor resources; HIV; infections like TB meningitis and cerebral malaria; and nutritional deficits. In LAMI countries very small premature babies are less likely to survive due to lack of sophisticated NICU care but LBW infants and birth complications are common. These infants survive and may have developmental outcomes similar to those of the more premature and ELBW infants who survive in developed countries. Information about long-term outcomes following neonatal high-risk conditions in LAMI countries is inadequate. (117, 118)

A 2007 review addressing childhood development and disability in low and middle-income (LAMI) countries (117) found large gaps in the knowledge of screening, service provision, policies and legislation. Only four

studies from South Africa were included, despite the wide eligibility criteria for papers (with no limitation on year, type of study or quality). In their review on LAMI countries the Ten Questionnaire (TQ) was the most commonly used screening tool. This tool was developed for use in identifying severe childhood disability and has been shown to be ineffective in identifying less severe forms of developmental disabilities such as moderate mental retardation. Most studies were cross-sectional and could not track changes over time. A need for longitudinal studies to assess outcomes from different types of service utilization was therefore highlighted. Longitudinal studies are also necessary for better estimates of the burden of childhood disability and the gaps that exist in health-related services in LAMI countries. No studies in the review addressed the issue of cost of early intervention and there was a lack of knowledge about the effectiveness of early intervention programmes. The review also found little information in the literature about health-related policies and legislation regarding childhood disability in LAMI countries, including South Africa. In the presence of limited budgets, research into the cost-effectiveness of interventions is essential with policies and legislation being critical in enabling effective utilization of available resources.

"*Successful school entry and primary school completion are substantive challenges facing many children in developing countries. Without identification and support, children with neurodevelopmental disabilities may not even begin to achieve these two central benchmarks of successful development*" (118)

10.5.1 Recommendations

There is a great need for culturally appropriate interventions that can be sustained in LAMI countries. Intervention is crucial for these children to improve their health related QoL and break the cycle of poverty. Failure to recognize the impact of developmental disabilities along with the lack of developmental experts and financial resources has resulted in very few EI programmes. There is therefore a need for research to identify and define the necessity for intervention in these countries by following the natural history and impact of various conditions, particularly the combined effect of negative insults experienced by children in LAMI countries.

Longitudinal monitoring of outcomes of interventions is also crucial to ensure their appropriateness and effectiveness. (40, 117, 118)

Recommendations for LAMI countries are to use tools that are easy to administer and that screen for disabilities in children less than 3 years of age. Tools must also provide knowledge and supportive services to families and their children. Using tools that have been standardised in other countries do not take cultural differences into account and decreased levels of validity and reliability may occur once the tests have been translated into the native language. A dilemma exists however, as the cost and time needed to develop culturally specific tools and perform extensive psychometric testing on them, particularly in light of the limited expertise and funding available in LAMI countries, is a barrier to validation studies. (114)

It has been suggested that children from different populations may attain developmental milestones at similar ages, possibly eliminating the need to standardize and validate instruments on developmental milestones in each country (119). Ethnic differences in the attainment of milestones does occur and socioeconomic factors have a marked influence (14). Validation studies on the ASQ in the USA, Canada, Korea, Holland and Norway have been done on a wide variety of infants. Although these are first world countries, populations representing all socioeconomic levels and many different ethnic groups were used in the validation studies (100). The ASQ was also chosen to follow-up children in a large, international multicentre trial (89), run in 33 countries (including South Africa) specifically because of low cost and flexibility across a wide range of cultures. In this study (involving 828 children, two-thirds of which were from developing countries), ASQ specificity was 82.3% and sensitivity was 87.4% showing acceptable psychometric properties in LAMI countries and supporting the ASQ for use in low-cost, large-scale screening and monitoring of childhood development.

Ertem et.al. (119) emphasized the need for using standardised tools in LAMI countries where studies have shown health care providers are not well equipped with knowledge about early childhood development. Family centered methods for monitoring child development, such as parent-administered questionnaires, are highly recommended in light of the fact that caregivers in LAMI countries are a key resource to support their child's

development. When selecting which screening tools to use, the authors caution against those developed for high-income countries due to lower literacy levels and cultural differences found in LAMI countries. Both the ASQ and PEDS have been validated and used in low socio-economic groups where literacy levels and cultural influences may be similar to LAMI countries, supporting their use in this context. (88, 102).

10.5.2 The South African Context

Private health care services in South Africa offer more first world medical care than is possible in the public sector. A recent study looking at NICU care in both public and private hospitals in South Africa found the private NICUs met many of the international standards. They had enough staff to exceed the nurse:baby ratios of 1:2; nursing staff were knowledgeable and worked under the supervision of neonatologists and all babies requiring ventilation were ventilated regardless of birth weight. All babies on oxygen were also appropriately monitored and screened on a regular basis for retinopathy of prematurity (ROP). In contrast public NICUs often had a shortage of monitoring equipment and staff. The nurse:baby ratio regularly reached 1:4 and units were understaffed or staffed by nurses with insufficient training. In some public NICUs there were no resident medical officers at night and only general duty doctors on call. Monitoring for ROP was frequently non-existent. There was a general policy of not ventilating ELBW babies and knowledge of standard NICU care by lower level staff (such as oxygen saturation levels) was insufficient (116).

Despite these differences in standards of care, Cooper and Sandler (120) reported comparable outcomes and rates of handicap at 18 months in a cohort of VLBW infants from a public sector hospital in Soweto, South Africa to those from developed countries. Similar to international studies, South African studies from 1985-1999 have shown improved survival of NICU infants but with a resultant high prevalence of developmental delay. Lower SES, particularly maternal education, was found to be a predictor of lower developmental scores for NICU infants in South Africa (120,121, 122, 123, 124,125).

The private health care sector serves only 15-20% of the population but is responsible for 61% of the total health expenditure (115). In 1981, a survey of all paediatricians registered with the South African Medical and Dental Council showed 45% of registered paediatricians worked in private practice (126), however no publications on the practice of general developmental screening by paediatricians in private practice in South Africa could be found. Studies on newborn hearing screening and screening for ROP in public and private hospital settings are available, as well publications on screening and EI related to speech, language and audiology. (115, 116, 127, 128, 129)

10.5.2.1 Early Intervention

In line with international research on EI described in this review, outcomes of children who received Cochlear implants in the Pretoria Cochlear Implant Programme indicate that key factors related to positive outcomes in South African children's audiological, linguistic, social and educational functioning were early age at diagnosis and early intervention (130).

HI-HOPES is an EI programme launched in 2006 at the Wits Centre for Deaf Studies in Gauteng, South Africa, servicing all economic sectors (131). Private practitioners referred 19% of infants on the programme, compared to 22% being referred via word of mouth and media advertising. The remaining infants were referred by public and private hospitals (56% and 3% respectively). Analysis of this programme shows the average age of initial registration with the programme was 25 months, with the majority of infants having profound or severe to profound hearing loss. The average age of initial diagnosis was 15 months, indicating a significant gap of nearly a year between diagnosis and intervention. This is despite the reported intervention guidelines for children with hearing loss recommended by the Health Professionals Council of South Africa. These guidelines state that the identification of potential hearing loss should occur by 1 month of age, diagnosis confirmation by 3 months of age and referral to EI before 6 months of age (131).

The value of these guidelines are shown in the test results of infants on the HI-HOPE programme. There was a marked difference in improvement of both receptive and expressive language in children identified before 7 months of age compared to those identified later. These findings of late identification and implementation of EI were similar to another South African Study looking at the identification and treatment of children with early onset hearing loss in Pretoria, where the mean age of diagnosis was 23 months and the mean age of initial hearing-aid fitting was 28 months, with referral to an EI programme only at a mean age of 31 months (132).

10.5.2.2 Developmental Screening

In assessing developmental screening practices in South Africa, only research on screening for the early detection of hearing loss and retinopathy of prematurity (ROP) was found (116, 133). These research reports noted that the vast majority of public sector hospitals in South Africa do not provide screening services. More than 80% of eligible premature babies were not examined for ROP and more than 90% of babies were not assessed for early detection of hearing loss.

Varughese, Gilbert, Pieper and Cook (116) estimated at least 260-300 babies were at risk for ROP each year in South Africa, with at least half of whom would become blind without treatment. Paediatricians in their study reported that they were not seeing blind babies coming back for follow-up after NICU discharge, but a study of children in blind schools in South Africa showed that ROP accounted for 10.6% of blindess. This clearly shows how ineffective screening practices result in lack of intervention and poor outcomes. Similarly, Swanepoel, Ebrahim, Joseph and Friedland (115) reported less than 7% of babies requiring hearing aids received them before 6 months of age, loosing critical time for improving language outcomes in the population. Both of these studies were published recently (2007 and 2008) showing a serious lack in implementation of screening programmes despite the government's commitment to the early identification of disabilities in 1997 (134).

Lack of equipment and staff shortages were the most frequently reported reasons for not having screening programmes. A lack of funding cannot be entirely to blame however as a similar lack of screening programmes

for hearing difficulties was noted in the South African private sector hospitals (115). Inconsistencies in the services provided, the type of screening used and the population groups targeted may well be due in part to lack of legislation or formal policies from the Department of Health and Government. Screening programmes have become part of standard childcare in other countries through definitive actions from policymakers. For example in North Carolina the states Medicaid programme has the right to refuse payment for a well-child visit unless the visit includes a developmental screen (62).

10.5.2.3 Resources

The dire situation of under-detection and under-treatment of children in South Africa was highlighted in a 2007 study looking at services for children in need of rehabilitation in a peri-urban township of South Africa. Only a quarter of children in need of rehabilitation actually received it and nearly half of all children with motor or intellectual impairments were not attending school (135).

In South Africa where resources are limited, rehabilitation can seem less of a priority than other essential health services. Properly allocating available resources may result in improved ability to provide rehabilitation services to South African children however, without increasing the financial burden on the country. A simple example of this is that the cost to society of looking after one blind child for life could have covered the costs of a significant amount of preventative screening and intervention programmes. (116)

The long-term implications of missed opportunities for EI need to be considered. The cost of childhood disability extend past the child and immediate family to affect the community and society as a whole. Children who do not reach their full potential are less likely to be productive adults. Poor levels of cognition and education are linked to lower earnings, negatively impacting the next generation. Improved education is linked to lower teenage pregnancy rates and improved outcomes for infants, which include improved health, nutrition, cognition and education. The total cost to society of poor early childhood development therefore needs to include the costs born by subsequent generations, before being weighed up against the immediate cost of early intervention (40).

10.5.2.4 Recommendations

Because the effect of interventions on the infant and child may only be realised in adulthood, long-term neurodevelopmental monitoring of children is essential to assess outcomes of interventions and inform policy. Without long-term monitoring of children, needs cannot be accurately identified and 'best practice' clinical guidelines cannot be determined (119, 136).

There is considerable empirical evidence that parent-administered questionnaires are an effective developmental monitoring tool (4, 57, 137). They have been shown to be as accurate as physician-administered screening tools in identifying delayed or disordered development. They are brief to administer and relatively inexpensive, making them practical to implement at regular intervals.

Early detection using standardised parent-administered screening tools has become an important health care strategy in high-income countries (119). In the South African context parent-administered questionnaires could be a practical and affordable solution to implementing more structured screening and monitoring, comparable with international standards.

> "*Unfortunately, death is the tip of the iceberg… failure of children to fulfill their developmental potential and achieve satisfactory educational levels plays an important part in the intergenerational transmission of poverty … In view of the high cost of poor child development, both economically and in terms of equity and individual well-being, and the availability of effective interventions, we can no longer justify inactivity."* (40)

10.6 Summary

The importance of early identification to facilitate early intervention, research and policy is highlighted, with the use of parent-administered questionnaires being well justified due to minimal costs and a strong family-centered approach, affording long term benefits to the child, family and society.

The PEDS COMBINED and ASQ are good quality parent questionnaires that have acceptable psychometric properties and are frequently referenced and recommended for use in the literature on developmental screening. Using these screening tools in a population of infants who have required neonatal intensive care can be expected to produce a substantially greater number of positive screens (30% - 70%) than using them in the general population (10%). Information on how well the two tests concur and on their use in the South African context is inadequate and further research into these areas is needed.

Information on developmental screening practices in South Africa by paediatricians in the private sector over the past 10 years is sparse. Considering the large body of scientific literature available on this topic internationally, investigation into the screening practices of South African paediatricians is warranted.

11 Aims & Objectives

The objective of this study is to look at the concordance between the ASQ, PEDS COMBINED and the PSA in the early detection of developmental delay in a group of high-risk infants in the private health care sector of Gauteng, South Africa.

This study does not attempt to determine which method is more psychometrically sound or accurate in detecting developmental delay and is not an attempt to standardize these screens for the South African population. The aim of this study is to add to the growing body of literature on parent-administered questionnaires and to identify some of the trends of pediatricians in Gauteng, South Africa in screening for developmental delay

> "*It is unlikely that randomized, controlled trials will clarify a single best tool or method for efficient and effective screening for all practitioners. Rather, the cumulative experiences shared across regions, practice settings, and with varying tools inform providers of the feasibility and effectiveness of the range of methods available for transforming developmental screening into an evidence-based practice."* (111)

12 Rationale and Research Questions

The ASQ and PEDS are two parent-administered questionnaires that are commonly recommended for use in the detection of developmental delay with favourably reported psychometric properties. They should therefore have a high convergent validity and concur on which infants or children in an assessed group are at risk for developmental delays. In September 2009, the first study to look at the concordance between the ASQ and PEDS was published (112). The data for the study were part of a larger study, not originally intended to compare the two questionnaires. The study found only moderate agreement and significant discordance between the ASQ and PEDS and called for further research to better understand the uses, limitations and performance of these two questionnaires.

The ASQ and PEDS COMBINED (using the PEDS:DM Assessment Level Version) are recommended for screening high-risk population groups, including infants in NICU follow-up programmes. Infants with perinatal complications requiring admission to a neonatal unit are at higher risk for developmental delay with 44% - 76% requiring special education by school-age compared with 10% of the general population (26, 66). A population of high-risk infants who had required NICU care was therefore chosen to increase the likelihood of positive screens in this study.

There have also been no papers published on the methods and ability of paediatricians in private practice in South Africa to detect developmental delay. The knowledge and use of standardised parent-administered screening tools used by these paediatricians is not known.

The current study is therefore a comparison of 3 different methods of screening for developmental delay in at-risk infants in the private health care sector of the southern Gauteng region of South Africa. It compares recommendations from the ASQ, PEDS COMBINED and paediatricians' subjective assessment (PSA) practices for a cohort of at-risk infants from neonatal intensive care units in this region. This study will also identify trends in developmental screening practices and in the knowledge and use of parent-administered screening tools in a diverse group of paediatricans working in the southern Gauteng region of South Africa.

The following hypotheses will be tested:

1. The number of infants identified with developmental concerns by the PEDS COMBINED and ASQ will be greater than that identified by the PSA.
2. The ASQ and PEDS COMBINED will have good agreement, better than that expected by chance
3. The PSA will have poor agreement with both the ASQ and PEDS COMBINED, less than expected by chance
4. Standardised parent-completed questionnaires are not routinely used by paediatricians in private practice in the southern Gauteng region of South Africa at present.

13 Methods

13.1 Population

This study used an in-subject design with each infant being it's own control. Subjects were selected through non-random criterion sampling using a single cluster design. Clusters included all private hospitals in the southern Gauteng region with a full neonatal intensive care unit. Criteria for selection of infants included all infants in these clusters who required more than 3 days care in the NICU post-partum and whose caregiver filled in a consent form. The criterion for paediatricians included any paediatrician who was identified by the infant's caregiver as the primary paediatrician. (Table 4)

All private hospitals in the Southern Gauteng region that support a neonatal intensive care unit (NICU) were contacted for permision to approach caregivers of infants in the hosptial's NICU. Signed consent was given by the hospital managers and either the NICU managers or paediatricians working in the specific units, depending on the individual hospital's policies. Following consent, the hospital was visited between four and six times at random from October 2007 until June 2008. At each visit, nursing staff were informed about the study and asked to hand out consent forms to caregivers. Posters describing the study with consent forms attached were also left in each of the neonatal units. All caregivers present in the unit at the time of the visit were approached, informed about the study and given an option to fill in a consent form. (Table 4)

The 6 months corrected age (6mnth CA date) was calculated using the formula recommended in the PEDS:DM professional's Manual

> *"subtract 1 month from chronological age if the child was 3 1/2 - 6 weeks premature, 2 months if 7 – 10 weeks premature, 3 months if 11 to 14 weeks premature, and 4 months if 15 – 18 weeks premature*" (104)

Caregivers were contacted to confirm their postal address approximately six weeks before the baby's 6mnth CA date. Questionnaires were posted two weeks later or faxed six weeks later with prepaid envelopes for returning

the completed questionnaires, except where the postal address could not be confirmed. Caregivers were reminded on two separate occassions by fax, sms or email to return completed questionnaires and were given the option to withdraw from the study at any time.

Questionnaires that were returned by the caregiver contained more detailed information about the infant's medical and developmental history. Those infants with reported genetic syndromes were excluded from the study, as were those infants who had spent less than 3 days in the NICU. The infants' chronological and corrected age at the time the questionnaire was completed were calculated by subtracting the birth date from the date of testing and then correcting for prematurity as recommended by the author of the PEDS / PEDS:DM (104). Returned questionnaires were scored according to the infant's corrected age and feedback was given to the caregivers via an email or faxed letter.

Returned questionnaires included a signed consent for the researcher to contact the infant's paediatrican. Details of their paediatrician were supplied by the caregiver; including the paediatrican's name, telephone number, date of their last visit and date of their next planned visit. Paediatricans identified in this way were informed about the study. They were asked to complete a consent form and fill out a short questionnaire regarding their knowledge of parent-administered questionnaires and their developmental screening and referral practices. The paediatrician was then asked to complete a form indicating any developmental concerns for their specific patient. This was in the form of a checklist that could be filled in and returned by fax or email, or the paediatrican could indicate their concerns telephonically. The information obtained in this way was used to produce the paedaitrician's subjective assessment (PSA) for the purposes of this study.

