Changes in walking ability, intellectual disability, and epilepsy in adults with cerebral palsy over 50 years: a population-based follow-up study

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AIM To determine if walking ability and presence of intellectual disability and epilepsy change from childhood to 50 years of age in individuals with cerebral palsy (CP), and if such changes are related to age, sex, or CP subtype.

METHOD This was a population-based follow-up study of 142 adults born from 1959 to 1978 (82 males, 60 females; mean age 48y 4mo, range 37–58y; 44% unilateral, 35% bilateral, 17% dyskinetic, and 4% ataxic CP) listed in the CP register of western Sweden. We compared childhood data with a follow-up assessment in 2016.

RESULTS At follow-up, walking ability had changed significantly ($p<0.001$). The proportion of participants walking without aids had decreased from 71% to 62%, and wheelchair ambulation increased from 18% to 25%. Walking ability was related to subtype ($p=0.001$), but not to age, sex, pain, fatigue, or body mass index. The proportion classified as having intellectual disability had increased from 16% to 22% ($p=0.039$) and the proportion with epilepsy from 9% to 18% ($p=0.015$). Of those with childhood epilepsy, 46% were seizure-free without medication.

INTERPRETATION Walking ability and the presence of intellectual disability and epilepsy had changed significantly since childhood. Life-long access to specialized health care is warranted for re-evaluation of impairments, treatment, and assistance.

Cerebral palsy (CP) is a lifelong motor disorder, '...often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems'.[1] It is commonly classified into spastic, dyskinetic, or ataxic subtypes, depending on the dominant neurological signs.[2] The severity of impairment and capability of physical and social independence vary considerably between individuals, and the risk of secondary health problems and mortality vary accordingly.[3,4]

Although children with severe impairments have an increased risk of early mortality, almost all children with CP reach adulthood, and today approximately 75% of individuals with CP are adults.[5] Nevertheless, CP is still in many ways considered a childhood condition and specialized health care for individuals with CP is rarely available for adults.[6]

Many parents of children with CP request prognostic information about functioning and participation in adulthood.[7] Because of the considerable individual variations in CP, such a prognosis requires tailoring to the impairment severity profile of the particular child, and the natural history of each impairment has to be known.

Information on the prevalence of impairments in children with CP is available through the many population-based CP registers around the world,[7] and the longitudinal development of specific impairments have been studied in children and young adults.[8,9] However, most CP registers have not been active long enough to include older adults. Systematic reviews of impairments in adults with CP rely on fewer and smaller studies with large variations[5,10] and little is known about the longitudinal development of specific impairments in adulthood.

Walking ability, intellectual ability, and epilepsy are important aspects of the impairment profile of an individual with CP, as they are associated with both participation and survival.[3,11,12] A decline in walking ability has been reported in 33% to 82% of adults with CP in cross-sectional studies[5] and this decline has been associated with CP subtype, pain, fatigue, and reduced balance.[13] Studies following up walking ability from childhood rarely extend

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further than the age of 30 years and follow-up studies of walking ability in adults are rare.

Intellectual disability has been reported in 34% to 64% of children with CP[7,14] and in 21% to 54% of adults with CP.[15,16] We have found no study describing how well assessments of children with CP predict their intellectual capability in adulthood. Epilepsy has been reported in 17% to 29% of children with CP[7] and in 22% to 42% of adults with CP.[15,16] With the exception of one study of 49 adults showing no change in the prevalence of epilepsy over 14 years,[17] the longitudinal development of epilepsy in adults with CP seems not to have been studied.

The CP register of western Sweden was started in the 1950s and is one of the longest-running population-based CP registers worldwide.[18] This register provides a unique opportunity to conduct a population-based study of very long-term outcomes.

The first aim of this study was to determine if walking ability, presence of intellectual disability, and epilepsy change from childhood to adulthood in individuals with CP. A second aim was to examine whether any such changes are related to age, sex, or CP subtype, and if walking ability in adulthood is related to pain, fatigue, or body mass index (BMI).

What this paper adds

- Changes in impairments in individuals with cerebral palsy (CP) over time are related to CP subtype.
- After 50 years, walking ability in CP may have deteriorated or improved.
- Intellectual disability in CP may not always be detected in early childhood assessments.
- Epilepsy in CP may develop after childhood or may be outgrown.

