Received: 8 September 2022 | Accepted: 12 June 2023

DOI: 10.1111/dmcn.15710

ORIGINAL ARTICLE

Classification of events contributing to postneonatal cerebral palsy: Development, reliability, and recommendations for use

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Abstract

Aim: This paper introduces the Surveillance of Cerebral Palsy in Europe (SCPE) classification of events contributing to postneonatally acquired cerebral palsy, presents its interrater reliability, and describes the cases identified in the SCPE database.

Method: The development of the classification, based on literature review and expert discussions, resulted in six main categories and 19 subcategories. The first chronological event designated as the primary event was mainly reported. Interrater reliability was assessed through online exercise providing 24 clinical vignettes representing single/complex pathways. Percent agreement and Gwet's AC1 index of reliability were estimated. Primary events were described using data of 221 children born between 2008 and 2012.

Results: Thirty-nine professionals (21 registries) participated in the reliability exercise. Substantial overall agreement was reached (0.75), with some contrast between complex (0.48, moderate agreement) and single events involved (0.89, almost perfect). The distribution of primary events showed that 32.1% were infections (category A), 23.1% head injuries (B), 15.4% related to surgery or medical interventions (C), 13.1% cerebrovascular accidents (D), 9.1% hypoxic brain damaging events of other origins (E), and 7.2% miscellaneous (F).

Interpretation: This classification allows all the events involved to be recorded while consistently reporting the primary event, and may be used in different settings.

Postneonatal cerebral palsy (CP) is due to non-progressive injury of the developing brain occurring beyond the neonatal period (i.e. more than 28 days after birth), sometimes stated as unrelated to factors in the ante-, peri-, or neonatal period.[1–3] To clarify, the Surveillance of Cerebral Palsy in Europe (SCPE) guidelines specify that postneonatal CP

This original article is commented on by Waight on pages 146–147 of this issue.

Abbreviations: CVA, cerebrovascular accident; SCPE, Surveillance of Cerebral Palsy in Europe.

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should be considered only in the absence of a causative brain-damaging event before 28 days after birth. Infants with contextual information, from during their first 28 days, of events very likely to have led to CP (such as a stormy course in the neonatal intensive care unit and/or lesions identified on neonatal imaging) should not be considered to have postneonatal CP, even if there is a new causative brain damaging event after 28 days. In addition, an upper age limit for brain damage of 2 years is set, in line with most of the literature.[1,4–8] It has been reported that 6% to 8% of cases of CP are postneonatally acquired in high-income countries[1,4,8,9] and up to one-third in low- and middle-income countries.[10–15] Birth prevalence for children born from 1995 onwards was estimated at 0.8 per 10 000 livebirths (95% confidence interval 0.6–1.0) in high-income countries in a recent meta-analysis.[16]

In contrast to CP attributed to disturbances in the development of the fetal or newborn infant's brain, the events involved in the etiological pathways following one another to lead to the brain lesion underpinning postneonatal CP are often easily identified. They provide an opportunity for preventive measures to be implemented in some cases. The first known event in this sequence does not (necessarily) directly cause the brain injury, but it is the starting point and, in this sense, it is particularly interesting because if it could have been avoided then the brain injury would not have occurred. This supports the ability to relate rate changes over time to successful preventive strategies. In the literature, infections, head injuries, and (cerebro) vascular episodes have long been identified as the three leading categories of events contributing to brain lesions.[1,2,4,17–19] On the other hand, the content of the subcategories differs considerably.[1,2,4,9] Confusion and lack of standardization of cerebrovascular events, consequences related to medical intervention, and hypoxic incidents in the literature make it difficult to compare and accurately monitor prevalence changes over time. These key categories should be clearly defined and individualized. A new classification should be able to reflect change in broad categories of events involved over long time periods while providing a fine-grained description. In addition, there is currently no international consensus on how the complex pathways leading to CP and underlying the main categories should be classified. An internationally agreed framework is needed that promotes the reporting of the most likely primary event in complex pathways for CP, whilst also capturing the details of other contributing factors. This would allow reproducible, reliable comparisons over time and across settings.

This paper aims to: (1) present the classification of events contributing to the brain lesion underpinning postneonatal CP developed by the SCPE network of CP registries, as well as guidance for designating and coding the primary event to be reported; (2) present its interrater reliability for designating the primary event; and (3) report on the distribution of these primary events using the last five validated birth years compiled in the SCPE database.

