ORIGINAL ARTICLE

Eight-year Developmental Follow-up in Children with Birthweights 1,251–1,500g in KK Women's and Children's Hospital

Winnie Li Chuen Choo,[1] *MMBS*, Mary Daniel Lourdes,[2] *MMBS, MMed(Paeds), Edm FAMS*

[1]National University of Singapore, Singapore

[2]Department of Child Development, Neonatology, KK Women's and Children's Hospital, Singapore

ABSTRACT

Background: Children of very low birth weight (VLBW, birth weight <1,500g) are at risk of developmental disability and learning difficulties. Regular follow-up to a school-going age allows timely diagnosis and intervention.

Methods: The duration, developmental assessment and service utilisation of 41 VLBW children with birth weights between 1,251–1,500g born in 2001 were retrospectively analysed.

Results: The median follow-up duration was 5 years 11 months. 56% were followed-up till preschool entry. Gross motor, fine motor and language domains were assessed in more than 95% of children. Information on behavioural and learning problems was only sought for in approximately 50% of children.

Conclusions: A significant proportion of these high-risk children were either not followed-up long enough or were not adequately assessed for learning and behavioural problems, thus justifying the development of a new standardised follow-up program for larger VLBW children.

Keywords: VLBW, Neurodevelopmental disability

INTRODUCTION

Children of VLBW are at risk of developmental disability in Singapore and other countries[1–3]. Severe disability includes profound intellectual, hearing or visual impairment and moderate to severe cerebral palsy[4]. Subtle neurodevelopmental disabilities involving cognition, learning, emotion and social skills tend to be missed or misdiagnosed until school age, thus delaying intervention[4]. The detrimental effect on development is cumulative and leads to poor adjustment to formal structured learning, poor academic achievement, the need for special education and reduced permanent employability[5–7]. Early developmental assessment does not reliably prognosticate long-term outcomes, as a diagnosis of developmental disability is unstable from the ages of two to eight[2]. Cognitive disability has the least stability of diagnosis or classification of severity compared to blindness, deafness and cerebral palsy[2]. VLBW children are a high-risk group and should be regularly and systematically monitored for developmental disabilities up to school age[8–9].

At KK Women's and Children's Hospital (KKH) in Singapore, infants weighing 1,250g or less at birth ("lighter VLBW children") undergo a standardised follow-up program from hospital discharge to eight years of age. This starts in a multidisciplinary neonatal clinic with the same neonatologist, a physiotherapist and dietician and continues for two years, after which they are internally transferred to a specialised VLBW follow-up clinic (VLBW FU clinic) in the Department of Child Development (DCD). Formal cognitive assessments are performed at two, five, and eight years with ready access to an educational psychologist for consultations and additional psycho-educational assessments. As children in Singapore enter formal primary education at age seven, this program allows identification of learning problems before/just after school entry.

Infants weighing 1,251–1,500g ("heavier VLBW children") are seen in a general neonatal specialist outpatient clinic (NN SOC) by a neonatologist for eight years. Clinical developmental assessments

are done twice a year until 24 months, then yearly till eight years of age. Referrals for cognitive assessments and to the Rehabilitation Department are performed whenever appropriate by the neonatologist in charge of the clinic, with an optional internal transfer to the VLBW FU clinic run by one of the authors in the DCD. Any child who does not attend a follow-up visit is recalled at least twice by the clinic staff.

Prior to hospital discharge, all VLBW children are referred to an early intervention programme (EIP) conducted by the hospital's Rehabilitation Department. The EIP discharges VLBW children at the age of two if they have no developmental disabilities. The infants also undergo outpatient VLBW screening consisting of an ophthalmological review, hearing screening with otoacoustic emission or automated auditory brainstem response and a cranial ultrasound scan at three corrected months of age.

The duration and quality of follow-up of the larger VLBW children over eight years was retrospectively studied to identify service gaps and to evaluate the need for a standardised programme.