Paediatricians were aware that their patients had filled in a parent-administered questionnaire, but were blinded to the recomendations from this questionnaire. To assist with follow-up of their patients they were emailed or faxed a short summary of recommendations from the questionnaries once the completed the PSA was recieved.

Table 4 Inclusion and exclusion criteria

Subjects:	Inclusion Criteria	Exclusion Criteria
Infants	* Ages between 5 months and 10 months * Must have required NICU care within the first day post-partum * Must have spent more than 3 days in the NICU	* Any infant with a genetic syndrome * Infants who spent less than 3 days in the NICU * Infants of caregivers who did not give consent
Paediatrician	* Identified by the caregivers of infants on the study as their infant's paediatrician	* Paediatricians who were not identified by one of the included infant's caregivers as being their paediatrician. * Paediatricians who did not give consent
Hospitals	*All private hospitals in Southern Gauteng with a fully functioning NICU	* Hospitals where the hospital management did not give consent

13.2 Measurements

(Appendix 18.3 – 18.7)

13.2.1 Ages and Stages Questionnaire (ASQ)

The ASQ (88) consists of a set of 19 questionnaires divided into the following age groups categorised by months 4,6,8,10,12, 14, 16, 18, 20, 22, 24, 27, 30, 33, 36, 42, 48, 54 and 60 months. The questionnaire matching the child's age is given to the care-giver, and requires the caregiver to respond 'yes', 'sometimes' or 'not yet' to a list of age appropriate activities. Thirty activities are divided into 5 developmental domains: Communication domain (Comm); Gross motor domain (GM); Fine Motor domain (FM); Problem solving domain (PS); Personal-Social domain (SE)

Caregiver responses are assigned points: 'yes' receiving 10, 'sometimes' 5 and 'not yet' 0. The points are added up in each of the five domains to give the domain scores, and then added together for a summary score. The domain scores and summary scores are compared to a derived screening cut-off score, recommended as 2 SD below the mean by the user's manual (88).

Seven open ended questions to elicit parental concerns are also included in the analysis of the questionnaires, but do not form part of the scored evaluation.

13.2.1.1 Rating the ASQ scores

(Table 5)

For the ASQ, a referral cut-off point of 2 SD below the mean for each domain is used to identify those babies who need further assessment. Children scoring above this are classified as age appropriate. The user's guide also recommends following-up infants whose caregivers have indicated in the open questions that the infant does not use two hands equally or does not stand on flat feet since these questions were specifically added to assist in detecting cerebral palsy (88).

For the purposes of this study the following scoring system was used (Table 5):

1- Any baby scoring at or more than 2 SD below the mean in a domain was given a rating of 1 for that domain. A rating of 1 was also given for the FM domain if a caregiver answered "*no*" to the questions about their infant's equal hand use and for the GM domain if the caregiver indicated the infant was toe-standing.

2- An arbitrary cut-off score of 1 SD below the mean was used to identify those babies that needed to be monitored more closely than normal. Any domain that was scored between 1 and 2 SD below the norm was given a rating of 2.

3- Any child scoring within the normal range and up to 1 SD below the norm was given a rating of 3.

13.2.2 PEDS COMBINED

The PEDS COMBINED consists of two questionnaires which can be used together, the PEDS and PEDS:DM.

The PEDS is a very simple tool, consisting of only ten questions which are designed to systematically elicit patental concerns in the domains of GM, FM, PS, SE, Comm (seperated into expressive and receptive language), as well as global and pre-school skills. Caregivers circle one of three provided options, namely '*yes'*, '*no'* or '*a little'*. These responses are then transferred to a scoring sheet which indicates which concerns are significant and predictive of developmental concerns according to the child's age, and recommends a course of action.

The PEDS:DM is a milestone checklist. It is designed to be used either as a screening tool or as an assessment level tool. The assessment level version uses the same items as the screening tool. It groups items by domains instead of ages and presents them as a continuous set of tasks so that items above and below the child's age can be scored. This results in the test being able to show a child's strengths and weaknesses. This also allows the user to determine age equivalents and percentage delay rather than just providing a cut-off score based on standard deviations below the mean. According to the author of the PEDS:DM the continuous item set allows for identification of emerging skills as well as mastered skills, which facilitates progress monitoring and is useful for programme evaluations and research studies. The PEDS:DM covers the same developmental domains as the PEDS.

The assessment level version was used for this study. It can be used continuously for infants up to 8 years and therefore contains many items not applicable to the age of the study population group. The author was contacted and gave permission via e-mail to restrict the item sets for this study.

13.2.2.1 Rating the PEDS COMBINED scores

(Table 5)

The PEDS COMBINED is designed to lead a practitioner down several paths requiring different actions depending on the scored results (104). These were summarized into one of three recommendations as shown in Table 5, with a resultant rating of:

1. Refer for in-depth assessment,
2. Monitor closely or
3. Age appropriate

13.2.3 Paediatricians

Paediatricians in this study were asked to rate the infants development based on their normal practice procedures and then to mark a checklist rating their opinion of the infant's development. The result was the PSA

13.2.3.1 Rating the PSA Scores

(Table 5)

The paediatricans were asked to rate the infant's development as,

1- Delayed - refer for in-depth assessment / intervention,

2- Concerned - monitor closely or

3- Not concerned at this time.

These were given the same rating of 1, 2 or 3 respectively for the purpose of this study (Table 5).

4- Infants could also receive a rating of 4 if the paediatrician had not seen the infant in more than four months and was therefore unable to comment on the infant's current development. This was determined if

   a. caregivers indicated on their forms that they had not seen their paediatrician for more than four months and had no follow-up visit planned, or

   b. the paediatrician reported that they had not seen the infant for more than four months.

13.2.4 Summary scores

The infant was determined to have failed (ie. They had a positive screen indicating a referral for further assessment was needed) if they failed any domain. This was recommended in the user manuals (88) and was also used by Sices, Stancin and Kirchner in their study comparing the ASQ and PEDS (112). In other words, they received a summary score of 1 if a rating of 1 was received in any of the domain scores. A summary score for each of the parent-completed questionnaires and for the PSA was therefore derived and determined by the lowest value of their respective domain scores (Table 5). A summary score was given as follows:

1- If the screen scored a 1 in any domain, it was given a summary rating of 1.

2- If the screen did not have 1's, but had a 2 in any domain, the summary rating was 2.

3- If the screen did not have 1s or 2s, the summary rating was 3

4- If the infant had not seen the paediatrician in more than 4 months and had no planned visits, the summary rating was 4

Table 5 Rating of Three Methods

Rating	ASQ	PEDS COMBINED	PSA
1	2 or more SD below mean OR toe-standing OR child not using hands equally well	Path A PEDS OR Fails 1 or more items on PEDS:DM	Delayed - refer for in-depth assessment / intervention,
2	Between 1 & 2 SD below mean	Path B, C or D on PEDS AND Passes all items on PEDS:DM	Concerned - monitor closely
3	Up to 1 SD below mean	Path E on PEDS AND Passes all items on PEDS:DM	Not concerned at this time
4	-	-	Unable to follow-up

13.3 Statistical Methods

Data was collected in clusters from each hospital and then analyzed at the individual infant level. To determine sufficient sample size we made the assumption (based on the literature) that in our high-risk group at 6 months corrected age 70% of infants should pass the screen (receive a rating of 3) (21). A sample size of at least 38 subjects would be required to have power in excess of 80% to detect excellent agreement ($\kappa \geq 0.8$) compared to the upper limit of poor agreement ($\kappa = 0.4$) when testing one sided at the 0.05 level of significance. When comparing the PEDS and ASQ, we had a sample size of 60 when comparing rating 1, 2 and 3; and a sample size of 43 when comparing rating 1 and 3 only. Our sample size was therefore sufficient to analyze the agreement between the PEDS and ASQ using the intraclass kappa coefficient. (Table 6)

In assuming a pass rate of 70%, a sample size of at least 23 subjects will have power in excess of 90% to detect poor agreement ($\kappa < 0.4$) compared to excellent agreement (($\kappa \geq 0.8$) when testing one sided at the 0.05 level of significance. When comparing the PSA rating and ASQ or PEDS COMBINED, we had a sample size of 32 and 35 respectively when for rating 1, 2 and 3; and a sample size of 24 and 27 for rating 1 and 3. Our sample

size was therefore sufficient to analyze the agreement between the paediatricians and each of the questionnaires using the intraclass kappa coefficient. (Table 6)

Table 6 Sample size calculations

	80% power to detect excellent agreement between questionnaires ($\kappa \geq 0.8$)		90% power to detect poor agreement between paediatrician and questionnaires ($\kappa < 0.4$)			
	ASQ:PEDS 1,2,3	ASQ:PEDS 1 & 3	ASQ:PSA 1,2,3	ASQ:PSA 1 & 3	PSA:PEDS 1,2,3	PSA:PEDS 1 & 3
Sample size required	38	38	23	23	23	23
Actual sample	60	43	32	24	35	27

The data were analysed to compare if the screening methods identified the same infants as developmentally appropriate (rating 3), being at-risk for developmental delay (rating 2) or developmentally delayed (rating 1). Comparisons were made between:

1 - ASQ & PEDS COMBINED

2 - ASQ and PSA

3 - PSA and PEDS

The kappa coefficient (κ) was used to assess if the agreement between the two screening methods in each pair was greater than that expected by chance, with a coefficient of ≤ 0.4 indicating poor agreement, 0.4 to 0.75 moderate agreement and ≥ 0.75 excellent agreement (138). To analyze how the number of children who passed each screen compared, a test of symmetry was used. The test decision was determined by a chi-squared calculation with a ρ-value < 0.05 indicating statistical significance. (Appendix 18.8.1)

The data were then analysed excluding the rating 2 as the ASQ is designed to screen positive or negative (rating 1 or rating 3). A rating of 2 was derived for this study in an attempt to compare the ASQ in a more similar manner with the PEDS COMBINED, but which had not been validated for the ASQ. A rating of 2 was given to an infant who was not age appropriate, but also not significantly delayed, with recommendations for closer monitoring, but not for a more in-depth assessment. To compare the ability of the thress screening methods to differentiate between those infants who should be referred for an in-depth developmental assessment and those

developing appropriately, the rating of 2 was removed from the analysis, as it did not fall into either category. Sices et al. (112) compared the ASQ and the PEDS in the same way.

If an infant had not been seen by their paediatrician for more than 4 months, a rating of 4 was given to the PSA. Data where infants scored a 4 on the PSA were not used in calcultions and analysis of kappa or symmetry when comparing the PSA to the questionnaires but were used in the descriptive analysis of results.

13.4 Ethical Considerations

The study was approved by the Human Research Ethics Committee (Medical)- Protocol M070416 (Appendix 18.1). Consent forms were obtained from participating hospital managers and NICU managers as well as all parents and of infants participating in the study. Consent forms were also obtained from paediatricians participating in the study. All participants were given the option to withdraw from the study at any time without consequence. (Appendix 18.2)

Parents were provided with contact details of the researcher in order to answer any questions or address any anxiety that may have occurred as a result of participating in this study. Parents received feedback in the form of a letter and were directed to their paediatrician to assist with any follow-up of recommendations from the screening. Paediatricians were provided with a summary of screening results to facilitate follow-up of any concerns raised by the questionnaires.

Hosptials, paediatricians' and infant-caregiver pair details were coded to ensure confidentiality. Caregivers were asked to use a specific code on the questionnaires and not to put their infant's names or other identifying information. Names and contact details were kept in a separate file, locked with an alphanumeric pin code.

No funding or financial support was received for this study.

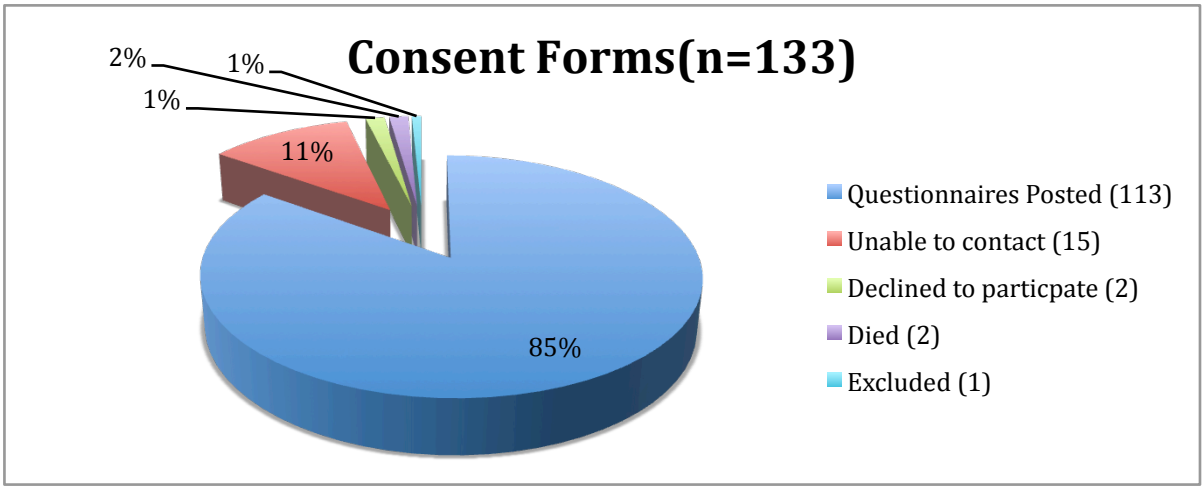
14 Results

14.1 Descriptive Analysis

14.1.1 Questionnaires

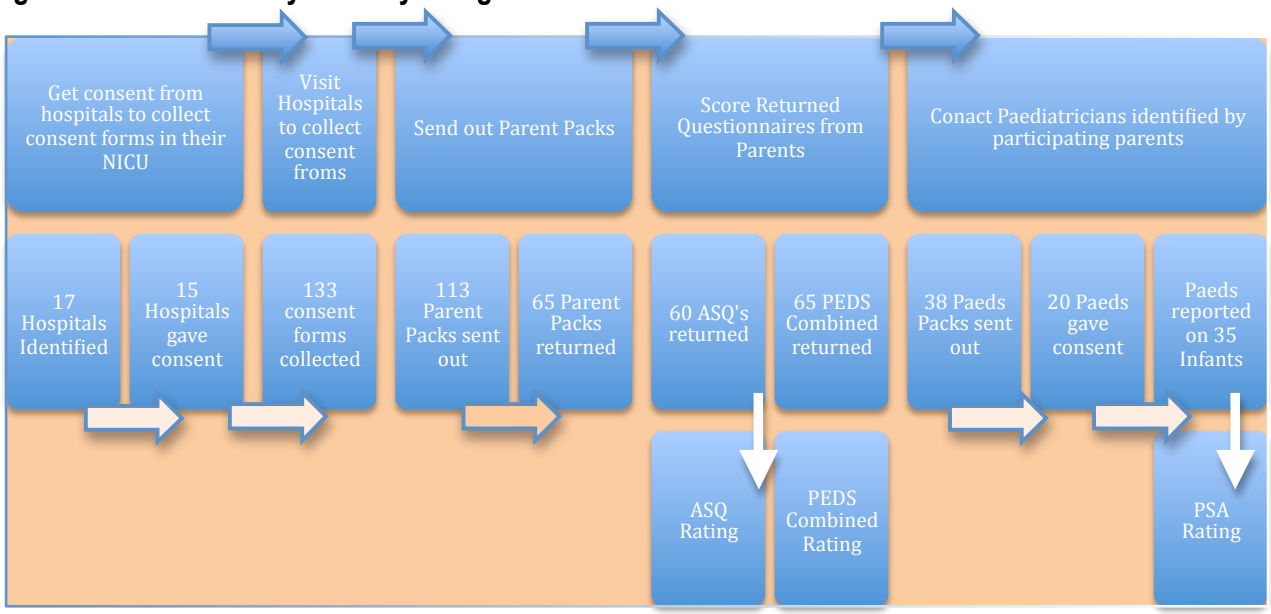
A total of 17 hospitals were identified as appropriate for this study and contacted for permisson to recruit subjects in their NICU's. Management from 15 hospitals gave consent and each of these hospitals was visited at random between 4 and 6 times by the researcher. At the end of the collection period, an average of 4 consent forms were obtained from each hospital (minimum: 0, maximum:16, SD: 5). This resulted in consent forms being obtained for 133 infants. Of these, 68 caregivers did not return questionnaires. Fifteeen caregivers could not be contacted and therefore were not sent questionnaires, 2 infants' caregivers declined to participate in the study, 2 infants had died and 1 infant was excluded from the study because of Down Syndrome. This resulted in 113 questionnaires being posted, of which caregivers returned 65 (57.5 %). (Figure 1)

Figure 1 Consent Forms Received



Of the 65 questionnaires that were returned, a total 60 ASQ forms were received (92%) and 65 PEDS COMBINED forms were received (100%). A total of 38 paediatricians were identified on the returned questionnaires and were contacted by the researcher. Of these 38 paediatricians 24 consented to particpate in the study, completing a total of 35 PSAs (54% of the 65 questionnaires returned). An average of two PSAs were completed per paediatrician (minimum: 1, maximum: 6, SD: 2). (Figure 2)

Figure 2 Summary of Study Design



There were 30 infants for whom no paediatrician's opinion was available. Of these 30 infants,16 (25%) did not receive a paediatrician's opinion because the paediatrician did not participate in the study, whilst the other 14 (21%) infants had not seen their paediatrician within the required time frame and thererfore received a PSA rating of 4. These infant's caregivers most often reported that they had not been back to the paediatrician due to financial restraints or because they did not see the necessity of a well-baby follow-up. Of these 14 infants who were not seen by a paediatrician for follow-up, ten were identified by the parent-administered questionnaires as having significant developmental delays. Infants who received a rating of 4 showed the same demographic variables as the rest of the studies cohort - they were mostly white, lower risk NICU infants and did not cluster around a particular hospital or paediatrician.