What this paper adds

- A standardized classification enables the description of the events contributing to postneonatal cerebral palsy (CP).
- The first chronological event in complex pathway leading to CP is coded.
- Category choice and coding of the primary event identify preventable situations.
- The detailed 2-level classification is easy to use in various settings.
- Substantial overall interrater reliability shows that main categories can be consistently differentiated.

METHOD

Development of a classification of events contributing to the brain lesion underpinning postneonatal CP

The first step in the development process was to determine the main categories and criteria to be used to classify events involved, after a review of the literature and a critical analysis of the data available in the SCPE database concerning children with postneonatal CP. In addition to the 'infection' and 'head injury' categories that are consistently found in the literature, we decided to introduce first a 'brain injury related to surgery or other invasive medical interventions' category (regardless of the mechanism involved, hypoxic or cerebrovascular), then a 'cerebrovascular accident (CVA)' category (not covered by other categories), and lastly, a 'hypoxic brain damaging event of other origin' category that included events with similar underlying pathophysiological mechanism. Other situations not assigned to a specific event-type group were classified in a 'miscellaneous' category.

Beyond these main categories, a detailed description in 19 subcategories was proposed to meet the dual objective of ease of use and precision in description. Two-level reporting was thus preferred: the letters representing the six main categories allow for reporting on trends over time, while the numbers used for the subcategories allow for precise reporting.

The classification was designed to be used in a two-step process in the case of a complex pathway leading to CP: (1) record in a chronological order all the known events potentially involved in the pathway leading to the brain injury and classify them (with the two-level reporting); (2) choose the first event in chronological order to be reported considering it as a point of origin and designating it as the 'primary event'. This 'primary event' should allow results to be reported in a homogeneous and reproducible way and should aim to focus possible preventive measures on one initial event that could be avoided. As an example, in the case of a child with a septic embolism resulting in CVA, (1) both infection and CVA

could be registered and coded, (2) 'infection' should be designated as the 'primary event' to be reported.

The development of the classification was a comprehensive process that combined proposals and decisions from the working group (mainly neuropaediatricians and epidemiologists) and discussions with the entire SCPE network until a consensus on classification was reached. The classification is presented in Table 1. Instructions for the use of the classification (Table S1), clarifying, in particular, the principle of the 'primary event' should always be used alongside the classification.

Interrater reliability of the primary event contributing to the brain lesion coding using the classification

Interrater reliability exercise focused on the primary event contributing to the brain lesion because it was the main event chosen to be reported. Members of the 24 active SCPE registries were invited to participate in an online interrater reliability exercise between March and April 2022. A total of 24 short clinical vignettes were written by clinician members of the working group to cover various situations (nine primary events of category A 'infection', three category B 'head injury', four category C 'brain injury related to surgery or other medical interventions', three category D 'CVA', three category E 'hypoxic brain damaging event of other origin', two category F 'miscellaneous). Participants were asked to code independently at least the main category even if they were unable to identify the subcategory, and to follow the coding instructions provided. The postneonatal CP cases were preliminarily divided into cases 'with a single identified event' (16 of 24) and cases 'with a complex pathway leading to CP where several events follow one another to contribute to the brain injury' (8 of 24). Participants were unaware of these distributions. In addition to the clinical vignettes, questions were asked to document the respondents' profile and experience with CP registration. The survey took approximately 30 minutes to complete. After 2 weeks, a reminder was sent stressing the importance of using the coding

TABLE 1 Classification of primary events contributing to brain lesions underpinning postneonatal cerebral palsy (CP) and their distribution among the 221 children with postneonatal CP with specified event known to be associated with development of brain lesion in the Surveillance of Cerebral Palsy in Europe database for the five last validated birth years (children born between 2008 and 2012).