METHODOLOGY

Patient Selection

The study cohort consisted of heavier VLBW children born in KKH from 1 January 2001 to 31 December 2001, who would have completed eight years of follow-up by 2009. Their names were obtained from a prospectively maintained VLBW database in the Department of Neonatology.

Data Collection

Patient records were retrieved from the medical records office after approval to conduct the study was granted by the Institutional Review Board of KKH. The following birth and neonatal details were obtained: gender, race, multiple gestations, gestational age, birth weight (absolute and as a percentile compared to the estimated gestational age), results of VLBW screening, referrals to medical specialists, EIP, dietician, psychologist for cognitive assessment, special schools, or to allied health services.

Between the ages of two to eight, documentation on the presence of gross and fine motor delays, cerebral palsy, speech impairment, expressive/ receptive language delay and the behavioural problem of temper tantrums were sought for. Between the ages of four to eight, case notes were reviewed for the presence of behavioural problems such as inattention, hyperactivity, attention deficit hyperactivity disorder (ADHD), autism spectrum disorders (ASD), emotional problems and poor social skills. The presence of learning difficulties was noted from the ages of six to eight. When any of these developmental problems were found to be present, the age of diagnosis, resolution, referral to allied health services and attendance at the referral service were noted. Absence of documentation regarding these disabilities was also noted.

The duration of follow-up, number of follow-up visits in NN SOC or the VLBW FU clinic and the reason for cessation of follow-up was recorded. An "open date" was a follow-up option where no specific date for follow-up was given to the patient, but where parents could return for a follow-up visit at any time within the next two years. "Recall" referred to the attempts of clinic staff to contact the family and offer another follow-up date when the child did not attend the appointed follow-up. The case notes of children who had been transferred to the DCD were not traced.

Statistical Analysis

The data was analysed using Microsoft Excel 2000.

RESULTS

Patient Demographics

There were 60 eligible heavier VLBW children. Patient records were available in 41 infants (68.3%) who formed the study cohort. 19 (31.7%) of the 60 records were not available from the medical records office during the time of the study as the patients were being seen in clinic, in the process of digitalised, or could not be retrieved.

The median gestational age was 31 weeks (range of 28.3–35.6 weeks). The median birth weight was 1,380g (range of 1,252–1,470g), and at the 10th centile (range below 3rd centile to 75th centile) based on gestational age. 11 (26.8%) were small for gestational age (SGA). 21 (51.2%) were male. 29 (70.7%) were Chinese, nine (22.0 %) were Malay, and three (7.3%) were Indian. 29 (70.7%) were singletons, while 12 were infants of multiple pregnancies where not all were heavier VLBW children. Six, four and two of them were part of twin, triplet and quadruplet pregnancies respectively.

Follow-up

Figure 1 shows the status of follow-up at age eight and the reasons for cessation of follow-up. 14 (34.1%) VLBW children defaulted follow-up after a median of nine follow-up sessions (range of 1–11) and a median duration of three years two months (range of two months to six years eight months). A median of four (range of 0–14) recalls were made for each of them. Five VLBW children defaulted follow-up within a year. A median of two recalls (range of zero to four) were made for each of them. Of the five early defaulters, four did not complete the VLBW screening. One defaulted the hearing screening test (compared to a cohort attendance of 92.7%), two defaulted the outpatient cranial ultrasound screen (compared to a cohort attendance of 80.5%), and one did not have an ophthalmology review (compared to a cohort attendance of 97.6%). One early defaulter had multiple co-morbidities. She was an SGA infant with neonatal referrals to the ophthalmology, endocrine, neurology, genetics and paediatric surgical clinics in the first year of follow-up. She attended the EIP but defaulted neonatal follow-up at 11 months of age despite six recall attempts. The other four were healthy VLBW children with no parental concerns or developmental delay in their first year of life.