METHOD

The present study was a long-term, follow-up study, comparing childhood CP data with a follow-up assessment conducted in adults aged 37 to 58 years. The study was part of a more extensive project surveying health and participation in adults with CP. All participants or their legal guardians gave informed consent. The study was approved by the Regional Ethical Review board in Gothenburg.

Participants

The focus of the current study was the five oldest cohorts in the CP register of western Sweden, adults born from 1959 to 1978 (n=723). The register is population-based and continuously registers all children born in Sweden with CP residing in western Sweden (Västra Götaland and Halland counties) at age 4 to 8 years.[18] Data are entered in the register at one time point for each child, and no follow-up data are added thereafter. Data from the register are published regularly, describing changes in prevalence and causes of CP.[18]

In 2016, all participants in the CP register born between 1959 and 1978 and still residing in the county of Västra Götaland, were identified through the Swedish Population Register and invited to a follow-up assessment consisting of an interview, a clinical examination, and questionnaires. Invitations were sent out by mail, followed by a telephone call and a second letter. The follow-up assessments were conducted from 2016 to 2019.

Data on living arrangements, education and employment, and medical issues such as seizures, pain, and medication, were gathered through interview. Walking ability was assessed, height and weight were measured, and BMI calculated. The Fatigue Severity Scale,[19] a questionnaire with nine items on a 7-point Likert scale, with higher scores indicating more severe fatigue, was administered. The childhood data from the CP register, describing walking ability and the presence of intellectual disability and epilepsy were compared to the data from the follow-up assessment in adulthood and analysed for associations with CP subtype, age, or sex. Walking ability in adulthood was also analysed for associations with pain, fatigue, or BMI.

Definitions

CP subtype was classified according to the Surveillance of Cerebral Palsy in Europe as unilateral spastic, bilateral spastic, dyskinetic, or ataxic.[2]

The classification of walking ability was based on self-reported, everyday performance. Walking impairment was originally defined in the register as mild (walking without aids), moderate (walking with aids), or severe (wheelchair ambulation). Translated to the Gross Motor Function Classification System (GMFCS), mild is GMFCS levels I or II, moderate is level III, and severe is levels IV or V.[20]

Intellectual disability was defined in the register as having an estimated or measured IQ <70. At follow-up, adults were classified as having intellectual disability if they claimed to have an intellectual disability, had attended a special school, resided at a group home, or were in sheltered employment designed for people with intellectual disability. In Sweden, all individuals with an IQ <70 are offered placements in special schools, sheltered employment, and group homes, designed for individuals with intellectual disability. All these services are free of charge and very few who are entitled to them opt out completely. We, therefore, consider the use of any of these services a good proxy of IQ <70.

Epilepsy was defined both in the register and at follow-up, as having antiepileptic treatment or recurring epileptic seizures. Presence of pain was defined as experiencing any pain, either constant or recurring, mild or severe.

Statistical analysis

Descriptive statistics summarized patients' characteristics and non-parametric methods were used for comparisons between groups. The participants were compared to those lost to follow-up with the χ^2 test for independence. Changes over time were analysed with McNemar's test for nominal data (intellectual disability and epilepsy) and the Sign test for ordinal data (walking ability). When analysing associations between impairments and age, the participants were divided into five age-groups according to the birth cohorts in the register. However, the mean age of

participants was calculated using the age of each participant at their follow-up. The χ^2 goodness-of-fit test was used to analyse the proportion of participants with declining walking ability and the proportion of new cases of intellectual disability or epilepsy, related to CP subtype. The distribution of walking ability related to age, sex, CP subtype, and pain were analysed with the χ^2 test for independence, or Fisher's exact test when observations were few. The distribution of walking ability in relation to fatigue and BMI was analysed with the Kruskal–Wallis test. The significance level was set at a two-tailed p-value of <0.05. Statistical analyses were conducted using IBM SPSS statistics 25 software (IBM Corp., Armonk, NY, USA).