Category	*n*	%
A Infection	**71**	**32.1**
A1 Encephalitis (all origins, except autoimmune origin) and/or meningitis (bacterial, viral, fungal…)	45	
A2 Severe sepsis, septicaemia, or septic shock	6	
A3 Other infections and consequences (Reye syndrome, gastroenteritis with severe dehydration, respiratory infections, malaria, encephalopathies with [suspected] infectious origin…)	20	
B Head injury	**51**	**23.1**
B1 Road traffic accident	5	
B2 Other accidental injury	5	
B3 Non-accidental injury	25	
B4 Unspecified	16	
C Brain injury related to surgery or other medical intervention	**34**	**15.4**
C1 Cardiac	17	
C2 Brain	11	
C3 Other organs	4	
C4 Unspecified	2	
D Cerebrovascular accident Non-traumatic (including ischaemic, haemorrhagic, or unspecified cerebrovascular accident)	**29**	**13.1**
E Hypoxic brain damaging event of other origin	20	**9.1**
E1 Near-miss sudden infant death syndrome	0	
E2 Near-drowning	9	
E3 Respiratory distress syndrome of non-infectious origin (traumatic or related to foreign body in respiratory tract…)	2	
E4 Cardiac arrest and heart infarction	7	
E5 Hypoxic of other origin (carbon monoxide inhalation, hypoglycaemic coma, electrocution…) or unspecified	2	
F Miscellaneous	**16**	**7.2**
F1 Status epilepticus, convulsions	10	
F2 Other	6	
Total	**221**	**100**

instructions. The coding instructions have been amended after analysing the results of this exercise to improve understanding of what should be designated as the primary event when several events occurred.

Statistical analysis

Interrater reliability was estimated with kappa-like statistics using the kappaetc package[20] in Stata 14.2 (StataCorp, College Station, TX, USA). As recommended,[21] the interrater reliability of the classification was assessed using complementary indexes. Percent agreement was estimated considering the six main categories. For each case, percent agreement was calculated as the proportion of pairs of raters (among all possible pairs of raters formed from the raters' sample) who assigned a given case to the same category. Percent agreement averages these proportions over all cases. Two agreement coefficients adjusted for chance were then computed along with their design-based standard errors and their 95% confidence intervals: the Fleiss' generalized kappa coefficient and the AC1 statistic originally introduced by Gwet.[22] Both coefficients quantify chance-corrected agreement but differ in the way the percent chance agreement is measured. The Gwet's coefficient was developed to overcome the limitations of the kappa coefficient, known as kappa paradoxes.[22] Results for both coefficients were interpreted using the Landis-Koch scale (<0.00 poor, 0.00–0.20 slight, 0.21–0.40 fair, 0.41–0.60 moderate, 0.61–0.80 substantial, 0.81–1.00 almost perfect agreement).[23] In order to search for outlying raters, influence analysis was applied by examining the agreement indexes, removing one rater at a time.

Interrater reliability statistics were estimated among the total sample of participants and for specific subgroups of raters defined according to their status (clinicians vs others), their experience in their registry (less vs 5 years and more), their mission in their registry (covering the inclusion of CP cases, yes vs no), and date of completion (before vs after reminder of the coding procedure). Agreement statistics for the total sample of raters were also estimated with stratification for case difficulty.

Application of the classification

Lastly, the classification was used to describe the events contributing to the brain lesions underpinning postneonatal CP included in the SCPE central registry for the last five validated birth years (children born between 2008 and 2012). Until now, the central registry described postneonatal cases via a clear text format and an International Classification of Diseases, 10th Revision code which the working group used to recode all cases applying the classification. For each case, the primary event was coded by three members of the group independently with discussion in case of disagreement. The proposed coding was sent back to each registry for validation.

Ethics statement

This study was based exclusively on pseudonymized registry data compiled at European level in the SCPE database. It did not require any contact with the registered persons. Therefore, ethical review and approval were not required for this study.

RESULTS

The classification included the following six major categories (Table 1): category A 'infection'; category B 'head injury (accidental or non-accidental)'; category C 'brain injury related to surgery or other invasive medical interventions'; category D 'all non-traumatic events, ischaemic, haemorrhagic or unspecified CVA'; category E 'hypoxic brain damaging events of other origin'; and category F 'miscellaneous'.

