A review of 404 'late walkers'

J DE Z CHAPLAIS AND J A MACFARLANE

Community Child Health Department of Paediatrics, John Radcliffe Hospital, Oxford

SUMMARY A survey of all known 18 month old Oxfordshire children who had not yet walked unassisted and who were born in the four year period between July 1, 1976 and June 30, 1980 was carried out. A total of 275 children aged 18 months with no previously suspected cause for late walking were referred by health visitors; 257 of these children were assessed neurologically and developmentally by a paediatrician at home. Nine cases of cerebral palsy (3.5%) and 6 cases of minor neurological abnormality (2.3%) were newly diagnosed. A register of all other 18 month old 'late walkers' (129) who were already known to paediatricians and were either normal (17) or had known causes for late walking (112) was compiled for the same four year period. The total incidence of pathology among all late walkers (404) in these two groups was 32%.

Methods

All health visitors and general practitioners in the area were asked to refer any child born after July 1, 1976 who had not taken five steps unassisted by 18 months of age (corrected for gestation). Referred children were visited at home by a paediatrician for examination and developmental assessment using the Denver scale. All children suspected of having a neurological abnormality were referred to Professor J P M Tizard at the John Radcliffe Hospital for final diagnosis.

The survey did not set out to be a fully comprehensive population screen since it had to allow for a population migration rate of 10% per year. From a previous survey of health visitor contact we had estimated that only 74% of all children in the area are assessed by child health clinic doctors and health visitors in clinics and a further 10% are seen at home. As it is also likely that referral of late walkers by hospital paediatricians for the register was incomplete, a small number of late walkers will inevitably have slipped through the net.

Results

The total number of late walking children traced during the four year period was 404, giving an incidence of 1.6% for late walking in the 18 month old Oxfordshire population. For reasons given above this is most probably an underestimate; the incidence of late walking among Newcastle born children, for example, is approximately 3%.¹

Seventeen of the 129 late walkers already being seen by paediatricians were normal and 112 had known causes for late walking. Table 1 gives details of the range of conditions responsible for late walking among these children.

Ten of the 275 children referred by health visitors were not seen because either parents or general practitioners were not in favour of them taking part in the survey; 8 children were not seen because referral for the survey was delayed. As much information as possible about these 18 children was gathered from questionnaires and they are presumed normal.

Table 1 *Conditions associated with late walking in children assessed by paediatricians before 18 months of age and living in Oxfordshire at 18 months of age between 1978 and 1982*

Cerebral palsy (all forms)	40
Neurologically abnormal	3
Educationally subnormal (moderate)	11
Educationally subnormal (severe)	5
Congenital syndromes or diseases (with or without mental retardation)	19
Congenital central nervous system abnormality	10
Arrested hydrocephalus	3
Congenital orthopaedic abnormality	8
Down's syndrome	9
Acquired disease	
Spinal tumour	1
Chronic renal failure	1
Muscular dystrophy	
Duchenne	1
Congenital	1
Total	112

A total of 242 of the 257 late walkers newly assessed for the survey at home had no obvious clinical or neuropathological cause for late walking at the time of assessment. (These children are referred to as 'idiopathic' late walkers.) Two children included in the normal group are currently under assessment, one for delayed speech with a probable specific language disorder and one for global developmental delay, possibly borderline educationally subnormal (M). The remaining 15 abnormal children included 9 with cerebral palsy and 6 with minor neurological abnormalities (see Table 2). Thus the total incidence of newly diagnosed neurological abnormality among 257 late walkers was 5.8%. One of the children with minor neurological abnormalities seemed to be normal when assessed at age 18 months but was subsequently referred to a paediatrician with gross and fine motor problems. The cases of cerebral palsy are classified in Table 3 together with details from the perinatal histories of these children.