RESULTS

Of the 417 adults with CP invited to the study, 142 (34%) chose to participate (Fig. S1, online supporting information). The mean age of participants at follow-up was 48 years 4 months (range 37–58y). Childhood registration had occurred at between 4 and 8 years of age. CP subtype assessed in childhood was spastic unilateral (44%), spastic bilateral (35%), dyskinetic (17%), and ataxic (4%). There was no significant difference between the participants and those lost to follow-up regarding sex, age, or walking impairment. However, there was a difference in CP subtype, intellectual disability, and epilepsy (Table 1).

At follow-up in adulthood, walking ability had changed significantly (p=0.001). The proportion of participants walking without aids had decreased from 71% to 62%, those walking with aids increased from 11% to 13%, and those using wheelchair ambulation had increased from 18% to 25% (Fig. 1). On an individual level, both decline and improvement was noted. In 22 (19%) of the 117 participants who walked with or without aids in childhood, walking ability had declined (Fig. 2). The proportion of participants showing walking decline was related to CP subtype (p=0.013), ranging from 45% in dyskinetic CP to 8% in unilateral CP. Simultaneously, four (10%) of the 40 participants who walked with aids or used wheelchair ambulation in childhood, showed improved walking ability (Fig. 2).

Walking ability in adulthood was associated with CP subtype (p<0.001), but not with age (p=0.602), sex (p=0.465), pain (p=0.820), fatigue (p=0.274), or BMI (p=0.143; Table S1, online supporting information).

In adulthood, the proportion of participants classified as having intellectual disability had increased from 16% to 22% (p=0.039; Fig. 1). Ten more participants were classified as having intellectual disability and two no longer met the criteria for intellectual disability. The proportion of new cases differed between CP subtypes (p=0.042). The presence of intellectual disability in adulthood was related to CP subtype (p<0.001), being most common in dyskinetic and ataxic CP, but not related to age (p=0.265) or sex (p=0.109).

The presence of epilepsy had increased from 9% to 18% (p=0.015; Fig. 1). Nineteen participants had developed epilepsy while six (46%) of the 13 participants with epilepsy in childhood no longer fulfilled the criteria. The proportion of new cases differed between CP subtypes (p=0.039), with a majority of new cases of epilepsy occurring in unilateral CP. The presence of epilepsy in adulthood was related to CP subtype (p=0.024), but not to age (p=0.746) or sex (p=0.665).

DISCUSSION

The present study was a follow-up assessment of individuals who were entered in a population-based CP register as children, 38 to 58 years ago. The main findings were that the decrease in walking ability was less than expected and that the presence of intellectual disability and epilepsy had increased. Further, some individuals showed improved walking ability or complete remission of epilepsy.

Table 1: Characteristics of participants and those lost to follow-up

	Adulthood, *n* (%)	Childhood vs adulthood, *p*	Childhood, *n* (%)	Childhood vs childhood lost to follow-up, *p*	Childhood lost to follow-up, *n* (%)
Total	142 (100)		142 (100)		275 (100)
Sex					
Female	60 (42)		60 (42)	0.898[a]	118 (43)
Male	82 (58)		82 (58)		157 (57)
CP subtype					
Spastic unilateral	62 (44)		62 (44)	0.001[a]	101 (37)
Spastic bilateral	50 (35)		50 (35)		132 (48)
Dyskinetic	24 (17)		24 (17)		19 (7)
Ataxic	6 (4)		6 (4)		23 (8)
Walking impairment					
Mild (GMFCS level I–II)	88 (62)	0.001[c]	102 (71)	0.126[b]	186 (68)
Moderate (GMFCS level III)	18 (13)		15 (11)		35 (13)
Severe (GMFCS level IV–V)	36 (25)		25 (18)		45 (16)
Unknown	0		0		9 (3)
Intellectual disability	31 (22)	0.039[d]	23 (16)	0.004[a]	80 (29)
Epilepsy	26 (18)	0.015[d]	13 (9)	0.006[a]	54 (19)

Significant difference, p<0.05, two-tailed. [a] Pearson's χ^2 test. [b] Fisher's exact test. [c] Sign test. [d] McNemar's Test. CP, cerebral palsy; GMFCS, Gross Motor Function Classification System.

