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Ability of independently ambulant children with cerebral palsy to ride a two-wheel bicycle: a case–control study

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ABBREVIATIONS

GMFCS Gross Motor Function Classification System

[Abstract]

AIM Limited information exists on the ability of children with cerebral palsy (CP) to ride a two-wheel bicycle, an activity that may improve health and participation. We aimed to describe bicycle-riding ability and variables associated with ability to ride in children with CP (Gross Motor Functional Classification System [GMFCS] I–II) compared with children with typical development.

METHOD This case–control study surveyed parents of 114 children with CP and 87 children with typical development aged 6 to 15 years (115 males, mean age 9y 11mo, standard deviation [SD] 2y 10mo). Kaplan–Meier methods were used to compare proportions able to ride at any given age between the two groups. Logistic regression was used to assess variables associated with ability to ride for children with CP and typical development separately.

RESULTS The proportion of children with CP able to ride at each level of bicycle riding ability was substantially lower at each age than peers with typical development (p<0.001). While most children with typical development were able to ride independently by 10 years of age, 51% of children with CP classified as GMFCS level I and 3% of those classified as GMFCS level II had obtained independent riding in the community by 15 years of age. Variables associated with ability to ride for children classified as GMFCS level I were age and parent-rated importance of their child being able to ride.

INTERPRETATION Some independently ambulant children with CP can learn to ride a bicycle, in particular if they are classified as GMFCS level I. Variables associated with ability to ride deserve consideration in shaping future efforts for the majority of this population who are not yet able to ride.

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What this paper adds

- Skill acquisition was delayed in children with cerebral palsy who had learnt to ride.
- At each age, a lower proportion of children with cerebral palsy (Gross Motor Functional Classification System [GMFCS] levels I–II) learn to ride compared with peers with typical development.
- More children with cerebral palsy classified as GMFCS level I were able to ride compared with those classified as GMFCS level II.
- Parent-rated importance and age are associated with ability to ride in children classified as GMFCS level I.

[Main text]

On average, children with cerebral palsy (CP) have lower levels of participation in physical and recreational activities than their peers with typical development.[1] Low levels of physical activity in childhood have been linked to the development of chronic disease in adulthood.[2] Physical activity has been associated with better quality of life and happiness in children with CP.[3] Bicycle riding may be a means of increasing physical activity and improving overall health and participation in this population. Existing literature on stationary bikes suggests improvements in terms of strengthening,[4] endurance,[4] muscle activation,[5] quality of life,[6] and bone mineral density.[7] Further, the positive effects of cycling as a peer, family, and community activity in the general population are well known.[8]

Clinical experience suggests that independently ambulant children with CP (Gross Motor Function Classification System levels [GMFCS] I-II)[9] can learn to ride a two-wheel bicycle given the right conditions. Learning to ride a two-wheel bicycle is a common childhood milestone, and can be a goal for children with CP who want to participate in a similar activity to their peers or family. However, there is no published literature documenting the proportion acquiring this skill and the age at which they do so. Moreover, there are no studies examining how different these outcomes are compared with children with

typical development, or the factors associated with skill acquisition. This information is important to consider in order to determine whether interventions to improve bicycle riding for children with CP can, and should, be implemented.

Some data are available on bicycle riding in the general paediatric population. Normative data collected for the Dutch calibration of the Paediatric Evaluation of Disability Inventory suggests 50% of Dutch children can ride a two-wheel bicycle for 50m, with help getting on and off, by the age of 4 years and 6months.[10] Results of a Norwegian study report the mean age at cycling debut for children with typical development as 5 years and 5 months, with 87% of children aged 4 to 15 years riding a bicycle for longer than 1 hour per week.[11] In Australia, 64% of children aged 5 to 14 years are reported to participate in bicycle riding at least fortnightly,[12] but data regarding proportions able to ride and age at skill acquisition are lacking.

The primary aim of this study was to determine the proportion of independently ambulant children with CP (GMFCS levels I and II) able to ride a two-wheel bicycle at any given age, and their average age at skill acquisition, compared with children with typical development. The secondary aim was to examine variables associated with ability to ride a two-wheel bicycle in each group. It was hypothesized that the proportions of children with CP able to ride would be lower than children with typical development at each age, and skill acquisition would occur at an older age, if at all.