14.1.2 Patient Characteristics

Population variables of the cohort are presented in Table 7. Infants were mostly white, lower-risk premature infants.The average age of the infants when the ASQ and PEDS COMBINED was completed was the same as the average age of the infants when the PSA was completed (6 months, SD:1 and 6 months, SD:2 respectively).

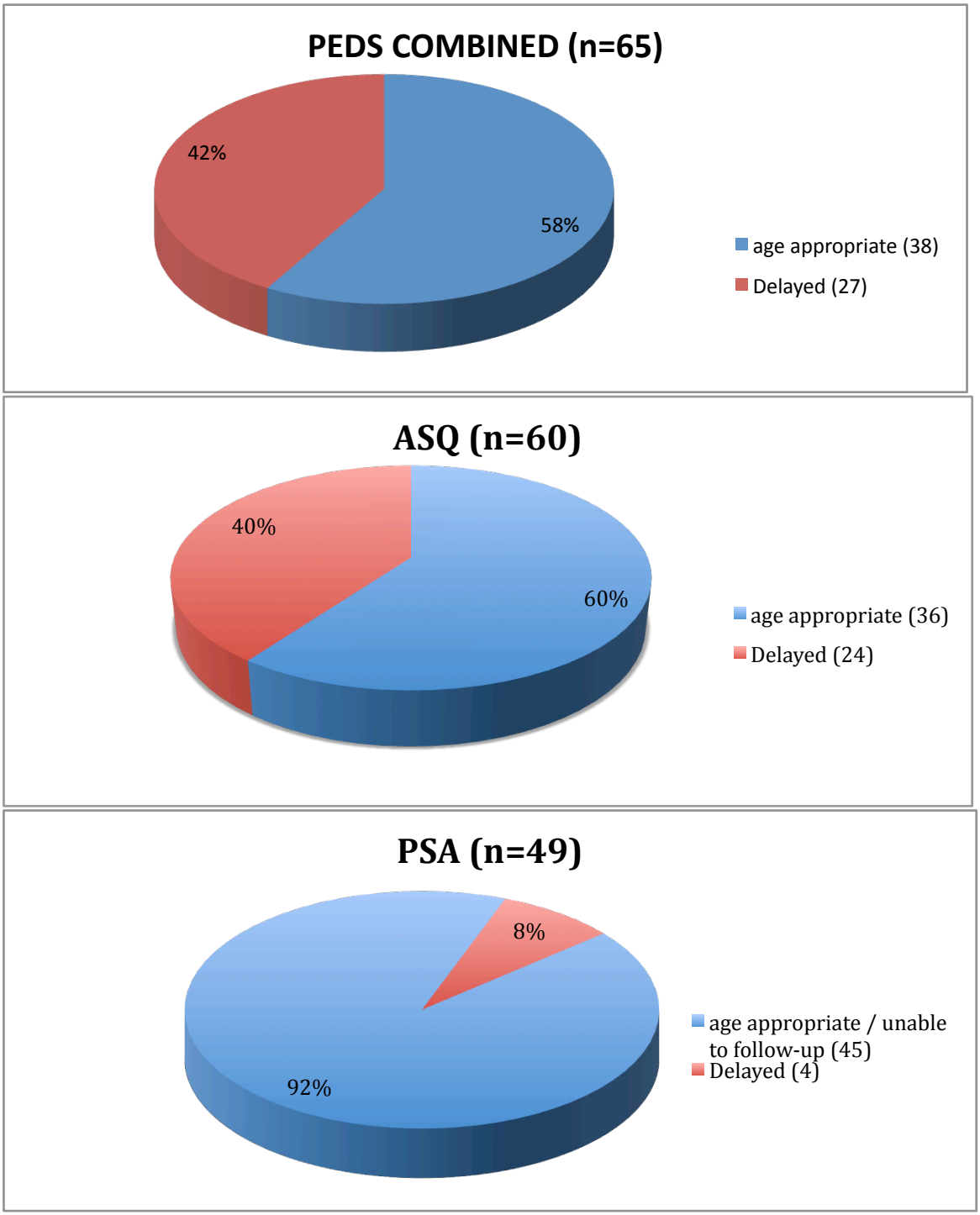
Table 7 Patient Characteristics

Patient Characteristics			
Total Number (n)		65	%
Birth weight (g)	mean ± SD	1796 ± 825	
	range	490 - 4270	
Gestational age (weeks)	mean ± SD	34 ± 3.4	
	range	26 - 42	
Sex (n)	male	27	42%
	female	32	49%
	unknown	6	9%
Race (n)	White	51	79%
	Black	10	15%
	Indian	4	6%
Reason for NICU Admissions (n)	Low birth weight / Prematurity	51	79%
	Respiratory Complications	11	17%
	Neonatal Infections	2	3%
	Congenital Abnormalities	1	2%
Number ventilated (n)		36	55%
Length of NICU Stay (days)	mean ± SD	27 ± 31	
	range	4 -131	
Age when questionnaire completed (months)	mean ± SD	6 ± 1	
	range	6 -10	
Age at paediatrician visit (months)	mean ± SD	6 ± 2	
	range	5 - 9	

14.1.3 Identification of Developmental Concerns

Of the total group of 65 infants whose caregiver's completed questionnaires, 6% (4) were identified with developmental concerns by the PSA. The ASQ identified 37% (24) and the PEDS COMBINED identified 42% (27) of the infants as having developmental concerns requiring further developmental assessment. (Figure 3)

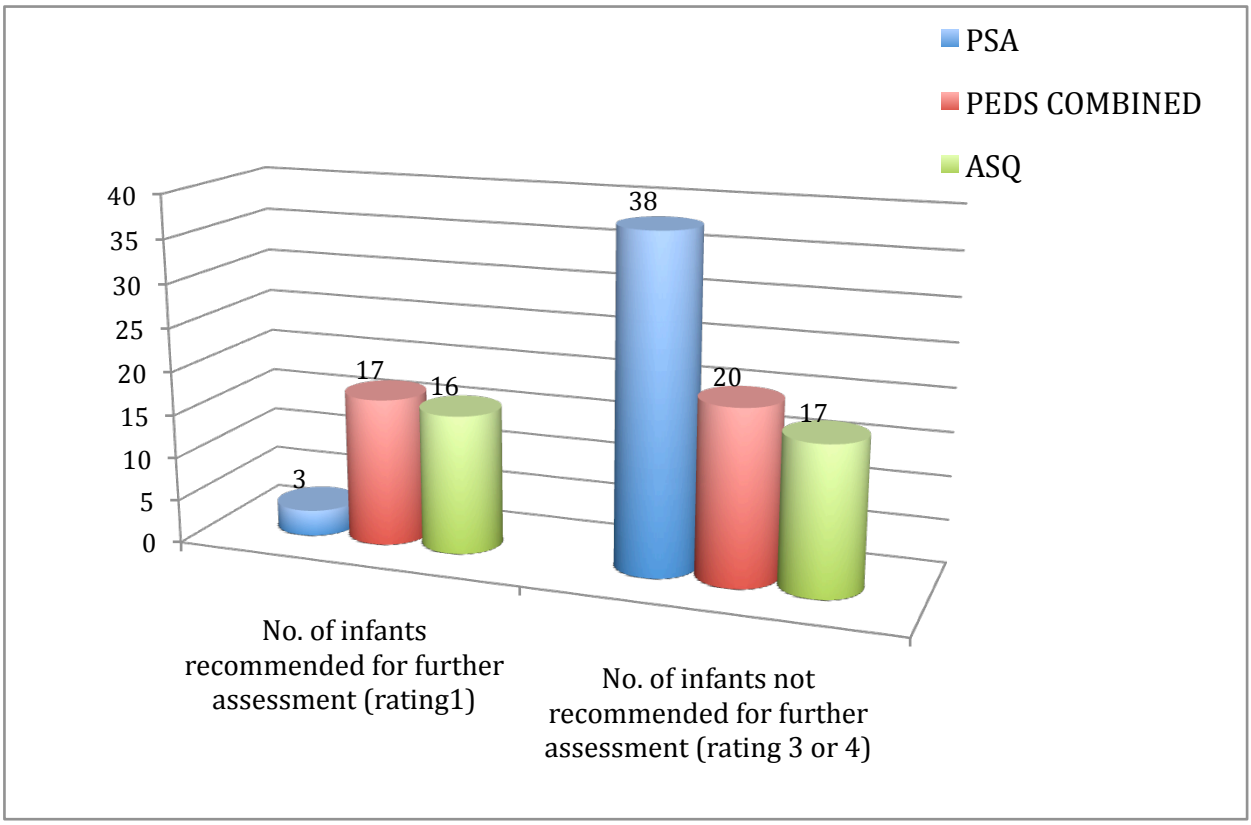
Figure 3 Identification of infants requiring in-depth developmental assessment



In total 44 of the 65 infant's (68%) had all three screening methods returned. In this group all infants had an ASQ and PEDS COMBINED and infants either had a PSA from the paediatrician or it was reported that they had not seen the paediatrician for more than 4 months. Of the 44 infants in this group, up to 38% were identified with

developmental concerns by the questionnaires, whereas only 6.8% were identified with developmental concerns by the paediatricians. (Figure 4)

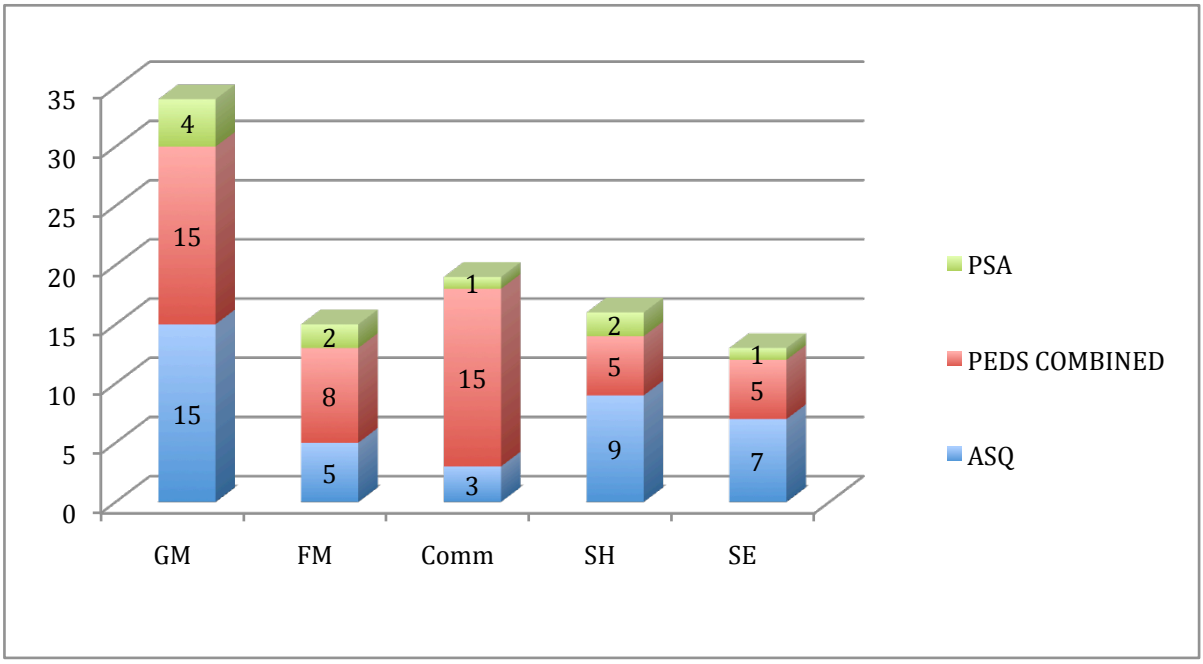
Figure 4 Comparison of recommendations by three screening methods



In the total group of 65 infants for whom questionnaires were received, gross motor concerns were the most frequently identified concerns by all 3 screening methods. The PEDS COMBINED also identified a higher number of children with communication concerns than the other screening methods. The paediatricians identified the least number of concerns in all domains . (Figure 5)

Figure 5 Number of concerns per developmental domain (rating 1)

(GM=Gross Motor, FM=Fine Motor, Comm = Communication, SH=Self Help, SE = Social Emotional)



14.1.4 Referral Practices of Paediatricians

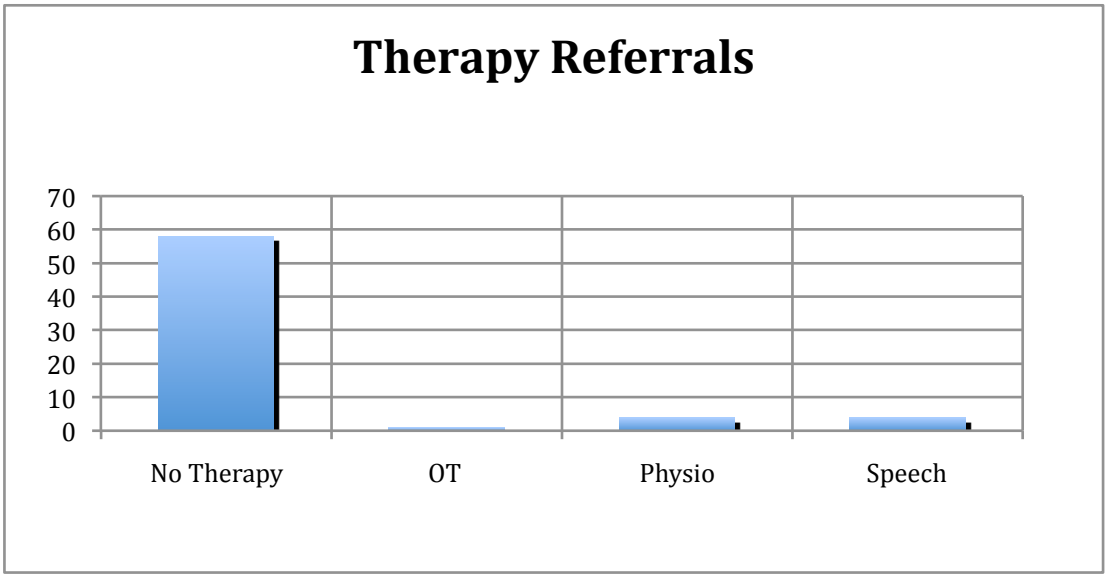
Paediatricans were selected for this study if they were identified by the caregiver as the infant's primary paediatrician. These paediatricians were asked to complete a short questionnaire describing their screening and referral practices and a total of 23 questionnaires were returned. Of these, the percentage of paediatricians who indicated that they would commonly refer babies under 12 months old with a non-specific, mild developmental delay for physiotherapy was 82.6%, for speech was 47.8% and occupational therapy was 39%.

Only three paediatricians indicated that they based their referrals on specific guidelines whereas all of the paediatricians indicated that they based their referrals on clinical judgment and experience. More than half of the paediatricians, (52%) felt that caregivers were unable to identify developmental delay in their own babies whilst 13% felt that some caregivers were able to and some were not. Less than 35% indicated that they thoughT caregivers were able to identify developmental delay in their own babies.

Knowledge about parent-administered questionnaires for developmental screening was limited amongst the paediatricians, with only five paediatricians (21%) indicating any knowledge about parent-administered questionnaires, and none of the paediatricians surveyed able to actually name the ASQ or PEDS. No paediatrician surveyed had experience using parent-administered questionnaires in their practice.

Of the 65 infants on the study, seven had been for therapy (11%). Of these, two went for both physiotherapy and speech therapy, two had only physiotherapy, two had only speech therapy and one had occupational therapy. Information on who had initiated the referral was not available. (Figure 6)

Figure 6 Therapy Referrals



14.2 Comparitive Analysis

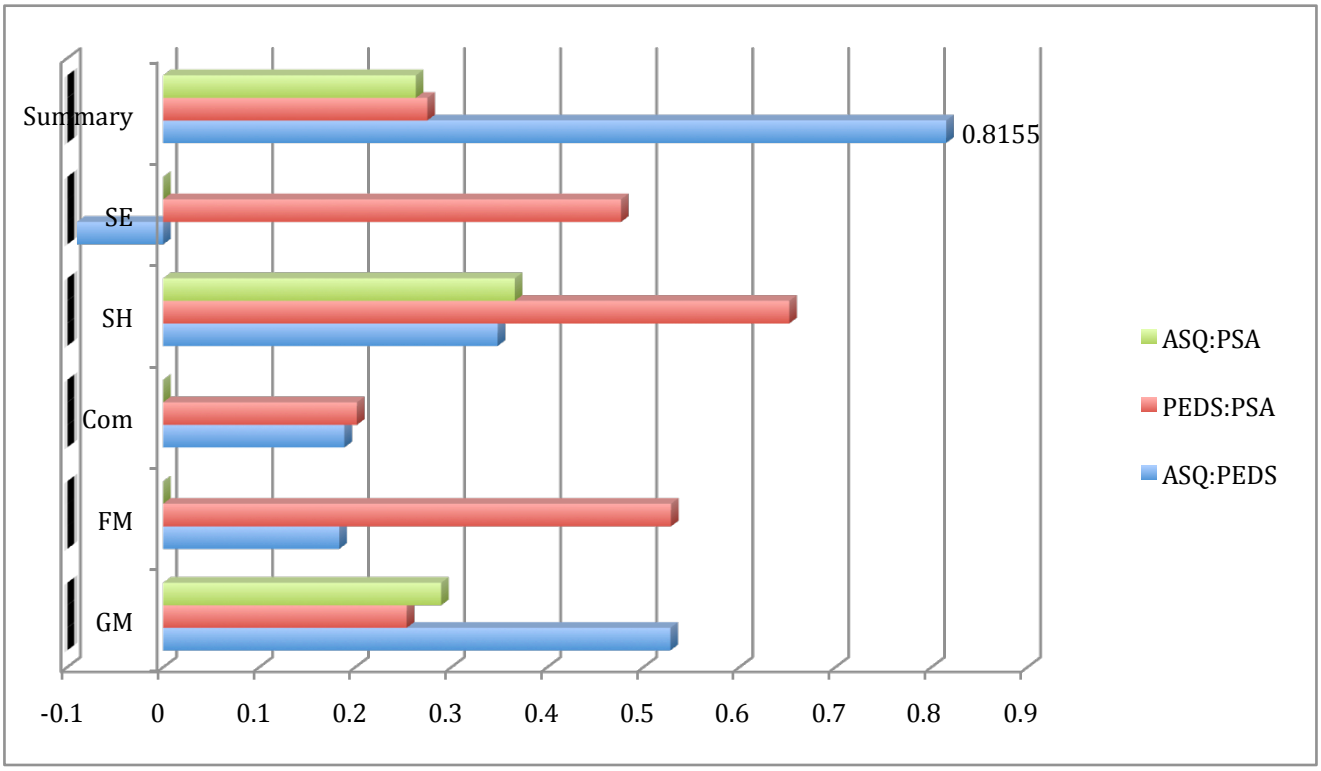
14.2.1 Comparison of rating 1 & 2

The ASQ and PEDS COMBINED showed excellent interrater agreement, agreeing on which infants required further developmental assessment (Figure 7). When there was discordance it was not random. Discordance occurred in the direction of the ASQ being more likely to identify an infant as delayed, except in the domain of communication where the PEDS COMBINED was more sensitive than the ASQ. The PSA was more likely to identify infants as age appropriate across all domains and for overall development, and therefore the least likely

to recommend further developmental assessment or intervention for an infant (Table 8). The PSA showed poor agreement with both of the standardised parent-administered screening tools overall in identifying infants as delayed (Figure 7). These results are described in more detail below.