11 (26.8%) VLBW children defaulted on at least one appointment but responded to a median of two recalls (range of one to eight). The highest total number of recall attempts (14) was made for a Malay singleton girl after a follow-up duration of three years nine months. She had multiple co-morbidities, an abnormal hearing screen and was on extended ophthalmological follow-up. She had also defaulted the EIP and a referral to the dietician.

28 (68.3%) VLBW children were referred to the EIP. Four (9.8%) did not attend. In 13 VLBW infants, (31.7%) there was no documentation of referral at hospital discharge. 38 (92.7%) attended outpatient hearing screening, five (12.2%) had abnormal hearing screening tests and one (2.4%) was diagnosed with hearing impairment. All 33 (80.5%) who underwent outpatient cranial ultrasound screening had normal results. None of the 40 VLBW children (97.6%) who attended at least one review in the Ophthalmology Department had significant visual impairment. Referrals to the dietician were made for 10 VLBW children (24.4%), including 4 of 11 SGA infants, with nine attending at least one session. 42 referrals to paediatric surgical and medical subspecialties were made for 27 VLBW children (65.9%), most frequently to otolaryngology (nine infants), cardiology (eight

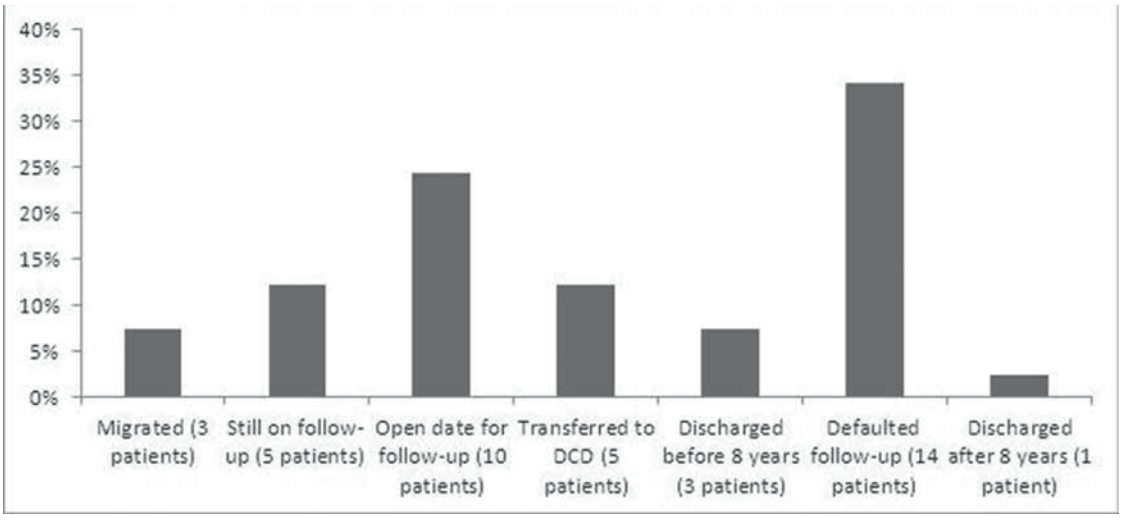

Fig. 1. Status of follow-up at age eight and reasons for cessation of follow-up

Table 1. Number and percentage of VLBW children on follow-up at different age groups.

	Number of VLBW children on follow-up		
Age (years)	NN SOC	VLBW FU	Total VLBW children on follow-up
1	36 (87.8%)	0 (0%)	36 (87.7%)
2	35 (85.4%)	0 (0%)	35 (85.4%)
3	27 (65.9%)	1 (2.4%)	28 (68.3%)
5	20 (48.8%)	3 (7.3%)	23 (56.1%)
7	11 (26.8%)	4 (9.8%)	15 (36.6%)
8	5 (12.2%)	6 (14.6%)	11 (26.8%)

infants), and paediatric surgery (six infants).