A total of 39 individuals from 21 registries and 19 countries provided complete coding (both main and subcategories) for the interrater reliability exercise (Table 2). They were mainly clinicians (79.5%) who had been working in registries for several years (76.9% more than 5 years) and were most often in charge of registry inclusions (64.1%). Slightly more than half completed the survey after the reminder. The distribution of responses is presented in Table 3. In each case, the most frequent response of a category corresponded to the coding expected by the expert group who prepared the clinical vignettes. In 14 of the 24 clinical cases, at least 37 raters coded the expected category. Percent agreement and chance-corrected statistics for interrater agreement are presented in Table 4. An overall substantial level of agreement was reached with percent agreement 0.79 and Gwet's AC1 0.75. Agreement differed according to the type of clinical vignette proposed. Agreement was moderate (Gwet's AC1 0.48) for the eight cases with complex pathways leading to CP (the kappa coefficient was even lower at 0.36), whereas

TABLE 2 Characteristics of participants in the interrater reliability exercise.

	n	%
Main professional activity		
Clinician	31	79.5
Non-clinician	7	17.9
Unknown/not stated	1	2.6
Length of time in the registry		
< 5 years	8	20.5
≥ 5 years	30	76.9
Unknown/not stated	1	2.6
Function in the registry: in charge of inclusion of children with cerebral palsy		
Yes	25	64.1
No	13	33.3
Unknown/not stated	1	2.6

TABLE 3 Results of the interrater reliability exercise for the classification of the primary event contributing to the brain lesion in the main categories.

Case	Short case description	Cat. A	Cat. B	Cat. C	Cat. D	Cat. E	Cat. F
1	Surgical removal of a brain tumour	0	0	39	0	0	0
2[a]	Meningitis complicated by seizures, hydrocephalus, and ventriculoperitoneal shunt	38	0	0	0	0	1
3	Meningitis and sepsis	39	0	0	0	0	0
4[a]	Brain surgery due to CVA (arteriovenous malformation)	0	0	8	31	0	0
5[a]	Sepsis complicated by CVA	30	0	0	6	1	2
6[a]	Sepsis after cardiac surgery	13	0	24	0	1	1
7	Electrocution, cardiac arrest	0	1	0	0	38	0
8[a]	Status epilepticus due to CVA	0	0	0	24	0	15
9	Encephalopathy with suspected infectious origin	38	0	0	0	0	1
10	Septic shock	39	0	0	0	0	0
11	Significant smoke inhalation requiring resuscitation, CVA	0	1	0	1	37	0
12	Status epilepticus	0	0	0	0	1	38
13	RSV bronchiolitis requiring intensive care	34	0	1	0	4	0
14	Fall on stairs, CVA, acute hydrocephalus requiring ventriculoperitoneal shunt	0	38	0	1	0	0
15	Cardiac surgery complicated by CVA	0	0	38	1	0	0
16	Pneumonia complicated by sepsis	38	0	0	0	0	1
17[a]	Brain surgery complicated by meningitis	16	0	21	1	0	1
18	Road traffic accident	0	39	0	0	0	0
19[a]	Gastrointestinal infection complicated by CVA	25	0	0	4	3	7
20	Cardiac arrest	0	0	1	0	38	0
21	Physical abuse	0	37	0	2	0	0
22	CVA due to sickle cell disease	0	0	0	34	2	3
23	Encephalopathy without infection identified or suspected	5	1	0	0	4	29
24[a]	Covid infection and myocarditis requiring ECMO followed by CVA	17	0	5	14	3	0

Abbreviations: CVA, cerebrovascular accident; RSV, respiratory syncytial virus; ECMO, extracorporeal membrane oxygenation.

The grey shaded fields represent the category expected by the expert group who prepared the clinical vignettes.

[a]Cases 'with complex causal chain'.

The clinical vignettes were divided into cases 'with complex pathway leading to cerebral palsy' ($n = 8$) and cases 'with single identified event' ($n = 16$) leading to cerebral palsy. Participants were not aware of this distribution.

TABLE 4 Interrater reliability for the six main categories of the classification overall and according to complexity of the pathway leading to cerebral palsy.

	Percent agreement	Gwet's AC1 coefficient (95% CI)	Scott/Fleiss' kappa coefficient (95% CI)
Total number of cases	0.79	0.75 (0.64–0.86)	0.73 (0.62–0.85)
Cases with complex pathway leading to cerebral palsy	0.55	0.48 (0.31–0.65)	0.36 (0.18–0.53)
Cases with single identified event	0.91	0.89 (0.81–0.97)	0.89 (0.80–0.97)

Abbreviation: CI, confidence interval.

it was almost perfect for the 16 cases with single identified events (Gwet's AC1 coefficient 0.89). Agreement was slightly better when the survey was completed after the reminder (22 responses, 56.4%). Finally, agreement did not markedly differ according to raters' profile (Table S2). Regarding the high participation of the Portuguese registry (11 of the 39

responses), analysis showed that their level of agreement did not differ from the overall results (data not shown). Likewise, influence analysis did not reveal any outliers.