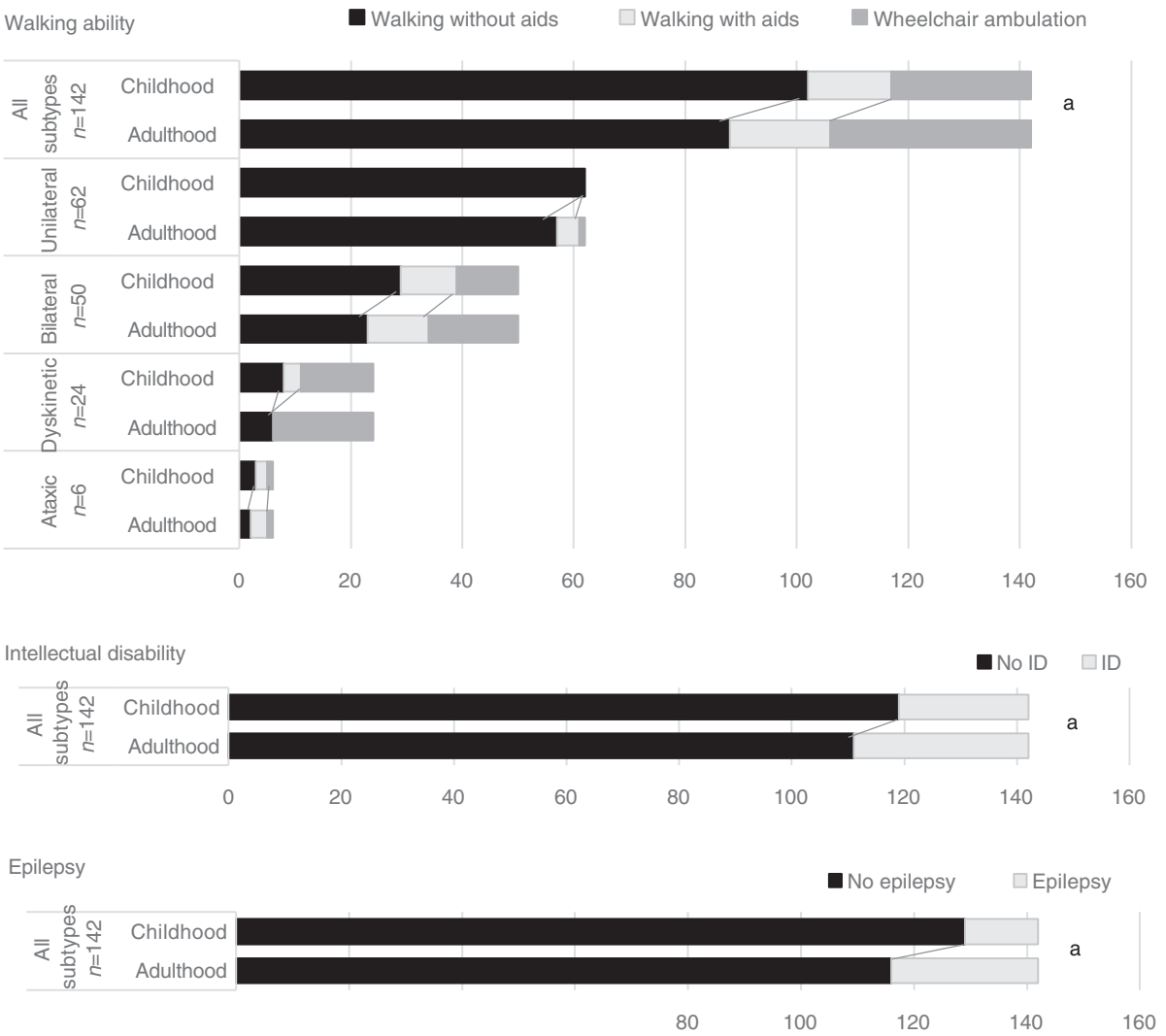

Figure 1: Changes in impairment from childhood to adulthood. [a]Significant change ($p<0.05$). ID, intellectual disability.