Figure 7 Kappa comparisons of Rating 1 & 3

(GM=Gross Motor, FM=Fine Motor, Comm = Communication, SH=Self Help, SE = Social Emotional)



14.2.1.1 Summary Scores

(Table 8)

The concordance between the ASQ and PEDS COMBINED expected by chance was 49.8%. The actual concordance between the two questionnaires was 90.7% (κ = 0.82, p = 0.05). The excellent agreement between the two questionnaires was therefore statistically significant and significantly greater than that expected by chance. When there was disagreement between the two questionnaires, the discordance of 9.3% was in the direction of the ASQ rating an infant as 1 when the PEDS COMBINED rated the same infant as 3 (ρ = 0.05)

The concordance between the PEDS COMBINED and PSA was poor and statistically significant (κ = 0.28, ρ = 0.03). Discordance was significantly in favour of the PEDS COMBINED identifying the infant as delayed when the PSA identified the infant as age-appropriate.

The concordance between the ASQ and the PSA was poor and statistically significant (κ = 0.26, ρ = 0.01). Discordance was significantly in favour of the paediatrican identifying the infant as age appropriate whilst the ASQ identified the same infant as needing further assessment.

14.2.1.2 *Domain Scores*

(Table 8)

For the GM domain, there was moderate agreement between the ASQ and PEDS COMBINED (κ = 0.66) and poor agreement between the PSA and both questionnaires (κ < 0.4). Discordance in this domain was not significant.

For the communication domain, poor agreement between the ASQ and PEDS COMBINED was significant (ρ = 0.01, κ = 0.19), with the ASQ being more likely to identify an infant as age appropriate whilst the PEDS COMBINED identified the same infant as requiring further assessment. Poor agreement between the PEDS COMBINED and the PSA was also significant for communication (ρ = 0.01 & κ = 0.20). Discordance was directional with the PEDS COMBINED more likely to recommend further assessment when the PSA identified the infant as age appropriate. The ASQ and PSA showed poor agreement, which was not statistically significant or directional.

For SH, SE and FM domains there was moderate agreement between the PSA and PEDS COMBINED which was not significant (ρ > 0.05) with random discordance. In other words the agreement was not different from that expected by chance. When the PSA and PEDS COMBINED disagreed, the disagreement was not in any

direction. There was also poor agreement and random discordance in these domains when comparing the ASQ to the PEDS COMBINED and PSA. The negative kappa score in the SE domain was not significant.

Table 8 Comparison of rating 1 & 3

(GM=Gross Motor, FM=Fine Motor, Comm = Communication, SH=Self Help, SE = Social Emotional)

Rating 1,3	Kappa	Symmetry : Summary	Symmetry: Domain
ASQ: PEDS COMBINED	Excellent agreement for summary (κ= 0.82) Moderate agreement for GM domain (κ= 0.53)	Borderline significant discordance is directional with a trend towards PEDS COMBINED being more likely to score a 3 (ρ = 0.05)	Statistically significant discordance is directional for Comm with ASQ more likely to score a 3 (ρ = 0.01) All other domains show random discrepancies with no statistical significance
Paed:ASQ	Poor agreement for summary (κ = 0.26) and poor agreement across all domains	Statistically significant discordance is directional with PSA more likely to score 3 (ρ=0.01)	Random discrepancies for all domains with no statistical significance
Paed: PEDS COMBINED	Poor agreement for summary (κ= 0.28) Moderate agreement for domains of: SH (κ=0.65) FM (κ=0.53) SE (κ=0.48)	Statistically significant discordance is directional with PSA more likely to score 3 (ρ=0.03)	Statistically significant discordance is directional for Comm with PSA more likely to score 3 (ρ=0.03) All other domains show random discrepancies with no statistical significance

14.2.2 Comparison of rating 1, 2 & 3

(Figure 8, Table 9)

Concordance was significantly reduced when infants who had received a rating of 2 were included in the statistical analysis, particularly when comparing domain scores (κ < 0.04). Although reduced, for the summary scores agreement was still fair between the ASQ and PEDS COMBINED and still statistically significant (κ = 0.58, ρ = 0.02). Poor agreement between the PSA and questionnaires for summary scores remained (κ < 0.26) and was still statistically significant for the ASQ (ρ = 0.02). ASQ remained the most sensitive screening tool overall, with PEDS COMBINED still the most sensitive for Comm concerns and the PSA the least likely to identify concerns.

Figure 8 Kappa comparison of Ratings 1,2 & 3

(GM=Gross Motor, FM=Fine Motor, Comm = Communication, SH=Self Help, SE = Social Emotional)

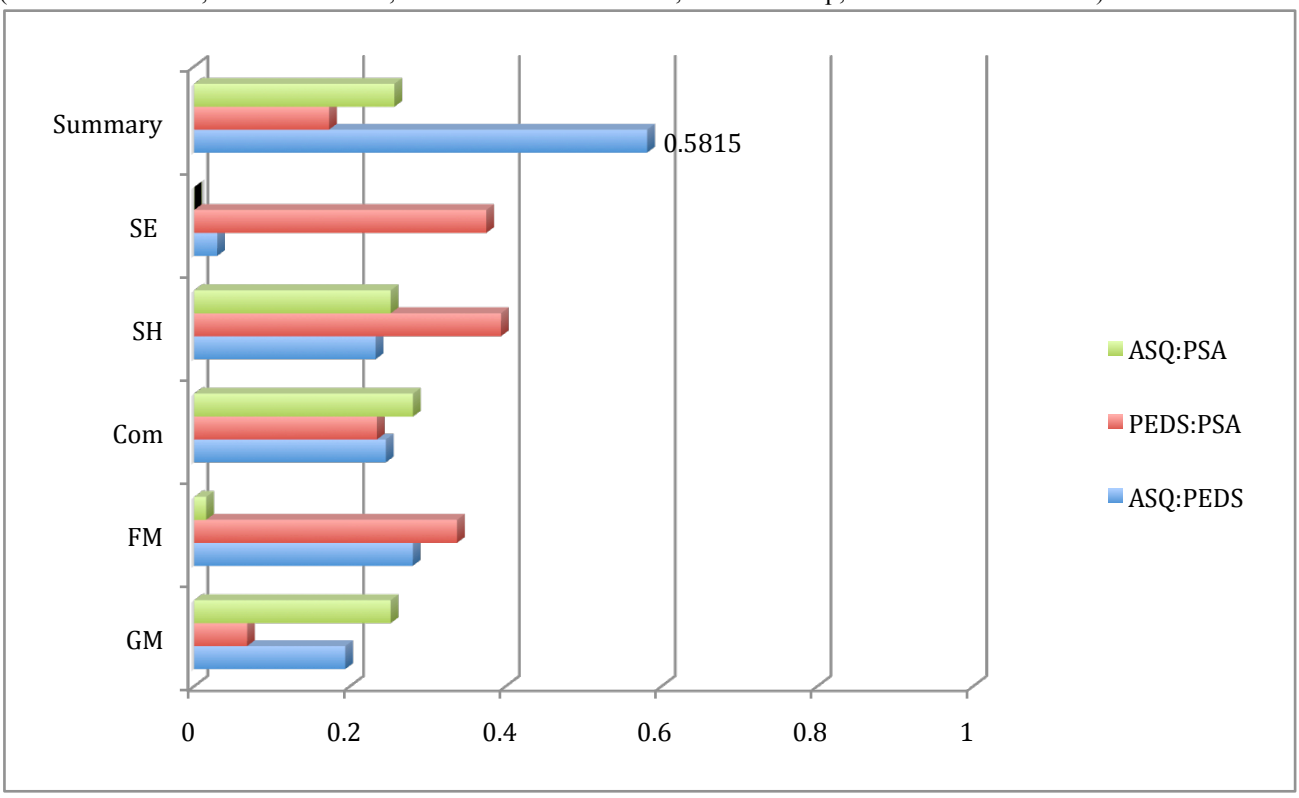


Table 9 Comparison of Rating of 1,2 & 3

(GM=Gross Motor, FM=Fine Motor, Comm = Communication, SH=Self Help, SE = Social Emotional)

Rating 1,2,3	Kappa	Symmetry: Summary	Symmetry: Domain
ASQ: PEDS COMBINED	Moderate agreement for summary (κ=0.58)	Statistically significant discordance is directional with PEDS COMBINED being more likely to score a 3 (ρ = 0.03)	Statisticallysignificant discordance is directional for FM & Comm. PEDS COMBINED more likely to score 3 for fine motor (ρ=0.04) and ASQ more likely to score a 3 for communication (ρ=0.00)
PSA:ASQ	Poor agreement for summary (κ=0.26) and across all domains	Statistically significant discordance is directional with PSA more likely to score 3 (ρ=0.02)	Random discrepancies with no statistical significance
PSA: PEDS COMBINED	Poor agreement for summary (κ=0.17) and across all domains	Not significant but trend towards PSA more likely to score a 3	Statistically significant discordance is directional for Comm (ρ=0.01) with PSA more likely to score 3

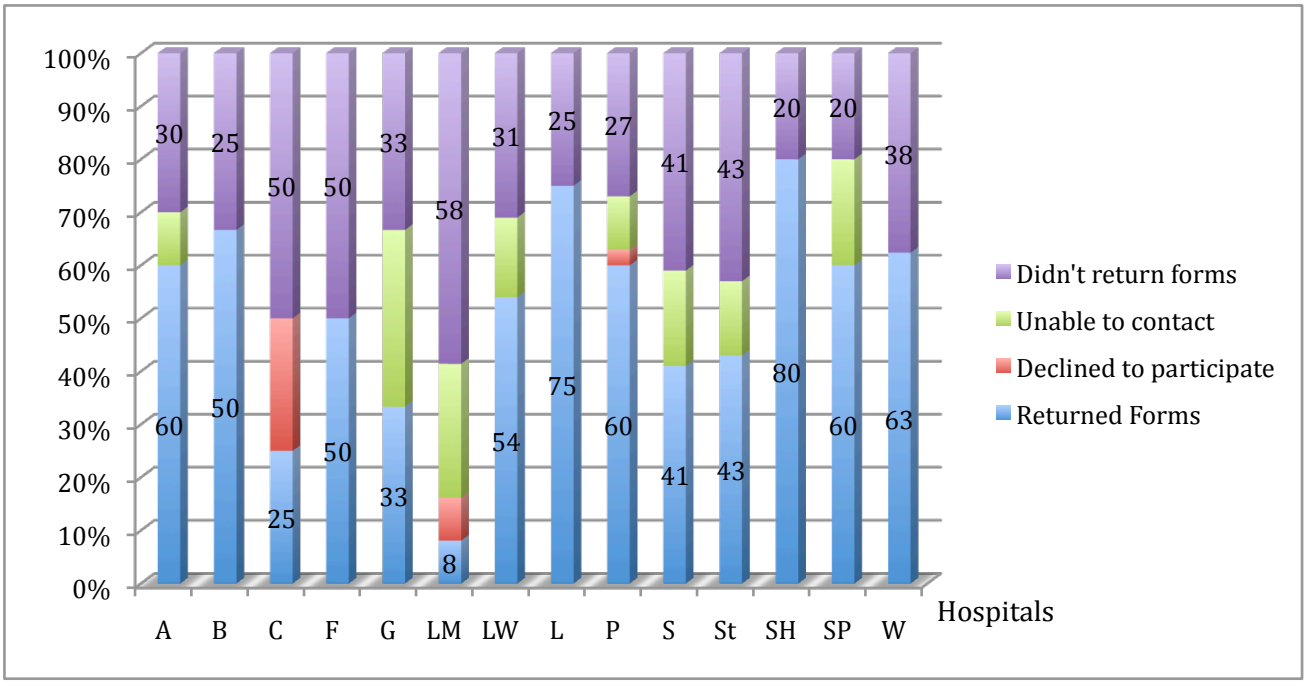
15 Discussion

15.1 Response rate of caregivers and questionnaire bias

The 57,5% return rate of questionnaires for this study may have resulted in non-respondant bias. According to the litrature, mail surveys have a mean response rate of 60%. This response is only slightly lower than the 60% and therefore indicates an acceptabe response rate (139). Of the 48 (42.5%) questionnaires not returned, non-respondents were spread fairly equally throughout the different hospitals. An average of 35% of questionnaires were lost to follow-up per hospital cluster, decreasing the likelihood of one hospital cluster of biasing the results. (Figure 9)

Figure 9 Percentage of Questionnaires Returned by Hospital

(one hospital was left out as there was only one consent form received, which was returned - giving 100% return rate)



The return rate rate in this study is also slightly higher than the response rate of 54% in the study reported by Hix-Small, Marks, Squires and Nickel (101) which looked at the implementation of the ASQ into a paediatric practice. In the Hix-Small study 13 demographic factors were assessed in non-respondents and non-respondent bias was found to be of no significance aside from two factors. A higher number of non-respondents were on Medicaid and were younger parents, suggesting that younger parents from lower income groups were less likely

to respond. In the current study all subjects were recruited from the private health care sector, indicating a higher economic status, however information on actual economic status and maternal age at birth were not obtained.

The fact that socio-economic data was not available in this study may be regarded as a limiting factor, but the ASQ and PEDS are recommended for use in infants from both high and low socio-economic groups (88, 101). Other studies have also reported that the accuracy of parental reporting on infants developmenal status is not influenced by sociodemographic factors or parental educational level (140, 141). The possibility that many of the non-respondents were younger mothers or from lower economic groups may make it less accurate to extrapolate the results from the study into lower SES groups in South Africa but does not affect the analysis of concordance between the screening methods of questionnaires that were returned. This study does not intend to address the validity of the questionnaires, but rather to identify how well they concur with each other and with the paediatrician's subjective assessment. For this study therefore, each infant served as it's own control and the agreement between the ASQ, PEDS COMBINED and PSA was analyzed at the level of the infant. Caregiver variables did not affect analysis.

A further limitation of this study is that the two questionnaires were posted to the caregivers in one package. Answering the one questionnaire could potentially have influenced the answers to the second questionnaire. The caregivers motivation for filling in forms was to determine the developmental level of their infant and therefore there was no incentive to answer questions incorrectly on either questionnaire. In the study by Sices et.al. (112) where the ASQ and PEDS were also filled out on the same day the agreement between the tests was no different to that expected by chance, indicating that completion of questionnaires on the same day does not increase concordance.

Further, the questionnaires are designed to be subjective and they ask different questions. The format of the questions is also different for each questionnaire. As an example, Table 10 compares the GM item set from the ASQ and PEDS:DM questionnaires:

Table 10 Comparison of Gross Motor Items on ASQ and PEDS:DM

Comparison of Gross Motor Items		
	ASQ – 6 month Questionnaire	**PEDS:DM – Assessment Level Version: 0-10 month items**
1	While on his back, does your baby lift his legs high enough to see his feet?	Does your baby try to keep his or her head steady?
2	When she is on her tummy, does your baby straighten both arms and push her whole chest off the bed or floor?	Does your baby roll from her back to her side?
3	Does your baby roll from his back to his tummy, getting both arms out from under him?	If your baby is lying on her back, can she pass a toy from one hand to the other?
4	When you put her on the floor, does your baby lean on her hands while sitting? (If she already sits up straight without leaning on her hands, check "yes" for this item.)	Can your baby get around on hands and knees or by scooting on his or her bottom?
5	If you hold both hands just to balance him, does your baby support his own weight while standing?	
6	Does your baby get into a crawling position by getting up on her hands and knees?	

15.2 Response rate of pediatricians

The percentage of paediatricians returning a rating on their patients was 54%. A higher response rate (up to 75%) may have been realized, but paediatricians were unable to give a PSA rating to 14 infants whose caregivers had returned questionnaires but not attended a six-month well-baby visit. A response rate of 54% is the same response rate as physician surveys published in the literature (139), and therefore taken as an acceptable response rate for this study.