Table 1 shows the percentage of VLBW children on follow-up at different ages and the location of follow-up. The six (14.6%) VLBW children who were internally transferred to the VLBW FU clinic for further evaluation were followed-up for a longer median duration (eight years seven months, [range of seven years three months to nine years five months]), and had more median clinic visits 13.5 visits [range of 11–17]) compared to those who were followed-up only in the neonatal specialist outpatient clinic (five years, one month [range of 2 months to 8years and 10 months]). Five (12.2%) were discharged to the DCD ("discharged to DCD") run by developmental paediatricians for developmental problems identified during routine assessments. 30 VLBW children (73.2%) were followed-up only in the neonatal SOC throughout the duration of their follow-up. Excluding five VLBW children discharged to DCD, the median duration of follow-up was 5 years 11.5 months (range of two months to nine years five months), with a median of 12 clinic visits (range of 1–18).

Developmental Assessment

Of the 36 VLBW children (87.8%) followed-up beyond one year, 32 (88.9%) were evaluated with a developmental screening tool (Denver Developmental Screening Test [DDST, Singapore] and/or the Ages and Stages Questionnaire) for developmental problems. The median age of walking was 14 months (range of 8–12 months) as documented in 21 (51.2%) VLBW children. One VLBW child (2.4%) was diagnosed with diplegic cerebral palsy. Two (4.9%) VLBW children were formally assessed by the educational psychologist for cognitive and learning abilities, and had intelligence quotient (IQ) scores of 109 and 76 at eight and nine years of age respectively. One (2.4%) VLBW child was scheduled for an IQ assessment at the time of review. One VLBW child was diagnosed with ADHD and was referred to but did not attend therapy services. The current state of his problem was not known as he defaulted on neonatal follow-up. There were no known diagnoses of ASD among the VLBW children studied, although prevalence of ASD is not known among those referred to the DCD.

Table 2 describes the developmental problems in the six VLBW children referred to the VLBW FU clinic. All had more than one developmental problem. All except one VLBW child with expressive language delay and tantrums attended rehabilitation and psychological services when referred. Developmental problems were detected throughout the course of follow-up, from the age of one year for fine motor delay for one VLBW child to nine years for learning difficulties in another.

DISCUSSION

Duration of Follow-up

The median duration of follow-up of 5 years 11 months is inadequate to definitively identify for late onset or subtle developmental disabilities, in particular neurodevelopmental disabilities which may cause significant academic difficulty[2,4]. Follow-up to a school-going age of eight years may allow problems related to transition from pre-school to primary school, and academic difficulties associated with subtle disabilities to be detected and addressed.

Cessation of Follow-up

Cessation of follow-up was most often due to defaulting appointments (34%) and failure to respond to a recall. The five VLBW children who

Table 2. Developmental problems in six VLBW children referred to the VLBW clinic in DCD.

Developmental problem	Number of VLBW children	Median age of detection	Referred to rehabilitation/psychological services
Learning difficulties	4	8 years (range of 8–9 years)	3
Inattention	4	5 years 6 months (range of 4–9 years)	1
Tantrums	4	4 years (range of 2–4 years)	1
Expressive language delay	3	1 year 11 months (range of 1 year 4 months to 9 years)	3
Receptive/auditory language delay	1	9 years	1
Fine motor delay	1	1 year	1
Hyperactivity	1	6 years	0
Emotional problems	1	6 years	1

defaulted on neonatal follow-up early within a year of life all missed routine outpatient screening services scheduled between three to five months of corrected age compared to the high take-up rates in the other VLBW children. It may be useful for doctors following-up future cohorts to enquire about the reasons for missing their screening as underlying issues may emerge, such as financial issues, social issues, or lack of understanding which might need referrals to medical social services or greater emphasis on the benefits of regular follow-up.

7% were discharged and 25% were given open dates for follow-up before eight years of age. This may be due to parental preference to visit a primary health doctor. This may also be due to unfamiliarity with the follow-up protocol amongst doctors during the 2001 SARS outbreak in Singapore when doctors were divided into outpatient and inpatient teams to avoid cross-infection.