At follow-up, walking ability had decreased for 19% of participants. This decrease in walking ability was counteracted by a healthy survivor effect, where the prevalence of a disease decreases over time because those who have the disease die. An earlier study of the same age groups in the CP register showed worse survival among participants who used wheelchair ambulation in childhood.[14] As a result, even though walking ability decreased for 19% of the participants, the distribution of walking impairment in surviving adults is very similar to the original distribution in the whole cohort of children with CP born 1959 to 1978.[14]

Previous studies of adults with CP reported a decrease in walking ability in 33% to 82% of similar aged individuals.[5] To a large extent, this difference could be due to our study being population-based instead of hospital- or service-based and, therefore, including adults who had not had reason to access healthcare. Another important explanation could be that there is a difference between a subjective decrease in walking ability and a shift in GMFCS level. In our study, walking ability was assessed with the GMFCS. Earlier studies have often been cross-sectional and reported on a subjective decrease in walking ability. Such a subjective decrease in walking ability might very well be troublesome for the individual, but may not affect their GMFCS level. In addition, adults choose their method of ambulation more than children. There are adults whose parents pushed them to walk, but as soon as they were old enough to decide for themselves, they chose wheelchair ambulation, thus shifting from GMFCS level III to IV without deteriorating in gross motor function. Moreover, there are adults who do not want to use walking aids, avoid taking more than a few steps indoors because walking more would cause pain or frequent falls, but who manage independent community ambulation by driving a car. These individuals stay in GMFCS level II even though they would have been classified in GMFCS level III or IV if they chose to use walking aids or a wheelchair. Therefore for these individuals, their GMFCS level does not

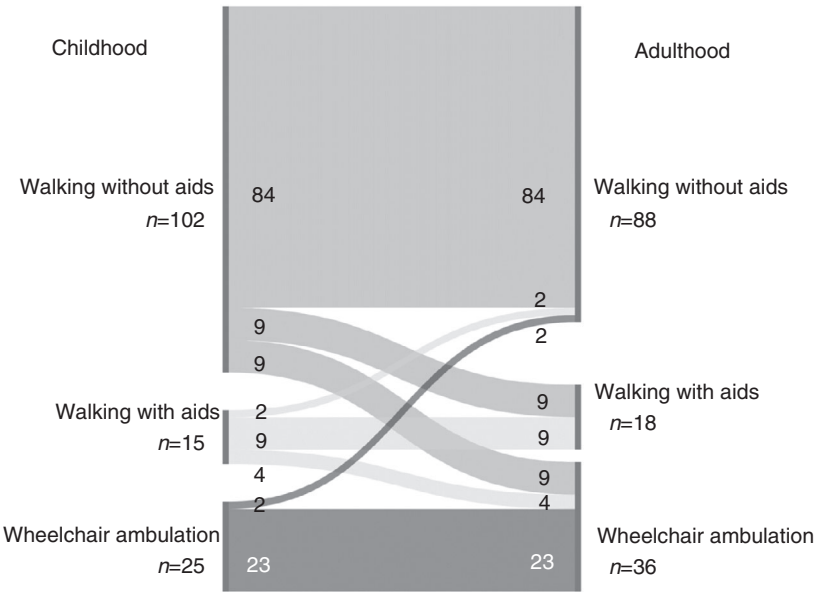

Figure 2: Individual changes in walking ability from childhood to adulthood.

necessarily give an accurate reflection of their walking ability.

Decreased walking ability has been associated with age, pain, and fatigue,[13,21] and recent research has discussed associated risk factors such as a sedentary lifestyle and obesity.[22] However, in our study sample, older age, the presence of pain, a higher level of fatigue, or obesity were not more common among participants with more severe walking impairments. This part of our study, however, was cross-sectional. There is still a possibility that pain or fatigue could cause participants to start using walking aids or wheelchair ambulation. If the use of assistive devices then alleviated the pain or fatigue, no difference between groups would have been detected.

It is important to mention that we also found individuals with improved walking ability, which is in line with previous studies.[13,21] However, they were too few and too disparate to allow for an analysis of the causes of their improvement.

At follow-up, there were 10 new cases of intellectual disability. There are several possible explanations for these new cases. First, the intellectual development of a young child with a normal IQ for their age might not always continue as expected. Second, a mild intellectual disability might not always become apparent and, therefore, not be assessed or diagnosed until older school age or adulthood. There are still adults with CP who have never been tested for intellectual disability. Among those who did not finish school and never attained an employment or independent living, there may be some who, if tested, would be diagnosed with intellectual disability. Third, access to better communication devices might have contributed to a more correct evaluation later in life. Consequently, testing only in early childhood might underestimate the presence of intellectual disability in adults. Offering renewed testing at older school age and in adulthood, as is recommended in emerging practice guidelines,[23] could help ensure that these individuals receive the support they need.