Because some paediatricians only returned one PSA, compared to others who returned up to six, data was re-analyzed by randomly selecting one infant from each paediatrician who had returned more than one assessment. This was done to determine if a single paediatrician-cluster biased the results of this study. The Kappa coefficient and test of symmetry was again used to determine the agreement between the 3 methods

with a sample size of 18 for the ASQ and 19 for the PEDS. The results were no different from the initial analysis indicating paediatrican-clusters did not bias the results.

15.3 Interventions

15.3.1 Physiotherapy

In a study by Kalie et al. (21), 28% of 12-month-old infants qualified for and received physiotherapy for GM delays. In this study, although nearly 83% of paediatricians indicated that they would refer an infant under 12 months with a mild, non-specific developmental delay for physiotherapy, only two of the 65 infants on the study were receiving physiotherapy, both of which had significant birth histories indicating cerebral palsy.

In the study cohort, 4 (6%) infants were identified by the paediatricians with GM concerns compared to 15 by the PEDS COMBINED and 15 by the ASQ (23% and 25% respectively). The detection rate for GM concerns by the questionnaires is very close to the percentage of 28% of children identified in the literature as requiring physiotherapy services in the first year following NICU care. The number of children requiring physiotherapy is likely to increase with increasing age as Reuner (10) found that by 17 years of age 55% of prematurely born children had required physiotherapy services. The rate of high-risk infants who will require physiotherapy is therefore greater than 23%, implying that questionnaires were unlikely to be over-detecting GM difficulties. Since most paediatricians indicated that they would refer infants under 12 months for physiotherapy but only 6% were identified with GM concerns, a large number of infants who could potentially have benefited were not referred to physiotherapy due to low detection rates by the paediatricians.

15.3.2 Occupational Therapy and Speech Therapy

Similar trends in infants who would have benefited from a referral to speech therapy or occupational therapy were noted in this study. Paediatricians identified 1 (1.5%) infant with communication difficulties, 2 (3%) infants with problem solving difficulties and 2 (3%) infants with FM difficulties. The questionnaires identified up to 14

(21.5%) communication delayed infants, 9 (14%) infants with problem-solving difficulties and 8 (12%) infants with FM difficulties. The literature shows that by 12 months, 16% - 32% of prematurely born infants received occupational therapy and 10% - 32% received speech therapy (21, 10). The detection rate of the questionnaires falls within the expected percentage of children likely to require occupational or speech therapy services as shown in the literature. This again highlights the low detection rates of paediatricians as well as the real possibility of referring infants early for intervention with the use of parent-administered screening tools.

15.4 Referral Practices

Paediatricians were asked to indicate if they commonly referred infants under 12 months of age with mild developmental delays to speech therapy, occupational therapy and physiotherapy. Less than half the paediatricians indicated they would refer infants to speech therapy and even less indicated they would refer to occupational therapy compared to more than 80% indicating that they would refer to physiotherapy. This was similar to the study conducted by Earls, Andrews and Hay (142) which found that providers were most comfortable referring infants with a clear motor skill issue and less certain of referral for problem solving issues and problems with personal social skills.

The trend of paediatricians to refer more frequently to physiotherapy was described by Reuner et al. (10) in 2009 as a gap between medical tradition which focuses on motor and neurological development and recent literature which highlights the impact of cognitive and neuropsychological deficits on developmental outcomes. In the current study, only one infant was identified by the paediatrician as having social-emotional concerns and this infant was diagnosed soon after birth with severe cerebral palsy. Difficulties identifying concerns in other domains were highlighted by one of the paediatricians in this study who commented that the infant was too young at 5 months corrected age to monitor areas other than gross motor.

In infants under 12 months of age, many skills are still emerging and traditional domains of GM, FM and Comm are more likely to be identifiable. Similar to other studies (10, 80, 142), in this study GM concerns were the most

frequently identified concerns by all three screening methods (ASQ, PEDS COMBINED and PSA), followed by communication concerns (Figure 5). The parent-administered questionnaires also identified 11% of SE concerns and 14% of infants with PS difficulties however, indicating that it is possible to identify and monitor more subtle developmental areas of cognition and emotional development in young infants with the use of parent-administered questionnaires.

15.5 Domains of Development

Some caution should be exercised when looking at specific developmental domains for an infant however, as especially at these early ages development in the different domains is interrelated and difficult to isolate. This can be seen in the choice of items chosen to represent specific domains by the different questionnaires. For example, PEDS:DM classifies the following question under the GM domain: "*If your baby is lying on her back, can she pass a toy from one hand to the other?*". In the ASQ a very similar question, "*does your baby pass a toy from one hand to the other?*" is classified under the PS domain.

In both the ASQ and PEDS COMBINED, a rating of 1 in the SE domain was always associated with a rating of 1 or 2 in the GM domain, highlighting the inter-relationship between the domains. As an example, in the ASQ the question, "*When on his back, does your baby put his foot in his mouth?*" is classified as a SE domain, but is highly dependent on gross motor skills. A case study from the cohort is given below to illustrate this point.

Infant 131B was born at 27 weeks gestation weighing 1040g. The child was ventilated for approximately 6 weeks and spent 108 days in the NICU suffering a Grade III Intraventricular heamorrhage, grade IV ROP and severe gastro-oesophageal reflux. The mother reported the infant underwent the following procedures whilst still in the NICU: VP shunt, laser for ROP, Nissan procedure and gastrostomy tube insertion.

From this history, the infant can be classified as high risk for developmental difficulties in all domains. Other difficulties also need to be considered including possible visual problems from ROP and feeding difficulties or oral aversion from severe reflux, which may affect normal infant behavioural responses and complicate the presentation.

The PEDS COMBINED and ASQ were completed by the mother when the infant was 6.5 months corrected age. This infant passed all domain items on the PEDS COMBINED except for RL. On the ASQ, the infant failed the GM and PS domain and scored as high-risk in the SE domain.

Table 11 illustrates the items failed in the two tests. Domains in which difficulties could possibly have resulted in failure of the items are checked. For example, the infant not holding out arms when mom says “come here” could be due to poor control of the trunk or shoulder girdle (GM), difficulty coordinating bilateral arms (FM), not recognizing the phrase or intonation (RL), not responding appropriately to social interactions (SE) or not yet understanding actions and consequences (PS). Possible other issues for failure of this item include an undetected hearing loss resulting in the infant not responding to speech. To determine which of these are actually responsible for the item failure requires a more in-depth assessment.

Table 11 Domains involved in failed items on ASQ and PEDS:DM

Domains possibly involved in failed items							
	GM	**FM**	**RL**	**EL**	**SE**	**PS**	**Summary**
PEDS Rating	3	3	**1**	3	3	3	**1**
Items failed	GM	FM	RL	EL	SE	PS	Other
Not excited to see breast or bottle			☑		☑	☑	☑ - Vision, oral aversion, lack of feeding experience due to gastrostomy tube
Not holding out arms when mom says "come here"	☑	☑	☑		☑	☑	☑ - Auditory
	GM	**FM**	**RL**	**EL**	**SE**	**PS**	**Summary**
ASQ Rating	**1**	3	3		2	**1**	**1**
Items failed	GM	FM	RL	EL	SE	PS	Other
Not prop-sitting, standing with support or pushing into 4-point-kneeling	☑						
Not smiling in mirror					☑		☑ - Vision
Not getting hands to feet or feet to mouth	☑	☑				☑	
Not passing toys from one hand to the other	☑	☑				☑	
Not playing by banging toys	☑	☑				☑	
Not reaching with bilateral hands	☑	☑				☑	

The ASQ rates GM and PS as 1, indicating a referral to physiotherapy or occupational therapy. The PEDS COMBINED rates RL as 1, indicating a referral to speech therapy or audiology. There is therefore poor agreement between the two questionnaires. If summary scores are taken into account however, both questionnaires have a rating of 1 and identify this child as requiring further assessment. To account for the complexity of developmental domains and their influence on development, as well as other environmental or genetic influences, it is essential that this next phase of developmental assessment be completed by practitioners or EI teams who are experienced in childhood development (4, 51).

The complexity of each milestone and the variety of skills that contribute to an items success makes it difficult to limit items to one domain. This may explain why summary scores for the ASQ showed excellent agreement with the summary scores for the PEDS COMBINED ($k = 0.82$); but domain scores showed only moderate agreement between the two questionnaires ($k < 0.53$).

15.6 Comparison of the ASQ & PEDS COMBINED

15.6.1 Summary Ratings

Differences in summary recommendation from the ASQ and PEDS COMBINED were analyzed. Statistical analysis revealed that although there was excellent agreement for summaries of the two questionnaires (90.7%, $\kappa = 0.82$, $\rho < 0.05$), if they disagreed the trend was l for the ASQ to be more sensitive than the PEDS.

When the PEDS COMBINED failed an infant (rating 1), the ASQ was likely to rate the infant as high risk (rating 2), indicating developmental concerns even though the infant did not fail the ASQ. Instructions in the ASQ User's Guide" suggest that if several areas of development receive scores that are low but not below the cutoff point, the child may need to be referred for assessment (88). Scoring in this manner was not used for this study as the recommendations were slightly vague and subjective. Four of the five infants who scored a 1 for the PEDS did however have more than one domain with a rating of 2 on the ASQ and could therefore have possibly received a summary rating of 1 for the ASQ also. This indicates that when the infant failed the PEDS COMBINED, they were very likely to show concerning development on the ASQ.

There were four infants that received a rating of 1 on the ASQ summary and a rating of 3 on the PEDS COMBINED summary. This sample size was too small to reveal any patterns between these discrepancies but show a trend for the ASQ to be more sensitive to developmental delays that the PEDS in this cohort. The opposite of this was found in the study by Sices et.al. (112) where the PEDS identified more children than the ASQ with developmental concerns. This difference was also not significant however and further investigation into the function of these two questionnaires is different settings may be beneficial in this regard.

15.6.2 Domain Ratings

Comparison of domain discrepancies did reveal a statistically significant pattern of disagreement between these two questionnaires for the domain of Comm. Discordance was in the direction of the ASQ scoring a 3 when the PEDS COMBINED scored a 1 ($\rho = 0.01$). This may be explained by the fact that the ASQ combines receptive

and expressive language into one domain, whilst the PEDS COMBINED separates these two components of language into two separate domains (RL and EL). The value of separating these two domains of development can be shown by using an example of one of the infants in this study.

> At 10 months corrected age, this infant was given a rating of 1 for the communication domain for the PEDS COMBINED as he failed the following RL items.
>
> *"When you say your baby's name does he or she stop and look at you?"*
>
> *"When you say things like, "Where's your bottle?" does your baby look around for his or her bottle?"*
>
> He was given a rating of 3 for the ASQ communication domain, but when individual items were analyzed, he had failed both the receptive language items:
>
> *"If you ask her to, does your baby play at least one nursery game even if you don't show her the activity yourself (eg. "Peekaboo)"*
>
> *"Does your baby follow one simple command, such as "come here", "give it to me" without your using gestures?"*
>
> The remaining four questions under the communication domain were passed, but related to expressive language and this pulled the infant's score up, resulting in a pass for this domain on the ASQ.

The ASQ was less sensitive than the PEDS COMBINED in detecting delays in communication, with the PEDS COMBINED detecting 15 infants with communication delays compared to 3 by the ASQ. The study by Sices et.al. (112) also identified the Comm domain as the most likely domain to be discordant between the two screens. The population group was older in the Sices et.al. study, with a mean age of 17.6 months (SD: 6.1 months), indicating that the discrepancies in the Comm domain persisted into the older age groups. The discrepancy in the current study between ratings in the Comm domain did not cause significant disagreement in the summary scores of the ASQ and PEDS COMBINED however, as all of the infants who passed the ASQ Comm domain failed in another ASQ domain or scored as high risk (rating 2) in at least two other ASQ domains.

The GM domain for the PEDS is another area where individual domains are analysed differently by the two questionnaires. The PEDS does not flag GM concerns as predictive of developmental concerns until 3-years of age. The reason for this is explained by Glascoe (143). The author points out that the original design of the PEDS was to identify children for special education. In the USA children with physical impairment only qualify for placement in special education when there are other developmental concerns and therefore predictive value of other parental concerns are given more weight than gross motor concerns. This poor ability to identify GM concerns before 3-years of age is addressed with the PEDS COMBINED however, which provides age-equivalent scores and percentage delays for all domains and resulted in similar number of children on this study being identified for GM concerns using either the ASQ or PEDS COMBINED with moderate agreement between the two screens (k=0.53). (102, 104).

15.6.3 Detection Rates

Use of the PEDS COMBINED also addresses problems found with using PEDS alone in a high-risk population group. Pritchard, Colditz and Beller (144) found that caregivers whose children were already receiving EI may have answered “no” to the questions on the PEDS regarding their concerns about their child’s needs because these were already being addressed. This resulted in a high false-negative rate in their study on prematurely born infants using just the PEDS. In the current study, adding the PEDS:DM assessment level version, which is specifically recommended for following up infants requiring NICU care (104), resulted in a high rate of positive screen and a similar percentage of infants being identified by the PEDS COMBINED (42%) and ASQ (40%) (κ = 0.82). The similarity in identification rate and agreement between the PEDS COMBINED and ASQ implies that the PEDS COMBINED is an appropriate tool for use in this population since the ASQ was originally designed for following up NICU infants and has been used with success in this population group (83, 91, 92, 93).

In a comparison of the ASQ and PEDS conducted by Sices et.al.(112) in 2009, the ASQ identified 27% of children at risk for developmental delay, whereas the PEDS identified 37%. Although the percentage difference

of children identified was not statistically different, the agreement between the ASQ and PEDS was only moderate and not different from that expected by chance (κ = 0.24). The difference between the current study and the study by Sices et. al., is the addition of the PEDS:DM. Using the PEDS COMBINED resulted in excellent agreement between the tests (κ = 0.82) with the PEDS identifying 37.97% of the infants as requiring further assessment and the ASQ identifying 36.36% from the cohort. The PEDS COMBINED therefore seems to address issues of parental interpretation and decreased GM sensitivity, resulting in excellent overall agreement between the ASQ and PEDS COMBINED and significantly decreasing discordance.

15.6.4 Practicalities when using the ASQ and PEDS COMBINED

There is some discussion as to which questionnaire is more appropriate to use in a particular setting. The decision about which of these tools to use has primarily focused on provider choice (56, 112). In this study the ASQ was easier to learn, use and score. It did not require additional score sheet like the PEDS COMBINED and the information summary for each questionnaire contained the cut-off points in an easy to use scoring system (Appendix 18.4 -18.6). The PEDS:DM assessment level version was more appropriate for screening infants by mail however as the continuous item levels meant that forms were meaningful regardless of the age of the infant when it was completed. When using the ASQ for this study, if caregivers did not complete the questionnaire at the right time, scoring became problematic. Reasons for this include forms that were received late due to the poor postal system, caregivers filling in forms late and one form where the caregiver completed the form too early, resulting in concerns that were no longer found when they filled the form in again at the correct time.

Another consideration for choosing questionnaires to use in the clinical setting or for research is the support by the authors of the questionnaires. The author of the PEDS COMBINED was always responsive to questions about the use and application of the tool. In comparison, it was very difficult to get a response to questions regarding the ASQ from the publishers as they did not respond to any questions other than those related to the copyright laws.

15.7 Identification of developmental delay

15.7.1 Comparison of three screening methods

The literature indicates that the percentage of children from a neonatal intensive care that would benefit from EI is between 30% - 70%, with an average of 42% needing educational support, as compared to 10% - 18% in the general population(10, 16, 26). In this study, the ASQ identified 40% and the PEDS COMBINED identified 42% of NICU infants as developmentally delayed, in-line with the expected prevalence of developmental delay of high-risk infants in the literature. The paediatricians identified only 6% of the NICU infants as developmentally delayed, well below the expected prevalence for infants requiring NICU care and even below the percentage expected in the general population with no risk. Other studies have also shown under detection of developmental delay when health care practitioners do not use standardized screening methods (62, 80) (Table 12).

Table 12 Comparison of ASQ, PEDS COMBINED and PSA screens

PEDS:ASQ	Questionnaires (n)	PSA pass (n)	PSA fail (n)	Lost to follow-up (n)
Pass:Pass	19	12	0	1
Pass:Fail	4	1	0	1
Fail:Pass	0	0	0	0
Fail:Fail	20	6	2	6

Of the infants in this cohort, 13% had not been to their paediatrician for a developmental assessment in more than 4 months and had no future appointments planned. For example, one mom reported that she would not be seeing her paediatrician unless her baby was sick as her paediatrician was too far away. This infant received a rating of 1 for GM concerns on the PEDS and a rating of 2 for GM concerns on the ASQ. Another infant had not seen their paediatrician due to financial constraints since the birth. This infant received a rating of 1 for GM, FM and SH skills on the PEDS (the ASQ was not completed). These examples highlight the benefit of a cost-effective systematic screening programme to identify those infants that may otherwise be lost to follow-up. They also highlight the fact that parental compliance rates affect the success of EI (17).

15.7.2 Accuracy of detection

The current study did not include a diagnostic measure to determine the accuracy of the questionnaires in detecting developmental delay. Other studies have looked at the accuracy and validity of the questionnaires in comparison to "gold standard" diagnostic measures and have found acceptable psychometric properties (88, 102).

The accuracy of the ASQ in a clinical setting was examined in a retrospective study by Marks, Hix-Small and Clark (17) in 2009. The authors compared the accuracy of paediatricians' screening practices to the ASQ in the referral of infants to EI and childhood special education. In their study nine infants were identified as age appropriate between 12 and 24 months by the board-certified paediatricans but as developmentally delayed by the ASQ. Only one of these nine infants went on to be assessed as not requiring either early intervention or early childhood special education by 60 months. When the authors looked retrospectively at children who were appropriately enrolled in intervention services, 52.9% of the earlier referrals were directly as a result of the use of the ASQ. The authors concluded that the ASQ was correct to capture a higher percentage of concerns and recommended that only children who are already enrolled in intervention programmes should bypass developmental screening.