Developmental Assessment

88.9% of 36 VLBW children followed-up beyond one year were assessed with a developmental screening tool. As the DDST (Singapore) is the only locally normed developmental tool and is available in part in the health booklet, it is the most commonly used tool. However, it may cause under-referral in high-risk populations like VLBW children, due to its high specificity but low sensitivity[10–11].

A literature review showed that clinically significant emotional and behavioural problems tended to be under-recognised by doctors seeing VLBW children, although they were present in 25–55% of VLBW children compared to a consistently lower control rate of 7%[12]. Only 4% of VLBW children were referred to a consultant psychiatrist[8]. Emotional problems were also under-reported by parents of VLBW adolescents[13], and parents of VLBW children may also be subject to the same trend of under-reporting emotional problems.

In this study, the prevalence of behavioural problems of 43.9% was higher than a non-referral outpatient population prevalence of 22.8% reported in the literature[14]. However, referral for behavioural management was rare at 4.9%, possibly due to conflicts in parental priorities, parental reluctance or lack of knowledge of appropriate services. In 9.8% of our patients, parents reported behavioural problems that subsequently resolved without intervention.

The rates of hearing screening (92.7%) and ophthalmic reviews (97.6%) were high, while approximately 20% did not undergo cranial ultrasound scanning. This could be due to parental default of appointments or failure of the doctor to order the scan. Parents may not have understood that cranial ultrasound was an important diagnostic tool for parenchymal damage with long-term consequences.

Table 3. Developmental problems in VLBW children (n=41).

Developmental problem	Documentation of presence or absence of delay	Prevalence	Referral to an allied health service	Attendance at allied health service	Documented resolution
Behavioural problems	24 (58.5%)	18 (43.9%)	2 (4.9%)	1 (2.4%)	4 (9.8%)
Language impairment	39 (95.1%)	14 (34.1%)	6 (14.6%)	6 (14.6%)	3 (7.3%)
Gross motor delay	40 (97.6%)	5 (12.2%)	5 (12.2%)	5 (12.2%)	3 (7/3%)
Fine motor delay	39 (95.1%)	5 (12.2%)	3 (7.3%)	3 (7.3%)	1 (2.4%)
Learning difficulties	24 (58.5%)	5 (12.2%)	3 (7.3%)	2 (4.9%)	0
Emotional problems	14 (34.1%)	2 (4.9%)	1 (2.4%)	1 (2.4%)	0
Poor social skills	30 (73.2%)	2 (4.9%)	0	0	0

Gross motor, fine motor and language domains were assessed more frequently than learning disabilities. Table 3 shows the former were assessed in more than 90% of VLBW children compared to learning difficulties in only 58.5% of them. It is possible that gross and fine motor delay were more easily and objectively elicited than neurodevelopmental disabilities, leading to a higher rate of assessment and documentation. However, learning difficulties should be proactively identified, as VLBW children are at risk of lower IQ scores, poorer academic performances and need for remedial education[15]. Although learning difficulties were reported in 12% and language difficulties in 34%, only 7.3% were formally assessed for cognitive and learning abilities with full-scale IQ assessments. This may suggest a lack of familiarity of the referral system, failure to identify children who may benefit from them or parental reluctance. As IQ and other cognitive assessments are useful in identifying specific learning disabilities, the threshold for administering them should be lower[7].

Referrals for Medical and Surgical Conditions

65.9% of VLBW children had a neonatal referral to a paediatric medical or surgical subspecialty. This may be attributable to a higher rate of chronic conditions in VLBW children and somatic conditions in preterm children[16–17]. The high rate of referrals, particularly to paediatric otolaryngology, may suggest that parents were more familiar with identifying somatic symptoms as worrying than signs of developmental delay.