METHOD

Ethics approval for this case–control study was granted by the Human Research Ethics Committee at the Royal Children's Hospital, Melbourne, Australia.

Participants

Parents or primary carers of children were eligible to participate if they met the following inclusion criteria: (1) had a child aged 6 to 15 years; (2) had a child able to walk 50m without a mobility aid or physical assistance (GMFCS level I–II if child with CP); (3) child and family living in the state of Victoria, Australia; (4) English-language proficiency; (5) current contact details were available.

Recruitment

Participants were recruited and data collected over an 8-week period in April and May 2015. Written informed consent was obtained from participants.

Children with cerebral palsy

Eligible parents or primary carers of children with CP were identified from the Victorian Cerebral Palsy Register, an ongoing register of individuals with CP who were born or resident in the Australian state of Victoria from 1970 onwards. Invitations and follow-up reminders to complete an online survey were sent by mail or e-mail to 662 families. The study was also advertised on a CP research centre website and through paediatric rehabilitation clinics.

Typically developing controls

Recruitment of parents or primary carers of children with typical development was primarily via snowball recruitment. Snowball recruitment meant parents invited through the CP register were also invited to respond on behalf of any 6 to 15-year-old siblings of children with CP and to pass on the invitation to other eligible parents of children with typical development. Additional methods of recruitment for the typically developing group included advertisements on the website of a children's research institute and invitation to a convenience sample of eligible parents known to the study investigators.

Participants were excluded from both groups if their child had other developmental disabilities, visual impairment, or medical conditions that could affect their ability to ride a bicycle.

Data collection

A customized web-based survey was designed and managed using REDCap electronic data capture tools.[13] The survey comprised 20 questions related to the following three domains: child characteristics (10 items; see Table SI, online supporting information), parent and family characteristics (3 items; see Table SI), and their child's bicycle riding ability (7 items [see Table I] + age at skill acquisition). Further details regarding survey administration can be obtained from the authors on request.

Child characteristics

Child characteristics included parent-rated GMFCS level (cerebral palsy group only),[9] age, sex, birth order, intellectual status,[14] schooling type, prior experience on other types of bicycles, and prior experience with bicycle-specific therapy or programmes.

Parent and family characteristics

Parent and family characteristics included sex of the participating parent and residential postcode, which was collected to provide a measure of social advantage/disadvantage and remoteness, using the Index of Relative Socio-economic Advantage and Disadvantage and the Accessibility and Remoteness Index of Australia respectively.[15,16] Parents/carers also provided a rating of importance of their child being able to ride a bicycle, and family interest in bicycle riding, on a five-point adjectival scale.

Child's bicycle riding ability

Participants reported their child's ability to ride by stating whether their child had or had not achieved each level of ability, using the descriptors shown in Table I, at the time of data collection. Age at skill acquisition was defined as the age at which the child could ride a two-wheel bicycle for 20m independently but may need help to start.

Statistical methods

Sample size

Sample size calculations indicated that 188 participants (94 per group) would be sufficient to detect a clinically meaningful difference of 20% in bicycle-riding ability between the CP and typically developing groups, based on the assumption that 70% of the children with typical development could ride a bicycle,[11] and assuming 80% power (α=0.05).

Statistical analysis

Descriptive statistics were used for presentation of within-group results. Group characteristics were compared using χ^2 tests for categorical data and *t*-tests for continuous variables. To compare ability to ride at each age between the children in the CP and typically developing groups, while accounting for a spread of ages in each group, the proportion able to ride at each age with its standard error was estimated and graphed using the Kaplan–Meier method for each group. χ^2 analysis was used to compare the proportion of children able to ride as a group, and the Wilcoxon rank-sum test was used to compare median age at skill acquisition.

Logistic regression analysis was used to determine variables associated with ability to ride within each group separately. Crude analysis was initially undertaken involving univariate logistic regression to examine the relationship between variables and ability to ride. Variables with significant differences ($p \leq 0.050$) in the proportion of children reported as

able to ride on univariate analysis were analysed for independence using a forward stepwise multivariate logistic regression model, adjusted for differences between the groups. Variables were removed from the model if the *p*-value was greater than 0.100. Data were analysed using Stata version 13.1 (StataCorp, College Station, TX, USA).

