

Article details: 2017-0013	
Title	Accuracy of administrative claims data for cerebral palsy: a retrospective cohort study
Authors	Maryam Oskoui MDCM MSc, Pamela Ng MSc, Marc Dorais MSc, Nicole Pigeon MD, Louise Koclas MD, Céline Lamarre MD, Francine Malouin PhD, Carol L. Richards PhD, Michael Shevell MDCM, Lawrence Joseph PhD
Reviewer 1	Guro Andersen MD PhD
Institution	The Cerebral Palsy Registry of Norway, Habilitation Center, Vestfold Hospital, Tønsberg, Norway
General comments (author response in bold)	<p>Very interesting and important study showing the usefulness of a registry. The manuscript is clearly and well written. Thank you.</p> <p>1. Other countries have performed similar studies (Hollung et al DMCN 2016 "Completeness and correctness of cerebral palsy diagnoses in two Health registers: implications for estimating prevalence"). supporting the conclusion. I recommend to include this and other possible references to increase the generalizability of the publication. References to other studies have been included in the Interpretation section for comparison with our findings.</p>
Reviewer 2	Lorna Fraser MSc PhD
Institution	Department of Health Sciences, University of York, York, UK
General comments (author response in bold)	<p>I welcome the authors' attempts to assess the accuracy of administrative health data but I have some major questions that would need to be addressed around the methodology before I could assess whether this study should be published. Therefore I have not listed the many minor issues with this paper in terms of readability but I would be happy to review again if the authors can answer my very important methodological questions below:</p> <p>Data sources</p> <p>1. More information on CP register eligibility, e.g. for residents of the region at the time when they are age 2 onwards or only those born in the region.</p> <p>2. The authors have assumed that the CP register is a high quality, complete dataset. Is there evidence to show this? The average of diagnosis of age 6 years seems very high to me. The average age of diagnosis of 6 years was not for the registry cohort, but for the false negatives (i.e. those children with a CP diagnosis in the administrative health databases, but not in the Registry). This has been clarified in the text.</p> <p>3. Also what if a child was originally diagnosed as CP then later given a different diagnosis? Does the register record if the child has died or moved to another region. The Registry only includes children with a confirmed diagnosis of CP at 5 years of age. If their diagnosis has changed they are removed from the database.</p> <p>4. How was the linkage undertaken? If it was undertaken as explained on page 5 and 6 then how did you know about children coded as CP in the AHD and not the register? Suggest that the authors also follow the Strobe RECORD reporting guidelines as more detail about the data sources and linkages are required. The strobe RECORD reporting guideline was followed. Thank you.</p> <p>5. Why was the study period from 1999-2002? surely if 5 years follow up was important then a more recent time period could have been used? As stated earlier, and in the manuscript, we chose the oldest cohorts in the registry (children born 1999-2002) to allow the longest followup period and also the most complete ascertainment. The data we obtained will be used for diagnostic validation, but also to look at early childhood healthcare utilization (which will be the subject of another manuscript).</p> <p>6. The results are very simplistic and could be expanded upon. What about recording of related ICD10 codes? It would have been useful to know if these children had no neurology related ICD9 or ICD10 codes recorded in the AHD or if they were recorded with very nonspecific neurological ICD10 codes or indeed some may have congenital anomaly codes. The goal of this study was to show the validity of the CP codes within administrative health databases. Several previous studies, including two prevalence estimates in Alberta and British Columbia, had simply used this code for the study. Our goal was to show the misclassification that this code represents. We are now working on elaborating a more sensitive diagnostic algorithm including these other related codes, with a Bayesian statistician, to create a more robust diagnostic algorithm.</p> <p>7. The data on those only found in the AHD dataset requires more explanation, did they only include those who had the same eligibility criteria as the register (> 2 years of age etc). This has been further explained in the manuscript.</p>
Reviewer 3	Sarah McIntyre PhD MPS BAppSc (Hons)
Institution	University of Sydney, Camperdown, Australia
General comments (author response in bold)	<p>Thank you for asking me to review this manuscript. In an era where there is an influx of the use of administrative data sets for epidemiological research (not their primary purpose), it is important to know "what is in these administrative data sets" and how much confidence we can have in their findings. The authors are to be congratulated for taking the time to look into the accuracy of CP in administrative health databases. Could the authors comment or include in their paper the following: Thank you</p> <p>Participant selection:</p> <p>1. It is currently unclear what years the administrative health records were searched for when trying to identify the CP cases from REPACQ. This has been clarified in the methods (from birth 1999 to 2002, to December 2012).</p> <p>Results:</p> <p>1. The false negatives suggest that one third of all those on REPACQ were not identified by the health administrative databases. This is a large proportion. If this is indeed the case, administrative data sets should never be used for "total population" research. As someone who does not live in Canada, I am unsure of whether "day procedures" such as botulinum toxin A would be included in these hospital data sets. Do the authors think that if they did this again in a later year cohort (ie birth years 2006-9), whether these milder children with GMFCS I-II and hemiplegia would be better ascertained? The administrative health data we obtained includes both RAMQ data for outpatient medical visits, and MED-ECHO</p>

data for hospitalizations. We are exploring other diagnostic code to increase the sensitivity of capturing children with hemiplegic CP (and all profiles of CP) by developing an algorithm with a Bayesian statistician. This will be a separate manuscript. Our goal here is to show that the commonly used CP codes should not be taken at face value as being valid, as they are increasingly used in research and misrepresent such a wide spectrum of CP disorders.

2. For the 11 that were unmatched, does this mean that they have never been to a hospital or claimed for anything medically related to their CP? Were these children mild, as well?

The 11 children who could not be matched: It is not related to their medical visits or severity, but rather a technical difficulty to link the two databases.

3. There is a presumption made that those not in REPACO (n=138) but had a code of CP in the administrative databases are false negatives, ie didn't actually have CP. How sure are the authors about this presumption? In a recent Norwegian article doing a similar investigation 60% of those in hospital records but not on the Register had a correct diagnosis of CP (39% did not). They concluded that using administrative data sets increases the completeness of a consent based register. Could this also be the case in Canada?

The study from Norway found that only 60% of children registered as having CP in one of the two national registries had confirmed CP after hospital record review by expert neurologists. They did not look at CP codes across the whole population in administrative health data. They showed that their registry underestimates prevalence estimates, and that admin data would over estimate prevalence estimates.

We acknowledge in our limitations that the registry could miss cases of CP (who died before two year of age, or who had such mild functional impairment that they did not come to medical or allied health attention).

Discussion:

1. Should (due to these findings) we be suggesting that administrative data sets only be used for those studies looking at CP with moderate to severe motor impairment? That we can't be confident that they are identifying those with mild CP (which is 50% of CP).

Yes. I would also suggest linking multiple data sources as the optimal method.

2. Is it a limitation of the study, that the investigators did not look into the false negatives?

The false negatives are described in the results section. We are now exploring other diagnostic codes to develop a more sensitive algorithm, and the codes found within this subgroup will be explored. This was not the goal of the current manuscript.

The paper is very well written, and the tables/figures are clear and concise. This is an important paper for those who wish to study the epidemiology, for health economists, those who plan services and health utilization.

Thank you.