According to our literature review, this study was the first follow-up of epilepsy in individuals with CP, spanning both childhood and adulthood. At follow-up, the number of participants with epilepsy had doubled and the majority of new cases were among individuals with unilateral CP. Some of these new cases may have occurred in childhood, after inclusion in the register, since onset of epilepsy has been shown to occur a little later in children with unilateral compared to bilateral CP.[9] But more importantly, at follow-up almost half of the participants with epilepsy in childhood had discontinued their medication and were seizure-free. This is in accordance with a previous study of children with epilepsy and CP,[24] and indicates that at least some of the adults with CP who are seizure-free on medication could benefit from an attempt to taper their medication.

CP subtype has been shown to be associated with survival.[3] According to our results, CP subtype is also associated with the development of impairments in those who survive into their 40s and 50s, making CP subtype a factor to consider in the prognosis for a child with CP.

Limitations

As was often the case before the introduction of the GMFCS,[21] our CP register classified walking ability into three categories, placing GMFCS levels I and II in the same category. A shift from GMFCS level I to level II would, therefore, go undetected. On the other hand, shifts

between GMFCS levels I and II have been reported to go both ways, with as many as 43% of adults in GMFCS level II shifting to GMFCS level I.[13] We, therefore, believe that the three levels used in the register provide a good classification of walking ability.

Much effort and adaptations were made to facilitate participation of adults with intellectual disability in the study, but adults with intellectual disability, and as a consequence also adults with epilepsy, were underrepresented in our sample compared to those lost to follow-up. Still, the presence of intellectual disability and epilepsy in adulthood was similar to that of an earlier study of the same age group.[17] In other respects such as age, sex, CP subtype, and mobility, the participants were representative of the population invited to follow-up.

The participants in our study were seen at two time points. In order to study longitudinal changes in more detail, repeated assessments would have been desirable. In the future, with increasing coverage of adults, the Swedish national quality register (CPUP)[23] might be able to provide such data.

CONCLUSION

Knowledge about the natural history of different impairments in CP throughout the lifespan is vital both to individuals with CP and to health care services, in order to be able to prioritize interventions today and prevent complications in the future. This study was conducted in Sweden and the results may be relevant for other high-income countries with similar health care systems.

Our results indicate that walking ability, the presence of epilepsy, and whether an individual is classified as having intellectual disability or not can change over time, confirming that CP is not a static condition. Further studies are needed to examine how other aspects of CP, such as associated health conditions and the possibility for participation and autonomy, change throughout the lifespan. Lifelong access to specialized health care for re-evaluation of functioning, treatments, assistance, and assistive devices is warranted.

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DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

SUPPORTING INFORMATION

The following additional material may be found online:

Figure S1: Study design flow-chart.