RESULTS

A total of 201 responses were collected from the parents of 114 children with CP and parents of 87 children with typical development. The response rate was 17% from 663 invited parents of children with CP. A response rate could not be determined for the control group given the snowball recruitment strategy.

Child, parent, and family characteristics are shown in Table SI. Excluding differences associated with CP, significant differences ($p<0.050$) between the groups were found in the following child-related variables: age, prior experience on a trike, and prior experience on a balance bike (also known as a walker bike); and the family-related variables of relative socioeconomic advantage/disadvantage (Index of Relative Socio-economic Advantage and Disadvantage)[15] and geographical remoteness (Accessibility and Remoteness Index of Australia).[16] The groups were well matched for child's sex and birth order, and other parent- and family-related variables.

Ability to ride a bicycle

Proportion able to ride at different ages

There were clear differences between the CP and typically developing groups in the proportions able to ride at each year of age ($p<0.001$; Fig. 1 and Table II). These results relate to the child being able to ride at level I (able to ride 20 m independently but may have help to start); however, similar proportions and between-group differences were seen for ability to ride at the other levels of riding ability described in Table I. Moreover, only six children in the CP group (GMFCS level I, $n=5$; GMFCS level II, $n=1$) and three in the typically developing group who could ride at the least challenging level (level I) had not yet accomplished riding at the most challenging level (level VI). Taking into account differences in ages between the groups and those who were not able to ride, 50% of the children in the CP group were estimated to be able to ride at age 10 years and 11 months versus 5 years for children in the typically developing group ($p<0.001$) (see Fig. 1).

Median age at skill acquisition

For those children with CP who did learn to ride, the median age at skill acquisition reported by parents was 6 years and 5 months (interquartile range 5y 2mo–9y 0mo; range 3y 6mo–12y 6mo). This was significantly later than the median age at skill acquisition reported by parents of children with typical development (median 5y, interquartile range 4y 5mo–5y 7mo; range 3y–10y 10mo [$p<0.001$]).

Variables associated with ability to ride

For this analysis, ability to ride was defined as the child being able to ride a two-wheel bicycle independently in the community, the most challenging level of ability (level VI; Table I). On univariate analysis, ability to ride in the CP group was associated with GMFCS level, current age, the importance parents placed on their child being able to ride a bicycle, and experience on a balance bike (Table III).

Only one child of the 33 (3%) classified as GMFCS level II was reported as able to ride independently in the community, whereas 41 of the 81 (51%) children classified as GMFCS level I were reported to ride at this level. Given that this child also had missing values for other variables included in the model, multivariate logistic regression analysis was undertaken for children with CP classified as GMFCS level I only. However, this fact, combined with the results of univariate analysis, provides strong evidence to suggest that children classified as GMFCS level II are less likely to be able to ride than children classified as GMFCS level I.

Results of the multivariate analysis (Table III) suggest that for children with CP (GMFCS level I), ability to ride independently increases with each year of age, and parents placing higher importance (important or very important) versus lower importance (not important to somewhat important) on their child riding. Furthermore, ability to ride independently is positively associated with having experience on a balance bike in a child with CP (GMFCS level I); however, this was not significant in the final model.

In the typically developing group, the child's current age and parental rating of importance of ability to ride a bicycle were associated with ability to ride independently in the community on univariate analysis (Table III). Both variables remained associated with ability to ride on multivariate analysis (Table III).

DISCUSSION

This is the first published study to examine the proportion of independently ambulant children with CP able to ride a two-wheel bicycle and age at skill acquisition. We found that

while the proportion of children able to ride increased with age across both CP and typically developing groups, the proportion was lower at any given age for children with CP. Given the motor impairments and activity restrictions associated with CP, these results are not unexpected. The findings highlight the challenges children with CP face when learning complex but functional motor skills such as riding a bicycle, by documenting the size of the gap between children functioning at the highest levels of motor function and children with typical development. The results also highlight that independently ambulant children with CP, in particular those classified as GMFCS level I, without additional comorbidities, can learn to ride a bicycle independently, given the right conditions. At the same time the study raises questions around why many of these children are currently not able to ride a bicycle.