Statistical analysis between the ASQ:PSA and the PSA:PEDS COMBINED show that the agreement between the paediatricians and the questionnaires was poor and statistically less than expected by chance ($\kappa = 0.26$ and $\kappa = 0.28$ respectively with $p<0.05$). In contrast, the agreement between the two questionnaires was excellent and statistically greater than expected by chance ($\kappa = 0.82$, $\rho < 0.05$). It is likely that the questionnaires were correct in identifying a significantly higher number of infants as developmentally at risk and the under-detection of developmental difficulties by the PSA may have resulted in many of the infants not being referred for EI at a time when they could have had substantial benefit.

In the study by Marks et al (17), paediatricians and the ASQ were significantly more likely to agree on developmental status in infants that were full-term than infants that were preterm. The authors concluded that

paediatricians probably experienced exaggerated difficulties identifying delayed development in lower risk, mostly late preterm infants. This can be related to the current study where the average gestational age of the infants was 34 weeks (SD = 2) and the average birth weight was greater than 1500g. Many of the infants from the study are therefore classified as lower-risk NICU infants and possibly more difficult to accurately assess for developmental difficulties without the use of a developmental screening tool.

15.8 Summary

This study has found that the use of both the ASQ and PEDS COMBINED was feasible in the South African private health care context. The questionnaires' rate of detection of developmental delay in a high-risk infant population group was in line with published international literature of expected rates for this population and the questionnaires were able to appropriately identify the same infants as developmentally delayed. The paediatricians identified a much lower rate of infants requiring further developmental assessment well below the expected rate of infants at-risk for developmental delay.

Olusanya stated that until those *'who are likely to be consulted first by caregivers are convinced or even aware of the value of early detection and intervention, only minimal progress will be realised'* (145). Considering the low rate of detection and lack of knowledge or use of parent-administered questionnaires by paediatricians in this study, this is an area that needs to be addressed. The need for improving knowledge and use of systematic developmental screening in South Africa with affordable and standardized tools is essential.

South African children face many challenges and should not be unnecessarily disadvantaged or deprived of EI opportunities simply due to under detection. Screening and monitoring developmental difficulties using parent-administered questionnaires is a feasible first step in early identification of developmental delay and a critical requirement for implementing and assessing early intervention services.

16 Limitations and Recommendations

16.1 Further follow-up studies on this cohort

This study looked at a select group of paediatricians and infants at-risk for developmental delay in the private health care sector in the southern region of Gauteng, South Africa. Optimal screening requires several screens at different time points. Infants in this study only received one screening each and further screening of these infants would have been useful in identifying the infants developmental trajectories and helpful in further identifying strengths and weaknesses in the performance of the ASQ and PEDS COMBINED.

It would also be helpful to assess the type of EI services available for these infants in the South African context as well as their pattern of usage, and the efficacy of the EI services. Studies documenting the long-term academic requirements of these infants who received NICU care in the private health care sector of South Africa would be valuable in comparing South African outcomes to those of other countries.

It would be beneficial to study the impact of identification and referral rates of paediatricians from this cohort following the introduction of a more formal screening programme into their practices.

16.2 Bias and Demographics

As discussed in section 15.1, the bias of non-respondents and the influence of completing both questionnaires at the same time could not be determined in this study. This may be addressed in future studies by identifying demographic variables and by asking the two main caregivers to fill in different questionnaires at the same time, or by changing the order of completion of the questionnaires in two homogenous groups of caregivers. Determining the demographics of future respondents and non-respondents will be beneficial in identifying which population groups would benefit from postal questionnaires and which would require screening at clinician visits.

Both questionnaires were scored by the researcher, introducing a possible bias. The questionnaires are designed to be subjectively scored according to the parent's concerns and are not open to interpretation by the

person scoring the tests however, decreasing the likelihood of this affecting results. Being blinded to the result of the other questionnaire would never the less further decrease the possibility of bias.

16.3 The South African Population

A limitation of this study is a small cohort that is not representative of the greater South African population in the public health sectors or in other regions.

Studies on the long-term outcomes of infants who graduated from an NICU in South African are needed. Information on the use of EI services in both the public and private health sector would also be beneficial, as well as information on the performance of existing early intervention programmes in terms of screening methods used, success in detecting developmental delay and efficacy of the services.

This information would assist in determining the impact of introducing more formal screening practices into these sectors and could also be used towards addressing the ethical considerations of ensuring intervention is available before implementing screening programmes.

16.4 Parent-administered Questionnaires

It has been shown in this literature review that parent-administered screening tools have been used in LAMI countries including South Africa. Validation studies in the private and public sectors of South Africa of these questionnaires has not yet occurred and be beneficial, particularly in the older age groups where the questionnaires are more likely to be influenced by different cultures.

This study looked only at the 6-month-old age group. Both the ASQ and PEDS COMBINED are recommended from 0 to 8 years of age and therefore investigation into the functioning of these questionnaires at different age groups and over different time frames is needed.

The practicalities and costs of using these parent-administered screening tools in different South African contextsalso needs further investigation.

The under-detection of developmental delay found in this study, together with the dearth of information on developmental screening practices and EI in South Africa highlights areas for further research. The use of parent-administered questionnaires for this purpose seems feasible and would play an important role in influencing policies to improve services for South African children.

17 Conclusion

This study has proved the hypotheses listed under Rationale and Research Questions in chapter 12:

1. The number of infants identified with developmental concerns by the PEDS COMBINED and ASQ was greater than that identified by the PSA.
2. The ASQ and PEDS COMBINED showed good agreement, better than that expected by chance
3. The PSA showed poor agreement with both the ASQ and PEDS COMBINED, less than expected by chance
4. Standardised parent-completed questionnaires are not routinely used by paediatricians in private practice in the southern Gauteng region of South Africa at present.

The excellent agreement between the two questionnaires indicates that these tools are effective in identifying children who require further developmental assessment. In contrast, the poor agreement of the paediatricians' subjective assessment with both questionnaires identifies a trend in paediatric private health care that could be improved and highlights the benefit of using these questionnaires to increase the early detection of children with developmental delay to improve early intervention and outcomes.

18 Appendix

18.1 Ethical Clearance

UNIVERSITY OF THE WITWATERSRAND, JOHANNESBURG

Division of the Deputy Registrar (Research)

HUMAN RESEARCH ETHICS COMMITTEE (MEDICAL)
R14/49 Silva

CLEARANCE CERTIFICATE	**PROTOCOL NUMBER M070416**
PROJECT	A Comparison of an Objective Standardized Parent-Administered Questionnaire to that of Subjective Screening Practices for the Early.....
INVESTIGATORS	Miss ML Silva
DEPARTMENT	Department of Physiotherapy
DATE CONSIDERED	07.05.04
DECISION OF THE COMMITTEE*	APPROVED UNCONDITIONALLY

Unless otherwise specified this ethical clearance is valid for 5 years and may be renewed upon application.

DATE 07.05.28 **CHAIRPERSON** .....
(Professors PE Cleaton-Jones, A Dhai, M Vorster, C Feldman, A Woodiwiss)

*Guidelines for written 'informed consent' attached where applicable

cc: Supervisor : Davies VA Prof

DECLARATION OF INVESTIGATOR(S)

To be completed in duplicate and **ONE COPY** returned to the Secretary at Room 10005, 10th Floor, Senate House, University.
I/We fully understand the conditions under which I am/we are authorized to carry out the abovementioned research and I/we guarantee to ensure compliance with these conditions. Should any departure to be contemplated from the research procedure as approved I/we undertake to resubmit the protocol to the Committee. **I agree to a completion of a yearly progress report.**

PLEASE QUOTE THE PROTOCOL NUMBER IN ALL ENQUIRIES

18.2 Information and Consent Forms

18.2.1 Hospital

Early Intervention Study For NICU Babies
Hospital Manager / Superintendent Consent Form

Dear

Hello, my name is Mindy Silva. I am a paediatric physiotherapist currently completing my masters degree in child health and neurodevelopment. I would like to invite your hospital to participate in my study, which will investigate the use of a parent questionnaire in detecting babies who would benefit from early intervention.

The rationale for the study is my belief that Standardised screening questionnaires would be a simple and cost-effective tool to introduce in both the private and public sectors in South Africa in order to facilitate timeous referrals for early intervention. Age-specific, standardised, parental screening questionnaires are easy to administer and cost effective, making them practical to implement at regular intervals.

Before leaving the NICU, parents will be invited to participate in the study by a letter (copy attached). When their baby is between 4 and 6 months corrected age, they will complete a parental screening questionnaire which will be scored by myself. Following this, when these babies next see their chosen paediatrician, I will contact the paediatrician to ask if they have identified any developmental concerns. Their concerns will then be correlated with the concerns identified by the parent-completed questionnaire.

All information will be completely confidential. Individual hospitals will not be identified. Babies will be assigned a number, and the baby's name will not appear on the questionnaire, nor on the feedback response from the paediatrician. Similarly, each paediatrician participating in the study will be assigned a letter. The paediatrician will not be asked for any information except that related to developmental concerns. Names, numbers and letters will be kept on a secure data base which is password protected and will be kept separate from the information generated by the completion of the forms and the telephonic report from the paediatrician. The study has been approved by the Human Research Ethics Committee (Medical)- **Protocol M070416**

Participating in this study will potentially assist in the introduction of a screening tool into the South African context, which may help many children in the future to gain from the benefits of receiving the intervention they need.

I am therefore requesting your permission to approach parents of babies being cared for in your NICU to ask them to participate in this study. You may of course withdraw your consent at anytime and discontinue participation without any reason or penalty.

Thank you for taking the time to read this. Please complete the attached form.

If you have any questions about this letter or the study, please contact me at CentaPaeds Therapy centre on 011-453-2624 or e-mail me at mindy@iburst.co.za.

Sincerely

Mindy Silva

Early Intervention Study For NICU Babies

Hospital Manager' / Superintendent Consent Form

Thank you for considering your participation in this study on the use of parent questionnaires to detect babies who would benefit from early intervention. Please complete the information below, delete which ever option is not applicable and fax to 011 453-2624.

I _____ *(name)* do / do not *(delete whichever is not applicable)* give permission for _____*(hospital name)* to participate in this study. I have read and understood the attached letter. I understand that I may withdraw my consent at anytime and discontinue participation without any reason or penalty.

_____ _____

Sign Date

Surname _____ Name _____

Hospital _____

PLEASE RETURN FORM BY FAX: 011 453-2624

18.2.2 Caregivers

Parent's Information Letter

Dear Parents or Gaurdian

Hello, my name is Mindy Silva. I am a paediatric physiotherapist currently completing my masters degree in child health and neurodevelopment. I would like to invite you to participate in my study, which will investigate the use of a parent questionnaire in detecting babies who would benefit from early intervention.

Professional organisations (such as The Committee on Children with Disabilities of the American Academy of Pediatrics) recommend screening of all healthy infants and children for developmental delay. The obvious benefit in this can be found in studies that have shown that a child will benefit more from intervention programmes that are begun as early as possible. Parent questionnaires are being used overseas as routine methods of screening infants. The purpose of this study is to introduce parent questionnaires into the South African paediatrician's office and to compare the ability of these questionnaires to that of the paediatrician in detecting developmental concerns.

If you agree to participate in the study you will be mailed 2 short questionnaires and a form requesting updated contact details and health information, which can be completed in your own home when your baby is between 4 and 7 months corrected age. The questionnaire will take you approximately 5 minutes each and consists of short questions about your baby's developmental milestones and concerns that you might have about your baby's development. You will then need to post this back to me. Cost of postage will be covered by myself. I will also need your consent to allow your paediatrician to give me a telephonic report on your baby's development following your routine check-up between the 4th and 7th month corrected age.

All information will be completely confidential. You will be assigned a number so that neither your name, nor your baby's name, appears on the questionnaire, or feedback response from the paediatrician. The paediatrician will not be asked for any information except that related to developmental concerns. Names and numbers will be kept on a secure data base which is password protected and will be kept separate from the information generated by your completion of the forms, and the telephonic report from your paediatrician. The study has been approved by the Human Research Ethics Committee (Medical)- **Protocol M070416**

By participating in the study, you will benefit from a holistic screening of your baby's development. You will also be given numbers to contact in the event of any concerns or anxiety that you may have as a result of participating in this study. Participating in this study will potentially assist in the introduction of a screening tool in South Africa, which may help many babies to gain from the benefits of receiving the intervention they need.

Signing and filling in the attached form indicates that you have read and understood all the information provided above; and that you agree to participate willingly. You may withdraw your consent at anytime and discontinue participation without any reason or penalty.

Thank you for taking the time to read this. If you have any questions about this letter or the study, please contact me at CentaPaeds Therapy centre on 011-453-2624 or e-mail me at mindy@iburst.co.za.

Sincerely

Mindy Silva

Parent's or Guardian's Consent Form

Thank you for considering your participation in this study on the use of parent questionnaires to detect babies who would benefit from early intervention. Please complete the information below and delete which ever option is not applicable.

I _____ parent / legal guardian of _____ agree / don't agree to willingly participate in this study. I further give consent for you to contact my baby's paediatrician for information on his/her development. I have read and understood the attached letter. I understand that I may withdraw my consent at anytime and discontinue participation without any reason or penalty.

_____ _____

Sign Date

Surname _____ Name _____

Baby's name _____ Paediatrician _____

Baby's date of birth _____ expected date of birth _____

Baby's birth weight _____

Name of hospital where baby was born _____

Name of hospital where baby was in NICU _____

How long did your baby stay in the NICU _____

Reason for NICU stay_____

How many days was your baby ventilated _____

Operations _____

Complications_____

Contact Details

	Mother's Details	Father's Details
Name		
Address	Residential Postal	
Home tel		
Work tel.		
Cell no.		
Fax no.		

18.2.3 Paediatrician

Early Intervention Study For NICU Infants

Dear Doctor

Thank you for taking the time to read this information. My name is Mindy Silva. I am currently completing my masters degree in child health and neurodevelopment. I would like to invite you to participate in my study, which will investigate the use of parent questionnaires in detecting babies who would benefit from early intervention.

The rationale for the study is that standardised, parent-administered screening questionnaires would be a simple and feasable tool to introduce in both the private and public sectors in South Africa, in order to facilitate developmental monitoring. These parental screening questionnaires are easy to administer and cost effective, making them practical to implement at regular intervals.

Parents on this study have babies who have required more than 3 days NICU care in any one of the private hospitals in the Southern Gauteng area. They have filled in parent-administered screening questionnaires regarding their baby's neurodevelopment, which they have received by post, fax or e-mail. They have also given consent for me to contact you in order to obtain a subjective report on their baby's neurodevelopmental status.

Details regarding the baby's neurodevelopment are not required, only wheather or not there are concerns in any of the developmental domains. You will not be asked for any information other than to indicate if the baby is doing well or requires a more in-depth assessment or closer monitoring (A sample form is included on page 4). Although I do not require any detailed information, any comments you make will be valuable to me in interpreting my data. A once-off short questionnaire regarding your knowledge of parent-administered questionniares is provided on page 3 and I would appreciate your input on this also.

You and your patient's details will remain anonymous and confidential. Information will be kept on a secure data base. The study has been approved by the Human Research Ethics Committee (Medical)- **Protocol M070416**

Participating in this study will potentially assist in the introduction of a screening tool into the South African context, which may help many children in the future to gain from the benefits of receiving the intervention they need.

Signing and filling in the attached form on page 2 indicates that you have read and understood all the information provided above; and that you agree to participate willingly. You may withdraw your consent at anytime and discontinue participation without any reason or penalty.

Thank you for taking the time to read this. If you have any questions about this letter or the study, please contact me at CentaPaeds Therapy centre on 011-453-2624 or e-mail me at mindysilva1@gmail.com

Sincerely
Mindy Silva

Doctors Consent Form

Thank you for considering your participation in this study on the use of parent-administered questionnaires to detect babies who would benefit from early intervention. Please complete the information below and delete whichever option is not applicable.

I _____ (name) agree / do not agree to willingly participate in this study. I have read and understood the attached letter. I understand that I may withdraw my consent at anytime and discontinue participation without any reason or penalty.

_____ _____

Sign Date

Surname _____ Name _____

Address of rooms 1

Address of rooms 2

Telephone number rooms 1

Telephone number rooms 2

Please make a note of preferred times and numbers that I can contact you on

_____

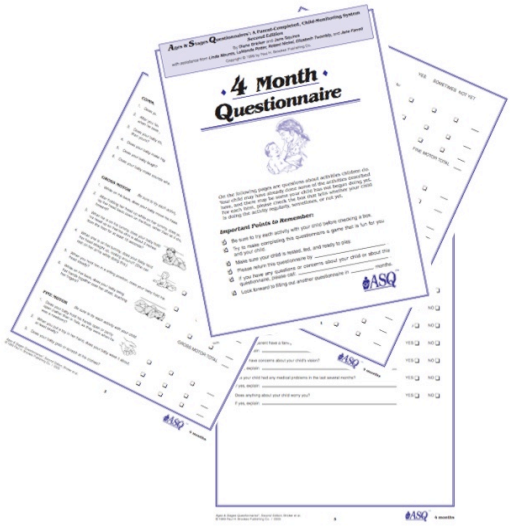
Would you prefer to give information by telephone, e-mail or fax? *(circle preferred choice)*

e-mail: _____ Fax: _____

18.3 Caregivers Pack

18.3.1 Information Sheet

18.3.2 Details Update Form



Please fill out this form, even if you have already done one previously

Initial Parent's or Guardian's Consent Form

Thank you for considering your participation in this study on the use of parent questionnaires to detect babies who would benefit from early intervention. Please complete the information below and delete which ever option is not applicable.