CONCLUSION

This audit showed significant service gaps in the follow-up of this high-risk group of VLBW children. The median follow-up duration was not long enough to ensure identification of learning difficulties prior to/after school entry. Inadequate emphasis was placed on learning and behavioural problems which can affect school performance. Based on these findings, a new standardised follow-up program for larger VLBW children is currently being developed.

Acknowledgement

The authors wish to thank Dr Yveline Chew for her help in developing the questions for the study.

REFERENCES

1. Daniel LM, Lim SB, Clarke L. Eight year outcome of very-low-birth-weight infants born in KK Hospital. Ann Acad Med Singapore 2003;32(3):354–61.
2. Roberts G, Anderson PJ, Doyle LW. Neurosensory disabilities at School Age in Geographic Cohorts of Extremely Low Birth Weight Children Born Between the 1970s and 1990s. J Pediatrics 2009;154(6):829–34.
3. Mu SC, Tsou KS, Hsu CH, Fang LJ, Jeng SF, Chang CH, et al. Cognitive development at age 8 years in very low birth weight children in Taiwan. J Formos Med Assoc 2008; 107(12):915–20.
4. Davis DW. Cognitive Outcomes in School-Age Children Born Prematurely. Neonatal Netw 2003;22(3):27–38
5. Sykes DH, Hoy EA, Bill JM, McClure BG, Halliday HL, Reid MM. Behavioral adjustment in school of very low birthweight children. J Child Psychol Psychiatry 1997;38 (3):315–25.
6. Schraeder BD, Heverly MA, O'Brien C, Goodman R. Academic achievement and educational resource use of very low birth weight survivors. Pediatr Nurs 1997;23(1): 21–5.

7. Løhaugen GC, Gramstad A, Evensen KA, Martinussen M, Lindqvist S, Indredavik M, et al. Cognitive profile in young adults born preterm at very low birthweight. Dev Med Child Neurol 2010; 52(12):1133–8.
8. Lind A, Korkman M, Lehtonen L, Lapinleimu H, Parkkola R, Matomäki J, et al. Cognitive and neuropsychological outcomes at 5 years of age in preterm children born in the 2000s. Dev Med Child Neurol 2011;53(3):256–62.
9. Fazzi E, Orcesi S, Telesca C, Ometto A, Rondini G, Lanzi G. Neurodevelopmental outcome in very low birth weight infants at 24 months and 5 to 7 years of age: Changing diagnosis. Pediatr Neurol 1997;17(3):240–8.
10. Ahsan S, Murphy G, Kealy S, Sharif F. Current developmental surveillance: Is it time for change? Ir Med J 2008;101(4):110–2.
11. Meisels SJ. Can developmental screening tests identify children who are developmentally at risk? Pediatrics 1989;83(4):578–85.
12. Hayes B, Sharif F. Behavioural and emotional outcome of very low birth weight infants – literature review. J Matern Fetal Neonatal Med 2009;22(10):849–56.
13. Dahl LB, Kaaresen PI, Tunby J, Handegård BH, Kvernmo S, Rønning JA. Emotional, behavioral, social, and academic outcomes in adolescents born with very low birth weight. Pediatrics 2006;118(2):e449–59.
14. Bhatia MS, Dhar NK, Singhal PK, Nigam VR, Malik SC, Mullick DN. Prevalence and etiology in a non-referral outpatient setting. Clin Pediatr 1990;29(6):311–5.
15. Botting N, Powls A, Cooke RW, Marlow N. Cognitive and educational outcome of very low birthweight children in early adolescence. Dev Med Child Neurol 1998;40(10): 652–60.
16. Hack M, Flannery DJ, Schluchter M, Cartar L, Borawski E, Klein N. Outcomes in young adulthood for very low birth weight infants. N Engl J Med 2002;346(3):149–57.
17. Tideman E, Ley D, Bjerre I, Forslund M. Longitudinal follow-up of children born preterm: somatic and mental health, self-esteem and quality of life at age 19. Early Hum Dev 2001;61(2):97–110.