The reasons for delayed skill acquisition in children with CP who can ride warrant further attention. For example, parental concern around the safety of their child with motor problems riding a two-wheeled bicycle may exist as a legitimate barrier to achieving independent riding in the community. The complexity of factors that contribute to motor skill acquisition in this population means it cannot be assumed that delay with other gross motor activities, such as walking,[17] is directly related to a later age at learning to ride. In addition to a later age at skill acquisition, Figure 1 shows a plateau in the proportion of children with CP able to ride after 10 years of age. This may indicate that children and their families are less inclined to pursue learning to ride a bicycle after the age of 10 years; a hypothesis consistent with research that indicates children with CP become less active as they transition into their teenage years.[1]

While this study does not attempt to disentangle the reasons why a lower proportion of children with CP are able to ride a bicycle, nor for the delay in skill acquisition, it does consider some factors associated with ability to ride. It was expected that as age increased, the proportion able to ride would also increase in both groups, given that older children had more time to acquire the skill. The result that children with CP of greater functional mobility (GMFCS level I) have increased odds of being able to ride is not surprising given the differences in locomotor ability seen between GMFCS levels. Consideration of the child's functional mobility level may help parents of children with CP and clinicians working with these families set realistic goals around bicycle riding. However, the variables that emerged as associated with ability to ride suggest the factors contributing to riding ability go beyond physical impairment. The findings that the importance placed on bicycle riding by parents and prior experience on a balance bicycle were associated with ability to ride in children classified as GMFCS level I indicates the potential influence of parental attitudes and

opportunities for task-related practice on motor outcomes. It appears many of the children with CP who had learnt to ride did so in informal settings with their families, in particular if their parents placed high importance on learning to ride. These results point to the influence the child's environment has on skill learning; however, further information regarding the barriers and facilitators to riding a bicycle perceived by families at both the skill-learning and participation levels would be beneficial to understand this relationship better.

Interestingly, bicycle-specific therapy was not found to be associated with ability to ride. However, a relatively small sample of children had experienced this therapy (*n*=27) and the specifics of the therapy were varied and poorly defined. Little is known about what current practices involve for clinicians or teachers when training bicycle skills in this population. What impact the paucity of evidence and lack of direction available to professionals and families has on the bicycle-riding ability in this population is unknown. Bicycle riding is a popular activity,[11] yet there are no peer-reviewed published studies focusing on learning to ride a bicycle in children with CP. Moreover, although research supports activity-focused, task-specific interventions directed by goals that are meaningful to the child and their family,[18–20] the evidence is minimal when it comes to approaches for learning specific complex gross motor skills in this population. The majority of task-specific training research conducted is related to improving hand function or self-care.[21] Direction is lacking in terms of how to approach bicycle riding skills, in particular regarding effectiveness of motor learning strategies including dosage, delivery, and responsiveness. This study highlights the need for further research into motor learning approaches for bicycle riding for the majority of children with CP (GMFCS levels I and II) who are not yet able to ride.

This is also the first known study to examine the ability of Australian children with typical development to ride a bicycle and their age at skill acquisition. The majority of children with typical development in this study were reported as able to ride independently in the community, indicating that learning to ride a bicycle is common in Australian children. The age at which 50% of children with typical development in this study could ride a bicycle 20m with help starting (5y) was later than the age at which 50% of Dutch children could ride a bicycle 50m with help getting on and off (4y 6mo).[9] Although differences in the outcome measured limit comparison of these results, both outcomes reflect the learning stage of bicycle riding. The later age found for children with typical development in this study may demonstrate differences in the cultural presence of bicycle riding in the Netherlands compared with Australia.[9]

Limitations

Attempts to reduce confounding between the groups was predominantly made by employing snowball recruitment of the control group via the Victorian Cerebral Palsy Register as the primary form of recruitment. However, there were limitations to this study design as differences in some characteristics between the groups were found, including age, socioeconomic status, and geographical remoteness. Other between-group differences were expected and likely due to the cognitive and physical impairments associated with CP,[13] given children with typical development were excluded if they had a disability.

While a number of potential confounding factors were accounted for through eligibility criteria and data analyses, the case–control design made it challenging to balance identification of all potential confounders with realistic recruitment and data collection. This meant some potential confounders may be left unaccounted for. Additionally, variables potentially associated with ability to ride in the CP group, such as upper limb function and motor type or distribution, were not collected. Potential for measurement bias exists in the parent-reported nature of the study. Parent-reported ability to ride has not been validated in either CP or typically developing populations, nor has the definition of levels of bicycle-riding ability developed by the authors. However, the survey method proved effective for recruiting sufficient sample size.