Please type in coloured areas

I _____ parent / legal guardian of _____ agree / don't agree to willingly participate in this study. I further give consent for you to contact my baby's paediatrician for information on his/her development. I have read and understood the attached letter. I understand that I may withdraw my consent at anytime and discontinue participation without any reason or penalty.

_____
Date

Surname		Name	
Baby's name		Paediatrician	
Baby's date of birth		Expected date of birth	
Baby's birth weight		Name of hospital where baby was born	
Name of hospital where baby was in NICU		How long did your baby stay in the NICU	
Reason for NICU stay			
How many days was your baby ventilated			
Operations			
Complications			

Contact Details

		Mother's Details	Father's Details
Name			
Address	Residential		
	Postal		
Home tel			
Work tel.			
Cell no.			
Fax no.			
e-mail			

Details update form – Please complete

Please type in coloured areas

Use as much space as you need

Baby's name:	
Date PEDS:DM assessment level booklet was filled in	
Date PEDS response form was filled in: :	
Date of last paediatrician's visit	
Date of next paediatrician's visit:	
Name of paediatrician	
Telephone no. of paediatrician	

Total number of days your baby was in NICU		Total number of days ventilated	

Please make a note of any serious illnesses or operations your baby has had since leaving the NICU:

Has your baby received an assessment, or been referred for an assessment, to a therapist or specialist due to concerns about development? Mark with an X		Yes		No

Please specify:

Developmental screening should be an ongoing process. Can I send you similar forms to monitor your baby's development in the future?		Yes		No

FOR OFFICE USE – DO NOT COMPLETE						
1 – Refer 2 – Monitor 3 – No concerns						
	Gross motor	Self help	Fine motor	Expressive	Receptive	Social-emotional
PEDS/PEDS:DM						

18.3.3 PEDS COMBINED Sample

Sample from PEDS

0. Please list any concerns about your child's learning, developments and behavior.

He seems little slow in some of his gross motor skills mainly crawling, trying to stand on his own with help of furniture

1. Do you have an concerns about how your child talks and makes speech sounds?

Mark with an X	No	X	Yes		A little	

Comments: He is very chatty.

2. Do you have an concerns about how your child understands what you say?

Mark with an X	No	X	Yes		A little	

Comments:

3. Do you have any concerns about how your child uses his or her hands and fingers to do things?

Mark with an X	No	X	Yes		A little	

Comments:

Sample from PEDS:DM

		Sometimes
or sees a pet?	X	Yes
4. How many different sounds such as "muh", "bah", "duh", or "guh" does your baby say?		None
		1
	X	2 or more
5. Does your baby put sounds together that sounds like talking?		No
		Sometimes
	X	Yes
6. If you offer your child something she likes, does she nod or say "yes"?		No
	X	Sometimes
		Most of the time
7. Does your child try to get your attention by pointing to things?	X	No
		Sometimes
		Most of the time

How your child is learning to use arms and legs: Gross Motor

Fill in an X		
1. Does your baby try to keep his or her head steady?		No
	X	A little
		Yes
2. Does your baby roll from her back to her side?		No
		Sometimes
	X	Yes
3. If your baby is lying on her back, can she pass a toy from one hand to the other?		No
		Sometimes
	X	Yes
4. Can your baby get around on hands and knees or by scooting on his or her bottom?		No
	X	A little
		Yes
5. If you hold **only one** of your baby's hands, can he or she take a few steps?		No
		A little
		Yes
6. Can your child walk without falling much?		No
		Falls a lot
		Doesn't fall often

Glascoe FP. Parents' Evaluation of Developmental Status. Nashville, TN: Ellsworth & Vandermeer Press LLC, 2006 www.pedstest.com

18.3.4 ASQ Sample

	YES	SOMETIMES	NOT YET	
COMMUNICATION *Be sure to try each activity with your child.*				
1. Does your baby make high-pitched squeals?	☑	☐	☐	___
2. When playing with sounds, does your baby make grunting, growling, or other deep-toned sounds?	☑	☐	☐	___
3. If you call your baby when you are out of sight, does she look in the direction of your voice?	☑	☐	☐	___
4. When a loud noise occurs, does your baby turn to see where the sound came from?	☑	☐	☐	___
5. Does your baby make sounds like "da," "ga," "ka," and "ba"?	☑	☐	☐	___
6. If you copy the sounds your baby makes, does your baby repeat the sounds back to you?	☐	☑	☐	___
		COMMUNICATION TOTAL		___
GROSS MOTOR *Be sure to try each activity with your child.*				
1. While on his back, does your baby lift his legs high enough to see his feet?	☑	☐	☐	___
2. When she is on her tummy, does your baby straighten both arms and push her whole chest off the bed or floor?	☐	☐	☑	___
3. Does your baby roll from his back to his tummy, getting both arms out from under him?	☐	☐	☑	___
4. When you put her on the floor, does your baby lean on her hands while sitting? (If she already sits up straight without leaning on her hands, check "yes" for this item.) 	☐	☐	☑	___
5. If you hold both hands just to balance him, does your baby support his own weight while standing? 	☐	☐	☑	___
6. Does your baby get into a crawling position by getting up on her hands and knees? 	☐	☐	☑	___
		GROSS MOTOR TOTAL		___
FINE MOTOR *Be sure to try each activity with your child.*				
1. Does your baby grab a toy you offer and look at it, wave it about, or chew on it for about 1 minute?	☑	☐	☐	___

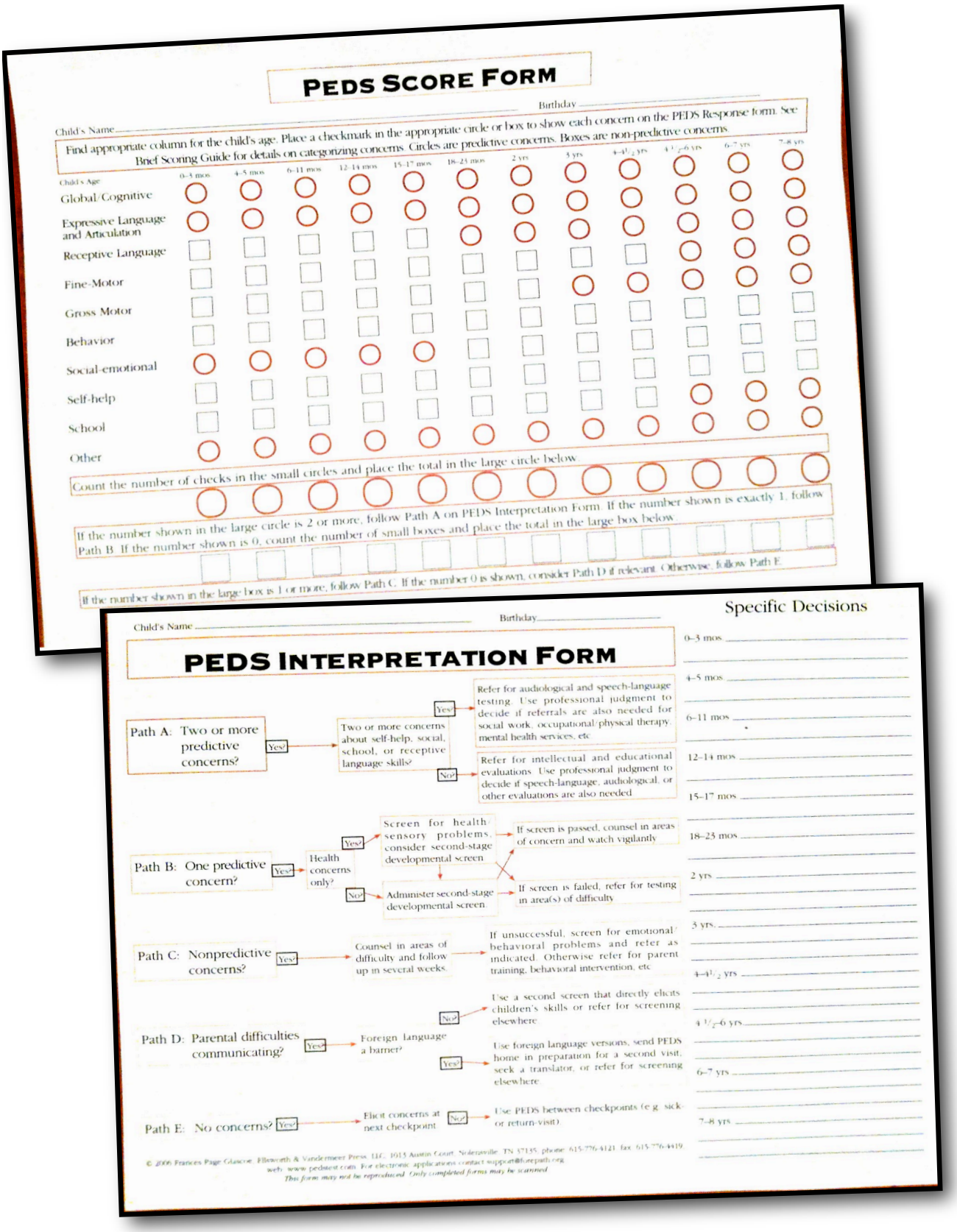
Ages & Stages Questionnaires®, Second Edition, Bricker et al.
© 1999 Paul H. Brookes Publishing Co. / 0305

3

6 months

Bricker D, Squires J. Ages and Stages Questionnaires. A Parent-Completed Child-Monitoring System Second Edition. Paul H. Brookes Publishing Co. Baltimore, Maryland 1999

18.4 Score Form PEDS



PEDS SCORE FORM

Child's Name _____ Birthday _____

Find appropriate column for the child's age. Place a checkmark in the appropriate circle or box to show each concern on the PEDS Response form. See Brief Scoring Guide for details on categorizing concerns. Circles are predictive concerns. Boxes are non-predictive concerns.

Child's Age	0–3 mos	4–5 mos	6–11 mos	12–14 mos	15–17 mos	18–23 mos	2 yrs	3 yrs	4–4½ yrs	4½–6 yrs	6–7 yrs	7–8 yrs
Global/Cognitive	○	○	○	○	○	○	○	○	○	○	○	○
Expressive Language and Articulation	○	○	○	○	○	○	○	○	○	○	○	○
Receptive Language	□	□	□	□	□	○	○	○	○	○	○	○
Fine-Motor	□	□	□	□	□	□	□	□	□	○	○	○
Gross Motor	□	□	□	□	□	□	□	□	□	□	□	□
Behavior	□	□	□	□	□	□	□	□	□	□	□	□
Social-emotional	○	○	○	○	○	□	□	□	□	□	□	□
Self-help	□	□	□	□	□	□	□	□	□	□	○	○
School	□	□	□	□	□	□	□	□	□	□	□	□
Other	○	○	○	○	○	○	○	○	○	○	○	○

Count the number of checks in the small circles and place the total in the large circle below.

If the number shown in the large circle is 2 or more, follow Path A on PEDS Interpretation Form. If the number shown is exactly 1, follow Path B. If the number shown is 0, count the number of small boxes and place the total in the large box below.

If the number shown in the large box is 1 or more, follow Path C. If the number 0 is shown, consider Path D if relevant. Otherwise, follow Path E.

Child's Name _____ Birthday _____

PEDS INTERPRETATION FORM

Path A: Two or more predictive concerns? Yes → Two or more concerns about self-help, social, school, or receptive language skills? Yes → Refer for audiological and speech-language testing. Use professional judgment to decide if referrals are also needed for social work, occupational/physical therapy, mental health services, etc. No → Refer for intellectual and educational evaluations. Use professional judgment to decide if speech-language, audiological, or other evaluations are also needed.

Path B: One predictive concern? Yes → Health concerns only? Yes → Screen for health/sensory problems, consider second-stage developmental screen. Yes → If screen is passed, counsel in areas of concern and watch vigilantly. If screen is failed, refer for testing in area(s) of difficulty. No → Administer second-stage developmental screen.

Path C: Nonpredictive concerns? Yes → Counsel in areas of difficulty and follow up in several weeks. If unsuccessful, screen for emotional/behavioral problems and refer as indicated. Otherwise refer for parent training, behavioral intervention, etc.

Path D: Parental difficulties communicating? Yes → Foreign language a barrier? No → Use a second screen that directly elicits children's skills or refer for screening elsewhere. Yes → Use foreign language versions, send PEDS home in preparation for a second visit, seek a translator, or refer for screening elsewhere.

Path E: No concerns? Yes → Elicit concerns at next checkpoint. No → Use PEDS between checkpoints (e.g. sick- or return-visit).

Specific Decisions

0–3 mos _____
4–5 mos _____
6–11 mos _____
12–14 mos _____
15–17 mos _____
18–23 mos _____
2 yrs _____
3 yrs _____
4–4½ yrs _____
4½–6 yrs _____
6–7 yrs _____
7–8 yrs _____

Glascoe FP. Parents' Evaluation of Developmental Status. Nashville, TN: Ellsworth & Vandermeer Press LLC, 2006 www.pedstest.com

child's name _____ date of birth _____ date of test _____

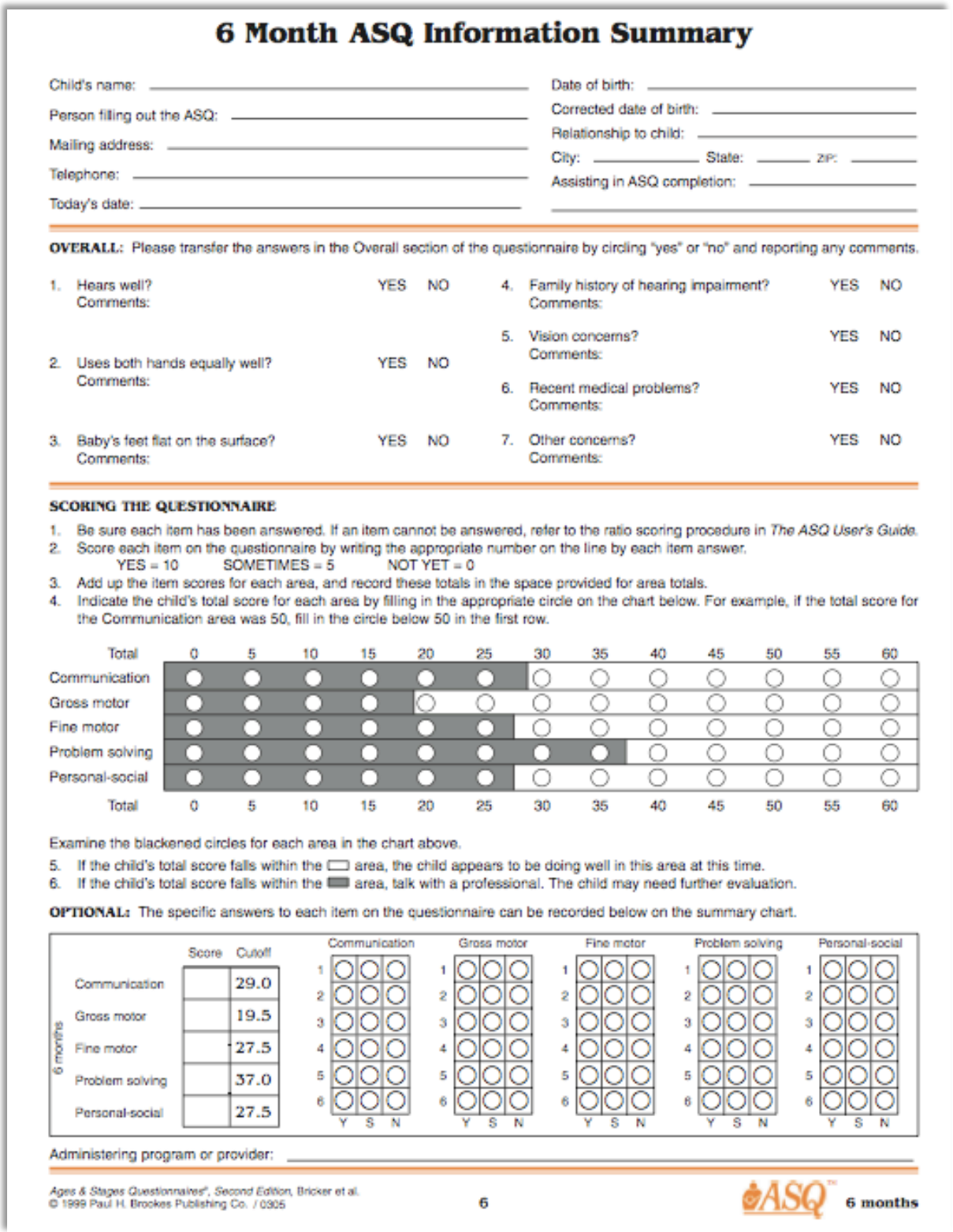
PEDS:DM ASSESSMENT LEVEL SCORING GUIDE

AGE	Fine Motor			Self Help			Receptive Language		
	Domain Item #	Give credit for:	Months value SE_M =+ .7	Domain Item #	Give credit for:	Months value SE_M =+ .7	Domain Item #	Give credit for:	Months value SE_M =+ .8
0 - 2 mos	1.	A little, Yes	2	1.	Sometimes, Yes	2	1.	Yes	2
3 - 4 mos	2.	Yes	2		Yes	4	2.	Yes	2
5 - 7 mos	3.	Yes	2	2.	Yes	2	3.	Sometimes, Yes	2
8 - 10 mos	4.	Yes	3	3.	Yes	3	4.	Most of the time	3
11 - 13 mos	5.	A little, Yes	3	4.	Yes	3	5.	Most of the time	3
14 - 16 mos	6.	Most of the time	3	5.	Most of the time	3	6.	Most of the time	3
17 - 19 mos	7.	3 or more blocks	3	6.	Most of the time	3	7.	2 or more	3
20-22 mos	8.	Sometimes, Yes	3	7.	Yes	3		2 or more	6
23-25 mos		Yes	6	8.	Sometimes Most of the time	3		3 or more	9
26-28 mos		Yes	9		Most of the time	6	8.	All	3
2-5 to 2-9	9.	Yes	5	9.	Most of the time	4	9.	3 or more	4
2-10 to 3-2		Yes	9	10.	Yes, mostly on right feet	5	10.	Knows both	5
3-3 to 3-7	10.	1 part 2 or more parts	5	11.	Most of the time	5	11.	Yes, does both	5
3-8 to 4-0		4 or more parts	10	12.	Says first and last name	5	12.	Does all three	5
4-1 to 4-5	11.	Yes	5	13.	Mostly Yes and does all fasteners	5	13.	All	5
4-6 to 4-10	12.	1 letter 2 or more letters	5	14.	Yes	5	14.	2 or more	5
4-11 to 5-5	13.	3 or more numbers	6	15.	15 or more minutes	6	15.	All	6
5-6 to 6-0	14.	3 or more	7	16.	Knows first or last name Knows first and last name	7	16.	3 or more	7
6-1 to 6-11	15.	All lines touch correctly	9	17.	Yes, Maybe	9	17.	1 2 or more	9
7-0 to 7-11	16.	2-3, 4-5	12				18.	Yes Some but not all	12
	Total Fine Motor		/90	Total Self-help		/78	Total Receptive Language		/90