Data on the pattern of locomotion before walking, family history of late walking, and Denver developmental screening test were analysed for the 242 idiopathic late walkers. Bottom shuffling was the predominant means of early locomotion and was found in 48% of children (Table 4). (The incidence of bottom shuffling in a normal population is reported to be between 8% and 9%.^{2 3}) Forty five per cent of late walkers crawled, 3.5% crept, and 3.5% had no means of locomotion before walking. A family history of walking later than 18 months was found among first degree relatives of 50% of all late walkers compared with 16% of normal walking 18 month old control children studied for the prospective series of the cerebral palsy survey. The 50% incidence of heredity among late walkers is presumably an underestimate since the family history was not always remembered, or grandparents were not available for information. A family history of late walking was equally likely to be found among crawlers as among the collective group with deviant early locomotion. (This information was collected from parents of 226 late walkers, the remaining 16 being not contactable by questionnaire.) The age of walking of 241 idiopathic late walkers is expressed in cumulative centile form in the Figure. (One child is

Table 2 Incidence of newly diagnosed neurological abnormality among 257 late walkers seen at home

Abnormality	No (%)
Cerebral Palsy	9 (3.5)
Minor neurological abnormality	6 (2.3)
Total neurological abnormality	15 (5.8)

Table 3 Perinatal histories of 9 children with cerebral palsy

Case No	
1	<i>Diagnosis: spastic paraplegia.</i> Mother had previous stillbirth. Born at term. Birth asphyxiated with cord round neck. Required intubation and resuscitation. Showed cerebral irritability for 48 hours requiring sedation with phenobarbitone. Referred to orthopaedic surgeons at 1 year with suspected talipes equinovarus deformity.
2	<i>Diagnosis: spasticity of left arm.</i> Normal term delivery. Presented at 3 months with idiopathic hydrocephalus controlled by ventriculoperitoneal shunt with no apparent complications and was normal neurodevelopmentally up to 1 year.
3	<i>Diagnosis: spastic paraplegia.</i> First of monovular twins born at 33 weeks' gestation. Birthweight 1880 g. A number of mild neonatal complications including transient hypoglycaemia, mild respiratory distress syndrome, non-haemolytic streptococcal infection of maternal liquor with colonisation of baby and umbilical infection; moderate jaundice.
4	<i>Diagnosis: spastic paraplegia.</i> Delivery induced at 36 weeks for haemolytic disease. Birthweight 2360 g. Moderately affected by rhesus haemolytic disease. Cardiac arrest for 6 minutes after exchange transfusion. Jittery episodes over following week. Seemed normal at 6 months follow up.
5	<i>Diagnosis: left hemiplegia.</i> Normal term delivery. Birthweight 3030 g.
6	<i>Diagnosis: spastic diplegia.</i> First of twins born to 18 year old unmarried mother at 29 weeks. Birthweight 1120 g. Mild respiratory distress syndrome; moderate jaundice. Had right sided focal fit at 5 weeks of age.
7	<i>Diagnosis: spastic paraplegia.</i> Spontaneous rupture of membranes at 28 weeks, followed by antepartum haemorrhage. Born by normal delivery at 31 weeks' gestation. Birthweight 1680 g. Mild respiratory distress syndrome in neonatal period. Prolonged jaundice due to haemolytic anaemia of unknown cause. Seemed normal when discharged from follow up at 8 months of age.
8	<i>Diagnosis: spastic paraplegia.</i> Mother had 1 previous stillbirth at 28 weeks and 1 neonatal death at 28 weeks lower segment caesarean section. Born by lower segment caesarean section after premature labour at 34 weeks. Birthweight 2120 g. No severe neonatal complications.
9	<i>Diagnosis: spastic diplegia.</i> Born at home at term. No records of delivery available. No reported complications.

Table 4 Method of locomotion before walking in 242 late walkers

Method	No (%)
Bottom shuffle	116 (48)
Creep	9 (3.5)
Sedentary	9 (3.5)
Crawl	108 (45)

still not walking aged 27 months.) Approximately 50% were walking by 19½ months and 97% by 2 years of age.

When assessing the overall development of the idiopathic late walkers it was decided to omit from

Figure Age of first walking of 242 late walkers.

data analysis 27 of the children who had been seen previously by paediatricians for reasons including developmental delay. This was because, not surprisingly, these children's Denver scores included a significantly higher proportion (37.4%) of abnormal or questionable scores than the sample as a whole (13.6%). The Denver scores were assessed according to the revised method of Frankenburg *et al*⁴ and their significance in terms of necessity for review or referral of children was assessed according to Bryant's criteria.⁵ Analysis of the scores of the 215 late walkers about whose development there had been no previous concern (see Table 5) showed that 192 (89%) had normal development in sectors other than gross motor (those who had started walking by the time of assessment had, of course, normal gross motor development as well). The remaining 23 (11%) had delays in social, fine motor, or language sectors which would together with a persistently abnormal gross motor performance qualify them for referral according to Bryant's criteria. This group included four children with abnormal or questionable scores in two sectors other than gross motor.