Table S1: Walking ability related to sex, age, subtype, BMI, fatigue, and pain

REFERENCES

1. Rosenbaum P, Paneth N, Leviton A, Goldstein M, Bax M, Damiano D, Dan B, Jacobsson B. A report: the definition and classification of cerebral palsy April 2006. *Dev Med Child Neurol Suppl* 2007; **109**: 8–14.
2. Surveillance of Cerebral Palsy in E. Surveillance of cerebral palsy in Europe: a collaboration of cerebral palsy surveys and registers. Surveillance of Cerebral Palsy in Europe (SCPE). *Dev Med Child Neurol* 2000; **42**: 816–24.
3. Himmelmann K, Sundh V. Survival with cerebral palsy over five decades in western Sweden. *Dev Med Child Neurol* 2015; **57**: 762–7.
4. Blair E, Langdon K, McIntyre S, Lawrence D, Watson L. Survival and mortality in cerebral palsy: observations to the sixth decade from a data linkage study of a total population register and National Death Index. *BMC Neurol* 2019; **19**: 111.
5. van Gorp M, Hilberink SR, Noten S, et al. Epidemiology of cerebral palsy in adulthood: a systematic review and meta-analysis of the most frequently studied outcomes. *Arch Phys Med Rehabil* 2020; **101**: 1041–52.
6. Young NL. The transition to adulthood for children with cerebral palsy: what do we know about their health care needs? *J Pediatr Orthop* 2007; **27**: 476–9.
7. Novak I, Hines M, Goldsmith S, Barclay R. Clinical prognostic messages from a systematic review on cerebral palsy. *Pediatrics* 2012; **130**: e1285–312.
8. McCormick A, Brien M, Plourde J, Wood E, Rosenbaum P, McLean J. Stability of the Gross Motor Function Classification System in adults with cerebral palsy. *Dev Med Child Neurol* 2007; **49**: 265–9.
9. Carlsson M, Hagberg G, Olsson I. Clinical and aetiological aspects of epilepsy in children with cerebral palsy. *Dev Med Child Neurol* 2003; **45**: 371–6.
10. Morgan P, McGinley J. Gait function and decline in adults with cerebral palsy: a systematic review. *Disabil Rehabil* 2014; **36**: 1–9.
11. Donkervoort M, Roebroeck M, Wiegerink D, van der Heijden-Maessen H, Stam H, Transition Research Group South West N. Determinants of functioning of adolescents and young adults with cerebral palsy. *Disabil Rehabil* 2007; **29**: 453–63.
12. Tan SS, van der Slot WM, Ketelaar M, et al. Factors contributing to the longitudinal development of social participation in individuals with cerebral palsy. *Res Dev Disabil* 2016; **57**: 125–35.
13. Opheim A, Jahnsen R, Olsson E, Stanghelle JK. Walking function, pain, and fatigue in adults with cerebral palsy: a 7-year follow-up study. *Dev Med Child Neurol* 2009; **51**: 381–8.
14. Jonsson U, Eek MN, Sunnerhagen KS, Himmelmann K. Cerebral palsy prevalence, subtypes, and associated impairments: a population-based comparison study of adults and children. *Dev Med Child Neurol* 2019; **61**: 1162–7.
15. Smith KJ, Peterson MD, O'Connell NE, et al. Risk of depression and anxiety in adults with cerebral palsy. *JAMA Neurol* 2019; **76**: 294–300.
16. Fortuna RJ, Holub A, Turk MA, Meccarello J, Davidson PW. Health conditions, functional status and health care utilization in adults with cerebral palsy. *Fam Pract* 2018; **35**: 661–70.
17. Benner JL, Hilberink SR, Veenis T, Stam HJ, van der Slot WM, Roebroeck ME. Long-term deterioration of

perceived health and functioning in adults with cerebral palsy. *Arch Phys Med Rehabil* 2017; **98**: 2196–205.

18. Hagberg B, Hagberg G, Olow I. The changing panorama of cerebral palsy in Sweden 1954–1970. *Acta Paediatr* 1975; **64**: 187–92.
19. Krupp LB, LaRocca NG, Muir-Nash J, Steinberg AD. The fatigue severity scale. Application to patients with multiple sclerosis and systemic lupus erythematosus. *Arch Neurol* 1989; **46**: 1121–3.
20. Palisano RJ, Rosenbaum P, Bartlett D, Livingston MH. Content validity of the expanded and revised Gross Motor Function Classification System. *Dev Med Child Neurol* 2008; **50**: 744–50.
21. Jahnsen R, Villien L, Egeland T, Stanghelle JK, Holm I. Locomotion skills in adults with cerebral palsy. *Clin Rehabil* 2004; **18**: 309–16.
22. Peterson MD, Gordon PM, Hurvitz EA. Chronic disease risk among adults with cerebral palsy: the role of premature sarcopoenia, obesity and sedentary behaviour. *Obes Rev* 2013; **14**: 171–82.
23. Alriksson-Schmidt AI, Arner M, Westbom L, et al. A combined surveillance program and quality register improves management of childhood disability. *Disabil Rehabil* 2017; **39**: 830–6.
24. Zafeiriou DI, Kontopoulos EE, Tsikoulas I. Characteristics and prognosis of epilepsy in children with cerebral palsy. *J Child Neurol* 1999; **14**: 289–94.