Caution should be exercised when generalizing some results to the CP population. Previous experience on a balance bicycle was found to be associated with ability to ride, while intellectual impairment and previous bicycle-specific therapy were not. The small sample of parents who reported their child had these specific variables limit the interpretation of these findings. Selection bias can also not be ruled out for either cases or controls. As the survey was delivered online, this may have excluded some participants without access to the Internet, or those who are illiterate. The authors suspect the small amount of missing data for questions relating to parent characteristics (bottom of Table SI) is a result of these questions being at the end of the survey.

CONCLUSION

A proportion of independently ambulant children with CP do learn the complex motor skill of riding a two-wheel bicycle, but a substantially smaller proportion is able to ride at any given age compared with their typically developing peers. If they do learn to ride, they do so at a later age. These findings support the need for further research into motor-skill learning approaches for ambulant children with CP, while working closely with families to empower

children and their parents, and understand their perspectives on what influences learning to ride a bicycle. Variables associated with ability to ride deserve consideration in shaping any future research or clinical efforts in this area.

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SUPPORTING INFORMATION

The following additional material may be found online:

Table SI: Child, parent, and family characteristics in cerebral palsy group, typically developing group, and total cohort.

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Table I: Descriptors of level of ability to ride

	Level of ability to ride	
Least challenging ⇩ Most challenging	I	Able to ride a bicycle (no training wheels) 20m independently but may need help to start
	II	Able to ride a bicycle 20m independently, no help needed to start
	III	Able to ride a bicycle 50m independently, no help needed to start
	IV	Able to ride a bicycle 50m independently and turn corners if required
	V	Able to ride a bicycle 50m independently, turn corners, and go up and down hills
	VI	Able to ride a bicycle independently in community, including on bicycle paths

Table II: Comparison of Kaplan–Meier estimates of cumulative proportion able to ride by selected ages

Age (years)	Cumulative proportions able to ride 20m independently but may need help to start						*p*
	Cerebral palsy group			Typically developing group			
	% estimate	SE	95% CI	% estimate	SE	95% CI	
4	1.8	0.01	0.4–6.8	12.9	0.04	7.4–22.1	0.002
6	14.3	0.03	9.0–22.3	76.5	0.05	67.0–84.8	<0.001
8	27.0	0.04	19.5–36.5	93.4	0.03	85.7–97.7	<0.001
10	45.1	0.05	35.1–56.4	95.6	0.02	88.0–99.0	<0.001
15	57.3	0.07	44.7–70.6	100	–	–	