The birth records of the 242 idiopathic late walkers showed that 205 children (85%) had had normal births. Of the remaining 15% with abnormal

perinatal events, 18 (7%) were preterm, 16 (7%) were small for dates, and 3 (1%) were severely birth asphyxiated. The preterm group included four twins and two triplets.

Discussion

Although the Oxford late walkers survey has shown a high overall incidence of pathology (32%) among the late walking population this was mainly concentrated in the third of children destined to walk late, who presented and were diagnosed before 18 months of age. Screening the remaining two thirds of late walkers at 18 months in the community brought to light a much smaller incidence of pathology amounting to 12.5% of the total pathology among all late walkers.

Incidence of neuropathology among late walkers.

Two previous surveys have recorded the incidence of cerebral palsy among selected groups of late walkers referred to specialist centres—Lundberg² found an incidence of 3.8% among 65 Swedish children with gross motor delay only and Robson⁷ found a 14% incidence of cerebral palsy among a group of 51 south London 'bottom shufflers'. In both surveys the groups of children were selected from records of hospital referrals for developmental delay or suspected neuropathology. The Oxford survey has attempted to include the whole range of late walkers in the community and its findings are therefore particularly useful for those working in developmental surveillance in the community. Neurologically abnormal late walkers in the Oxford survey (5.8%) had not been picked up at routine 18 month developmental checks because they did not stand out clinically as an abnormal group and their neurological signs when first seen were subtle. The 8 children with cerebral palsy represent the mild end of the spectrum, most moderate and all severe cases being diagnosed at less than 1 year of age regardless of age of walking. Robson³ points out the considerable degree of overlap in terms of motor milestones between the normal motor delayed and the mildly motor handicapped child populations. The mean age of walking for hemiplegic children in Robson's

Table 5 Denver test performance of 215 'idiopathic' late walkers

	No (%)
Children with gross motor delay only	192 (89)
Children with abnormal or questionable performance in 2 or more Denver categories	23 (11)
Total	215

Table 6 Mean ages of diagnosis and proportions of late walkers among 15 diplegic and 15 hemiplegic children in Oxfordshire (1976 to 1980).

	Diplegic	Hemiplegic
Mean age of diagnosis	17 months	12.5 months
% late walkers	100%	50%

sample at Guy's Hospital was 15 months and that for diplegic children was 24 months, hence the normal late walking child population falls between these two means. Table 6 shows the respective mean ages at diagnosis and the proportions of late walkers among 30 diplegic and hemiplegic children born between 1976 and 1980 and resident in Oxfordshire at 18 months of age. The later mean age of diagnosis of diplegic children may be partly related to the later age of walking of this group compared with the hemiplegic group. It also reflects the unpredictable and sometimes fluctuating neuropathological process described by Robson⁷ during which the initial phase of hypotonia in infancy progresses via a phase of intermittent extensor activity to overt spasticity by age 18 to 24 months. In the case of three late walkers referred for the Oxford survey the gradual evolution of signs of spasticity did not allow a definite diagnosis to be made for periods of between 6 and 18 months after initial referral. Furthermore, one child confidently diagnosed at 18 months as having ataxic cerebral palsy showed subsequent regression of neurological signs and was reclassified at 2 $\frac{3}{4}$ years of age as having a minor motor disorder, but not cerebral palsy.

Clinical distinction between pathological and idiopathic late walkers. Since it may be extremely hard to discriminate the normal from the abnormal late walker at 18 months of age those working in the community have to steer a course between under and over referral. Granted that there is considerable overlap in clinical signs and developmental patterns between 'idiopathic' and 'symptomatic' cases at 18 months, a number of factors seem to favour a normal outcome. These include a normal pre-, peri-, and postnatal history; shuffling with heredity for shuffling; and an abnormal or late pattern of learning to sit.² To this could be added a family history of late walking in either parent or a sibling, which might enable reassurance to be given to parents of half of neurologically normal crawlers seen in the community. With regard to clinical presentation normal, bottom shuffling late walkers characteristically show a mild to moderate degree of hypotonia with an extended range of joint mobility resulting in the frequent finding of hyperextended knees and planovalgus feet. This syndrome, which made up 25% of Lundberg's late walking population, causes great concern to some parents and health visitors but it should be the most reassuring finding. In the Oxford survey the feet of normal children who walked late would invariably dorsiflex with ease past the neutral position with legs extended, in contrast to the findings in 6 of 7 diplegic children who walked late who showed increased

resistance to passive dorsiflexion at the ankle together with a tendency to stand or walk round the furniture on the toes. A history of bottom shuffling per se should not, however, be taken as reassurance since according to Robson⁶ one in 8 children presenting with spastic diplegia is a 'shuffler'.