Among the 237 children with postneonatally acquired CP born between 2008 and 2012 included in the SCPE central registry, the age at onset of brain damage remained unspecified for eight cases (with the possibility that brain injury occurred after 2 years) and for another eight, no description was available. The final sample comprised 221 children with postneonatal CP. The distribution of the primary event is presented in Table 1. Category A 'infection' was the most prevalent (32.1%) with mainly subcategory A1 reflecting either encephalitis with direct nerve cell infection or meningitis, and subcategory A3 comprising the consequences of other infectious diseases that can lead to CP by unspecified mechanisms (such as respiratory infections, gastroenteritis) or all infection-related encephalopathies, when the pathogen does not directly infect the central nervous system. Subcategory A2 'severe sepsis, septicaemia, or septic shock' leading to CP by unspecified mechanisms was rare. Category B 'head injury' accounted for almost a quarter of cases (23.1%). Category C involved 15.4% of cases (mainly related to cardiac or brain surgery/procedures). The CVAs (category D) accounted for 13.1%, whereas category E (hypoxic events) accounted for 9.1%. Lastly, 7.2% of the events were classified in the 'miscellaneous' category.

DISCUSSION

This paper introduces a classification of events contributing to brain lesions underpinning postneonatally acquired CP developed by the SCPE network. This classification aims to record the 'primary event' leading to CP with sufficient detail (19 subcategories) to accurately describe events involved for prevention purposes, while allowing the prevalence and evolution of the six main categories to be studied. While overall interrater reliability was substantial, it was only moderate for the most complex cases but was almost perfect when one single identified event was involved.

In addition to the classic categories of infection and brain trauma, we chose to individualize a surgical sequelae category (C) and a hypoxic category (E) to identify situations amenable to preventive measures (e.g. cardiac surgery) or that could have been so in the past (e.g. sudden death). On the other hand, category D includes non-traumatic and non-related to surgery cerebrovascular events which represent mainly non-easily preventable causes of stroke. Adding these categories also allows for a more homogeneous record that is easier to compare over time and across different settings. Another essential point concerns the choice of the 'main' event to be reported which, especially in the case of complex pathways leading to CP, affects the results and can differ significantly between studies without being generally clearly specified. This demonstrates the need for a common international shared understanding of classifying complex pathways and the need for clear coding instructions. However, it is essential to leave the possibility of capturing the full chain of events for more detailed studies focusing on a particular event. We therefore recommend using this classification in a two-step process: first selecting all the categories corresponding to the events potentially involved, and second choosing the primary event to be reported first.

Within the SCPE network, the expert panel will analyse all cases of postneonatal CP newly entered in the SCPE database (free text describing the clinical pathway leading to CP, classification of all events potentially involved, and designation of the 'primary event') to ensure accuracy and reproducibility of coding over time. However, the more general aim is the widest use possible. Using the same classification should simplify international comparisons. In addition, international agreement on an upper age limit of 2 years after birth for brain injuries is needed, as some surveillance systems record cases with a higher upper age limit.[24]

The interrater reliability exercise was successful, with 39 participants from 21 of the 24 registries approached. The results of the exercise were also very encouraging. The substantial overall interrater reliability showed that the professionals working in CP registries consistently distinguished between the different main categories of the classification and suggested that variations between ratings were fairly low. The choice of the clinical vignettes was crucial. Overall, the exercise was designed to represent as many different clinical situations and examples of subcategories as possible. For simple cases, agreement was almost perfect. For complex cases, the kappa coefficient was found to be lower than Gwet's AC1, which was expected since the kappa coefficient is known to be negatively influenced by the fact that cases were not evenly distributed across categories.[22] Nevertheless, the stability of the Gwet coefficients is quite reassuring, and even if the agreement for complex cases is decreased compared with the simplest ones, it remains moderate. Lastly, the difficulty of explaining the concept of choosing the primary chronological event should probably also be stressed, although many examples are provided in the coding instructions. The (albeit moderate) increase in agreement observed after the reminder supports the need for following these instructions to improve reliability. Finally, in the interrater reliability exercise, participants were asked to directly designate the 'primary event'. However, we believe that giving the opportunity to record several involved events will improve the reliability of the designation of the primary event, as participants may feel more comfortable if they are confident that no important information is lost.