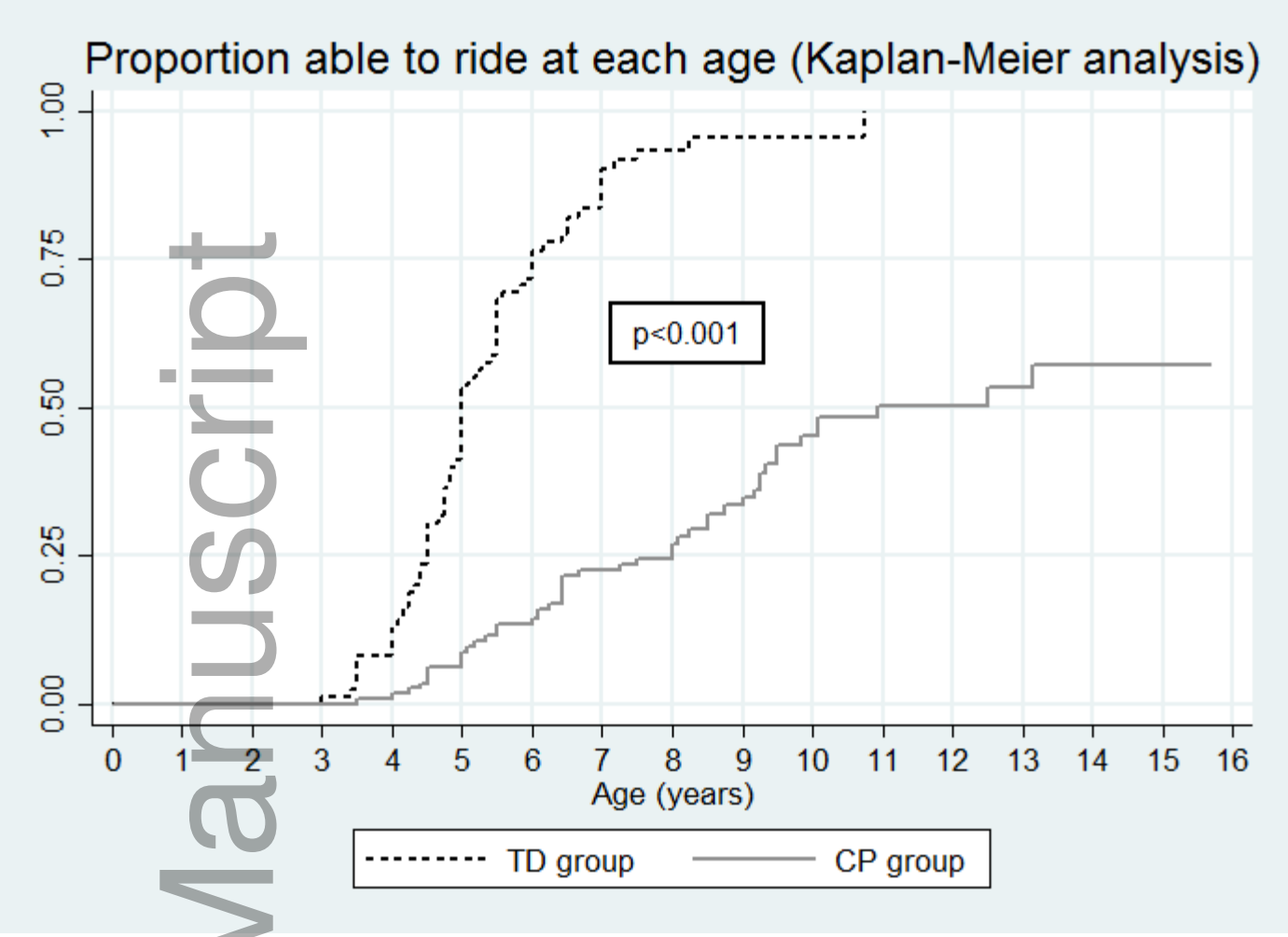
SE, standard error; CI, confidence interval.

Table III: Statistical analysis of variables associated with ability to ride independently in the community

Variable associated with ability to ride	**Observations (*n*)**	**OR**	**95% CI**	**SE**	***p***
Cerebral palsy group					
Univariate (crude) analysis					
GMFCS	113	0.03	0.0–0.2	0.03	0.001
Parent rating of importance	103	2.20	1.3–3.7	0.60	0.002
Age at time of study	113	1.30	1.1–1.5	0.10	0.004
Prior experience on balance bike	103	3.90	1.2–12.8	2.40	0.020
Multivariate logistic regression (GMFCS level I only)					
Parent rating of importance	*n*=71; pseudo r^2=0.2	7.20	1.9–27.3	4.90	0.004
Age at time of study		1.30	1.1–1.7	0.20	0.008
Prior experience on balance bike		4.90	0.1–24.5	4.00	0.060
Typically developing group					
Univariate analysis					
Parent rating of importance	82	2.60	1.2–5.9	1.10	0.020
Age at time of study	87	1.80	1.1–3.0	0.50	0.030
Multivariate logistic regression					
Parent rating of importance	*n*=82; Pseudo r^2=0.3	16.10	2.6–98.0	14.80	0.003
Age at time of study		2.00	1.0–4.0	0.70	0.040

OR, odds ratio; CI, confidence interval; SE, standard error; GMFCS, Gross Motor Functional Classification System.

Figure 1: Kaplan–Meier curve of estimates of the proportion able to ride at any given age by group. These results relate to the child being able to ride at level I (Table SI); however, similar proportions and between-group differences were seen for ability to ride at the other levels of riding ability described in Table SI, including 'Able to ride a two-wheel bicycle independently in community including on bicycle paths'. TD, typically developing; CP, cerebral palsy.



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