Scoring Guide (continued)

AGE	Expressive Language			Gross Motor			Social Emotional		
	Domain Item #	Give credit for:	Months value SE_M =+ .7	Domain Item #	Give credit for:	Months value SE_M =+ .5	Domain Item #	Give credit for:	Months value SE_M =+ .6
0 - 2 mos	1.	Yes	2	1.	A little, Yes	2	1.	Sometimes Most of the time	2
3 - 4 mos	2.	Sometimes, Yes	2	2.	Sometimes, Yes	2	2.	Sometimes Most of the time	2
5 - 7 mos	3.	Sometimes, Yes	2	3.	Sometimes, Yes	2	3.	Sometimes Most of the time	2
8 - 10 mos	4.	2 or more	3	4.	Sometimes, Yes	3	4.	Yes	3
11 - 13 mos	5.	Sometimes, Yes	3	5.	Yes	3	5.	Often	3
14 - 16 mos	6.	Most of the time	3	6.	Doesn't fall often	3	6.	Often	3
17 - 19 mos	7.	Sometimes, Most of the time	3	7.	Feet stay on the ground, One or both feet off the ground	3	7.	Yes	3
20-22 mos	8.	2 or more	3		One or both feet off the ground	6	8.	Yes	3
23-25 mos		5 or more	6	8.	Walks up on own, holds rail or needs one hand held	3	9.	Yes, uses two toys together	3
26-28 mos		8 or more	9	9.	Most of the time	3	10.	A little, Yes	3
2-5 to 2-9	9.	2 or more	4	10.	Yes	4	11.	Sometimes, Yes	4
2-10 to 3-2	10.	About half, Most	5	11.	Yes	5	12.	15-20 minutes	5
3-3 to 3-7	11.	One correct Both correct	5	12.	Puts one foot on each step	5	13.	Sometimes, Yes	5
3-8 to 4-0	12.	Knows 'last' or 'first', Knows 'last' & 'first'	5		Puts one foot on each step	10	14.	Yes	5
4-1 to 4-5	13.	Some, All	5	13.	Yes, not much arm-waving	5	15.	Yes	5
4-6 to 4-10	14.	No mistakes on at least one sentence	5				16.	Most of the time	5
4-11 to 5-5	15.	Yes	6				17.	Yes	6
5-6 to 6-0	16.	16-25, All	7						
6-1 to 6-11	17.	No mistake on at least one sentence	9						
7-0 to 7-11									
	Total Expressive Language		/78	Total Gross Motor		/51	Total Social-Emotional		/62

18.6 Score Form ASQ



6 Month ASQ Information Summary

Child's name: _____ Date of birth: _____

Person filling out the ASQ: _____ Corrected date of birth: _____

Mailing address: _____ Relationship to child: _____

Telephone: _____ City: _____ State: _____ ZIP: _____

Today's date: _____ Assisting in ASQ completion: _____

OVERALL: Please transfer the answers in the Overall section of the questionnaire by circling "yes" or "no" and reporting any comments.

1. Hears well? YES NO
   Comments:
2. Uses both hands equally well? YES NO
   Comments:
3. Baby's feet flat on the surface? YES NO
   Comments:
4. Family history of hearing impairment? YES NO
   Comments:
5. Vision concerns? YES NO
   Comments:
6. Recent medical problems? YES NO
   Comments:
7. Other concerns? YES NO
   Comments:

SCORING THE QUESTIONNAIRE

1. Be sure each item has been answered. If an item cannot be answered, refer to the ratio scoring procedure in *The ASQ User's Guide*.
2. Score each item on the questionnaire by writing the appropriate number on the line by each item answer.
   YES = 10 SOMETIMES = 5 NOT YET = 0
3. Add up the item scores for each area, and record these totals in the space provided for area totals.
4. Indicate the child's total score for each area by filling in the appropriate circle on the chart below. For example, if the total score for the Communication area was 50, fill in the circle below 50 in the first row.

Total	0	5	10	15	20	25	30	35	40	45	50	55	60
Communication	●	●	●	●	●	●	○	○	○	○	○	○	○
Gross motor	●	●	●	●	○	○	○	○	○	○	○	○	○
Fine motor	●	●	●	●	●	●	○	○	○	○	○	○	○
Problem solving	●	●	●	●	●	●	●	●	○	○	○	○	○
Personal-social	●	●	●	●	●	●	○	○	○	○	○	○	○
Total	0	5	10	15	20	25	30	35	40	45	50	55	60

Examine the blackened circles for each area in the chart above.

5. If the child's total score falls within the ▭ area, the child appears to be doing well in this area at this time.
6. If the child's total score falls within the ▬ area, talk with a professional. The child may need further evaluation.

OPTIONAL: The specific answers to each item on the questionnaire can be recorded below on the summary chart.

6 months	Score	Cutoff
Communication		29.0
Gross motor		19.5
Fine motor		27.5
Problem solving		37.0
Personal-social		27.5

Communication				Gross motor				Fine motor				Problem solving				Personal-social			
	Y	S	N		Y	S	N		Y	S	N		Y	S	N		Y	S	N
1	○	○	○	1	○	○	○	1	○	○	○	1	○	○	○	1	○	○	○
2	○	○	○	2	○	○	○	2	○	○	○	2	○	○	○	2	○	○	○
3	○	○	○	3	○	○	○	3	○	○	○	3	○	○	○	3	○	○	○
4	○	○	○	4	○	○	○	4	○	○	○	4	○	○	○	4	○	○	○
5	○	○	○	5	○	○	○	5	○	○	○	5	○	○	○	5	○	○	○
6	○	○	○	6	○	○	○	6	○	○	○	6	○	○	○	6	○	○	○

Administering program or provider: _____

ASQ™ 6 months

Squires J, Bricker D, Potter L. Ages and Stages Questionnaires User's Guide. 2nd ed. Baltimore: Paul Brookes Publishing, 1999.

18.7 Doctors Pack

18.7.1 Short Questionnaire

18.7.2 PSA

Knowledge And Use Of Parent-Completed Screening Questionnaires

1. Do you commonly refer babies under 12 months old with a non-specific, mild developmental delay for any of the following:
    a. Physiotherapy
    b. Occupational therapy
    c. Speech therapy

2. What do you normally base your referral on?
    a. Specific guide lines
    b. Clinical judgement and experience

3. Do you think the parents in your practice are able to identify developmental delays in their babies? Yes / No

4. Do you know about parent administered questionnaires for developmental screening? Yes / No
    a. If yes, can you name any?
    List _____
    _____
    _____

5. Have you ever used them in your practice? Yes / No
    a. If yes, which ones have you used?
    List _____
    _____
    _____

    b. What was your experience with it?
        i. Beneficial
        ii. Not beneficial *Expand*

PSA

Baby's name: Date:

Date of last appointment:

Mark an X in the appropriate column for EACH point.(1 – 6).

Make any relevant comments underneath each point

NB! Please indicate if there is any area of development which you will be monitoring more closely than usual

	Delayed - Refer for in depth assessment / intervention	Concerned :– monitor carefully	Not concerned at this time
1. Fine motor			
Comments			
2. Self help			
Comments			
3. Receptive			
Comments			
4. Expressive			
Comments			
5. Gross motor			
Comments			
6. Social emotional			
Comments			

18.7.3 Examples of Feedback Forms

18.7.3.1 Paediatrician

October 10, 2008

Dear [redacted]

[redacted], , has been a participant in research on the early detection of developmental delay using parent administered screening questionnaires at age 6 months corrected (the PEDS;PEDS:DM and Ages & Stages). These are screening tools recommended by the American Academy of Pediatrics.

Analysis of the forms show that he is falling significantly below his age level for a gross motor skills development and has several other areas of concern including receptive languages skills.

As these questionnaires are a screening tool and not an in-depth assessment, the parent has been given feedback on the results and asked to contact you for continued monitoring of these developmental areas.

Thank you for your participation in this research,

Regards,

Mindy Silva
011 453-2624

18.7.3.2 Caregiver

October 2, 2008

Dear Parent(s),

The forms that you have completed look carefully at how your baby is learning, developing and behaving. She is doing well in the areas below:

❑ using hands and fingers to do things
❑ listening and understanding
❑ Using the larger muscles in her arms, legs and body
❑ talking and speech
❑ learning to take care of herself
❑ getting along with others and behaving

As this questionnaire is a screening tool and not a comprehensive assessment, we would advise you to make an appointment with your peadiatrician should there be anything that concerns you about your baby's development.

Because they have such a difficult start, developmental monitoring is especially important for a baby who has spent time in a NICU. The earlier difficulties are identified and addressed, the better their ability to overcome them and reach their full potential. Please ensure your child has regular developmental monitoring.
If there are difficulties, good help is available.

Please feel free to contact me at mindy@iburst.co.za or (011)453 2624 should you have any questions or concerns.

Thank you,

Mindy Silva

18.8 Other

18.8.1 Kappa and Symmetry

To assess the accuracy of any particular measuring 'instrument', it is usual to distinguish between the *reliability* of the data collected and their *validity*. Reliability is essentially the extent of the agreement between repeated measurements, and validity is the extent to which a method of measurement provides a true assessment of that which it purports to measure. When studying the variability of observer *categorical* ratings, two components of possible lack of accuracy must be distinguished. The first is inter-observer bias, which is reflected in differences in the marginal distributions of the response variable for each of the observers (Cochran's Q-test is the appropriate test for the hypothesis of no inter-observer bias). The second is observer disagreement, which is indicated by how observers classify individual subjects into the same category on the measurement scale (Kappa coefficient is one of the most common approaches). In this part, we will focus on the Kappa coefficient (or Kappa statistics).

Kappa Statistics: an index which compares the agreement against that which might be expected by chance. Kappa can be thought of as the chance-corrected proportional agreement, and possible values range from +1 (perfect agreement) via 0 (no agreement above that expected by chance) to -1 (complete disagreement).

Hypothetical Example: 29 patients are examined by two independent doctors (see Table). 'Yes' denotes the patient is diagnosed with disease X by a doctor. 'No' denotes the patient is classified as no disease X by a doctor.

		Doctor A		
		No	Yes	Total
Doctor B	No	10 (34.5%)	7 (24.1%)	17 (58.6%)
	Yes	0 (0.0%)	12 (41.4%)	12 (41.4%)
Total		10 (34.5%)	19 (65.5%)	29

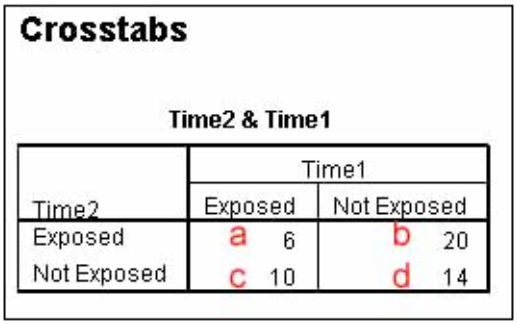
Kappa = (Observed agreement - Chance agreement)/(1 - Chance agreement)
Observed agreement = (10 + 12)/29 = 0.76
Chance agreement = 0.586 * 0.345 + 0.655 * 0.414 = 0.474
Kappa = (0.76 - 0.474)/(1 - 0.474) = 0.54

<http://www.dmi.columbia.edu/homepages/chuangj/kappa/ >
[Accesed 26 June 2009]

McNemar test

Symmetry. McNemar's test is sometimes called *McNemar's test of symmetry* or *McNemar symmetry chi-square* because it, and the marginal homogeneity test which extends it beyond dichotomous data, apply to square tables in which the diagonal reflects subjects who did not change between the before and after samples (or matched pair samples). The test variable may be nominal or ordinal. The test of symmetry tests whether the counts in cells above the diagonal differ from counts below the diagonal. If the two counts differ significantly, this reflects change between the samples, such as change due to an experimental effect between the before and after samples.

Computation of the McNemar Test. For the McNemar test, data are arranged as below (for



Crosstabs

Time2 & Time1

Time2	Time1 Exposed	Time1 Not Exposed
Exposed	a 6	b 20
Not Exposed	c 10	d 14

convenience, cell letters have been added in red):

Note that you cannot have a simple table where the columns are "Time 1" and "Time 2", and the rows are "Exposed" and "Not Exposed", because then observations would appear twice in the table! The McNemar test uses the chi-square distribution, based on this formula: Chi-square = $(|b - c| - 1)^2/(b + c)$ degrees-of-freedom = (rows - 1)(columns - 1) = 1 The formula above is the usual *continuity-corrected* version of the McNemar test. One occasionally encounters the uncorrected version, sometimes used for large samples, in which case the "-1" term in the numerator is omitted. *Example:* Chi-square = $(|20 - 10| - 1)^2/(20 + 10) = 81/30 = 2.7$ d.f. = (2 - 1)(2 - 1) = 1

< http://faculty.chass.ncsu.edu/garson/PA765/mcnemar.htm > [Accessed 25 Jan 2010]

Sample from statistical print out (Piet Becker, Biostatistics Unit, MRC)

```
->  symmetry peds_sum asq_sum if (( 1 == peds_sum | peds_sum == 3) & ( 1 ==
asq_sum | asq_sum == 3) & ( 1 <= paed_sum & paed_sum <=
> 3)& (visit ==1))

-------------------------------
           |       asq_sum
 peds_sum  |    1       3    Total
-----------+--------------------
         1 |    8       0        8
         3 |    1      13       14
           |
     Total |    9      13       22
-------------------------------

                                                chi2     df      Prob>chi2
-------------------------------------------------------------------------
Symmetry (asymptotic)                    |      1.00      1         0.3173
Marginal homogeneity (Stuart-Maxwell)    |      1.00      1         0.3173
-------------------------------------------------------------------------

->  kap peds_sum asq_sum if (( 1 == peds_sum | peds_sum == 3) & ( 1 == asq_sum |
asq_sum == 3) & ( 1 <= paed_sum & paed_sum <= 3)& (
> visit ==1))

             Expected
Agreement    Agreement     Kappa    Std. Err.         Z      Prob>Z
-----------------------------------------------------------------
  95.45%       52.48%     0.9043      0.2122       4.26      0.0000
```

18.8.2 Chronological Age Computation and Adjustment for Prematurity

A Guide to Chronological Age Computation and Adjustment for Prematurity

Chronological Age Calculation (with no prematurity)
Write the date of testing as follows: years, months, days and then the child's birthday in the same order just below the testing date. Subtract the birth date from the date of testing, borrowing years, months, and days as needed, (e.g., borrow 1 year to create 12 more months, borrow 1 month to create 30 more days). Then round the days into the nearest month: eliminate days if 15 or fewer, or add one month if 16 or more days.

in the example below, the date of testing (2/7/2006) is expressed as 2006 years, 2 months, 7 days and the date of birth (5/23/2005) is expressed as 2006 years, 2 months, and 7 days. In order to subtract the days, 1 month was borrowed and converted into 30 days and added to the 7 days. Subtracting months required converting 1 year into 12 months and added that to the remaining 1 month. At this point the birthdate could be subtracted. Then the days were rounded (in this case down).

Example of chronological age computation

	Year	Month	Day
	2005	(12 + 1 =13)	(+ 30) = 37
Date of Testing	~~2006~~	~~2~~	~~7~~
Birthdate	2005	5	23
Age	0	8	14
with rounding:			***8 months***

Correcting for Prematurity
For children less than 2 years of age and who were born 4 or more weeks prematurely, adjustments need to be made to chronological age. To do this:

1. Compute chronological age as above.
2. Subtract months premature. If prematurity is reported in weeks, round to the nearest month, e.g., round down for 1 month + 1 - 2 weeks; 2 months, + 1 – 2 weeks, etc. and round up a month if 1 month + 3 – 4 weeks, 2 months + 3 – 4 weeks, etc.. Similarly if prematurity is reported in days, convert days to months (of 30 days each), rounding down if fewer than 15 days beyond the month, and round up to the next highest month if 16 or more days beyond a month.

Example of correction for prematurity.

	Year	Month	Day
	2005	(12 + 1 =13)	(+ 30) = 37
Date of Testing	~~2006~~	~~2~~	~~7~~
Birthdate	2005	5	23
Age	0	8	14 ***days***
with rounding:		***8 months***	
subtract months premature		2	
adjusted age		***6 months***	

8

The guidelines provided throughout the rest of this chapter was drawn from Glascoe FP. Technical Manual for the BRIGANCE© Screens-II. North Billerica, MA: Curriculum Associates, Inc. 2006, and adapted with permission.

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