Predictive value of assessment of 18 month old late walkers. The importance of early delays in motor development in terms of future performance is not fully understood. The population of children who walk late is mixed—some children are genetically destined to show an isolated gross motor delay, some have more generalised or global delay, in some development has been temporarily delayed by adverse perinatal events, and some have minor neurological abnormalities that may or may not persist. Moreover, the effects of social deprivation and understimulation interacting with the above factors are also important, though difficult to quantify.

Neligan and Prudham⁷ compared the performances of early (before 10 months) and late walkers (later than 17 months) at school entry age. They found a significant difference on testing for language and fine motor skills among boys but not among girls.

Silva⁸ carried out a three year assessment of 1037 children in Dunedin, New Zealand who were followed up prospectively from birth. The early motor delayed group were 5 to 6 times more likely to be represented among those with very low language and fine motor performance at three year follow up compared with the normal range of children. Between half and a third of children with early motor delay scored within the average or better than average range on later testing. (It is not clear whether this survey included any children with neuropathological or other reasons for late walking.) The long term outcome of the idiopathic group of Oxfordshire late walkers must await the forthcoming school entry age follow up study.

This work was funded by the Leverhulme Trust. We gratefully acknowledge the help of Miss R King.

References

- Neligan G, Prudham D. Norms for four standard developmental milestones by sex, social class and place in family. *Dev Med Child Neurol* 1969;11:413-22.
- Lundberg A. Dissociated motor development. Developmental patterns, clinical characteristics, causal factors and outcomes, with special reference to late walking children. *Neuropediatrics* 1979;10:161-82.
- Robson P. Screening for children. Developmental paediatrics. *R Soc Health J* 1978;98:231-7.
- Frankenburg MD, Goldstein AD, Camp BW. The revised Denver developmental screening test. *J Pediatr* 1971;790: 988-95.

- ⁵ Bryant GM. Use of the Denver developmental screening test by health visitors. *Health Visitor* 1980;**53**:2–5.
- ⁶ Robson P, MacKeith RC. Shufflers with spastic diplegic cerebral palsy: a confusing clinical picture. *Dev Med Child Neurol* 1971;**13**:651–9.
- ⁷ Neligan G, Prudham D. Potential value of four early developmental milestones in screening children for increased risk of later retardation. *Dev Med Child Neurol* 1969;**11**:423–31.
- ⁸ Silva PA. The significance of early delays in motor development. *New Zealand Journal of Health, Physical Education and Recreation* 1979;**12**:78–83.

Correspondence to Dr J A Macfarlane, Community Health Offices, The Radcliffe Infirmary, Oxford OX2 6HE.

Received 20 February 1984

Fifty years ago

The intradermal tuberculin reaction

J W E CORY

‘The significance of negative reaction.—A negative skin reaction to an ordinary diagnostic dose of tuberculin (0.1 cc of 1 in 100 dilution) may mean:

- (1) That the child has not been infected with tuberculosis.
- (2) That the test has not been properly performed or the tuberculin used is inactive.
- (3) That the child is already so ill with tuberculosis that its skin sensitivity is too much diminished to react.
- (4) That, although the child is suffering from tuberculosis, an intercurrent fever, such as measles has caused a temporary diminution of the skin sensitivity and that if repeated during convalescence a positive result may be obtained with the same dose.

The findings here reported agree with those of Mantoux who wrote in 1910. ‘A negative test except in measles, meningitis and miliary tuberculosis and advanced cases with marked toxæmia is an argument of the first order in excluding clinical tuberculosis; contrary to most clinical methods the value of the intracutaneous test lies in its negative results.’

(Completely true 50 years later. RONALD ILLINGWORTH)

Archives of Disease in Childhood 1934;**9**:177–88.