One limitation is that our results are specific to this particular study; they depend on both the provided clinical vignettes and the professionals involved, which could limit the generalizability of the results. However, these 24 vignettes covered a wide range of chain of events contributing to brain lesions with some complex ones for which agreement was expected to be lower. Moreover, the participants were mostly professionals experienced in registry work, which we believe is a reassuring element in the practical application of this classification. Another limitation is that this classification was developed based on the descriptions reported

in the SCPE, thus reflecting events involved in high-income countries. However, while the prevalence of postneonatal CP cases in low- and middle-income countries may be higher and the potential events contributing to cerebral lesions may differ in their distribution,[14] the range of possible identified events remains almost identical, and the classification is sufficiently detailed for all potential events to be easily recorded in any setting.

The distribution of events contributing to cerebral lesions for the last five validated birth years was presented to better introduce the classification and its applicability. Comparison with data published elsewhere is challenging. First, since events involved in postneonatal CP are assumed to change over time, it is obvious that any comparison with publications reporting data for children born before 1999[1–5] cannot be done without complementary data on prevalence. This was not the objective of this work and a future study will present in detail the long-term trends in the SCPE network. Second, comparisons are difficult because of disparities in the content of the classifications, in the number of categories, and in the choice of the event to be considered. This underlines the value of an international use of a single classification such as the one proposed. For now, even when comparing with data corresponding to similar generations[9,25] the proportions observed are different. For instance, depending on the classifications used, some identified events may be categorized as CVA, surgical sequelae, or even hypoxia. In the Australian report,[9] CVA are involved in 33.6% of the cases and included CVA associated with surgery, with cardiac complications or spontaneous. For comparison with the data presented here, category D and category C should be combined for a relatively similar total of 28.5%. However, it is not identical since category C includes all consequences of surgery, whatever the mechanism (i.e. CVA or hypoxia), whereas in the Australian report,[9] perioperative hypoxia is classified separately.

Conclusion

With new structured main categories and subcategories, the ability to describe in detail all the events involved in postneonatal CP and then designate the primary event to be reported in a reproducible way is a strength of this new classification.

Adoption of such a classification by a large number of registries should contribute to measure the impact of prevention initiatives in different settings.

ACKNOWLEDGMENTS

This study was performed on behalf of the SCPE collaboration. We are grateful to all registries across Europe contributing data and/or participating to the interrater reliability exercise: France (C Arnaud, M Delobel, D Klapouszczak, A Ego, A Montovert, C Tronc), Northern Ireland (O Perra, K McConnell, C Kerr), Sweden (K Himmelmann, M Påhlman), Ireland (O Hensey, V Dowding), Tubingen (I Krageloh-Mann, V Horber), Norwich (M J Platt), Denmark (G Rackauskaite, B Laursen), Italy (M Marcelli, M Tacke), Norway (G L Andersen, S Julsen Hollung), Spain (J De La Cruz), Slovenia (D Neubauer, A Troha Gergeli), Portugal (D Virella, T Folha), Latvia (A Greitane, L Ceiciniece), Hungary (K Hollódy, E Nagy), Iceland (S Sigurdardottir), Belgium (E Ortibus, I Franki, E Dhondt), Croatia (V Mejaški Bošnjak, I Daković), Switzerland (C Kuenzle, A Tscherter), Greece (A Papavasiliou, M Petra, S Mastroyianni), UK (K Horridge, C Harvey), Moldova (E Gincota Bufteac). This publication was possible thanks to the data collection and management performed by the JRC-SCPE Central Registry, part of the EU Platform on Rare Diseases Registration.

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

FUNDING INFORMATION

Research was conducted by the authors within their academic institutions.

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.

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SUPPORTING INFORMATION

The following additional material may be found online:

Table S1: Instructions for use of the SCPE classification of events contributing to the brain lesion underpinning of postneonatal CP.

Table S2: Interrater reliability for the six main categories of the classification by raters' characteristics.

How to cite this article: Pudig L, Delobel-Ayoub M, Horridge K, Gergeli AT, Sellier E, Ehlinger V, et al. Classification of events contributing to postneonatal cerebral palsy: Development, reliability, and recommendations for use. Dev Med Child Neurol. 2024;66:250–257. https://doi.org/10.1111/dmcn.15710