

innovation without evaluation: the IDEAL recommendations. *Lancet* 2009; **374**: 1105–12.

3. Hepp ZS, Haas VM, Latal B, Meuli M, Möhrlein U, Schauer SM, et al. Early childhood neurodevelopmental outcome after open prenatal spina bifida aperta repair. *Dev Med Child Neurol* 2021; **63**: 1302–7.
4. Worley G, Greenberg RG, Rocque BG, Liu T, Dicianno BE, Castillo JP, et al. Neurosurgical

procedures for children with myelomeningocele after fetal or postnatal surgery: a comparative effectiveness study. *Dev Med Child Neurol* 2021; **63**: 1294–301.

5. Espinoza J, Shamshirsaz AA, Sanz Cortes M, Pammi M, Nassr AA, Donepudi R, et al. Two-port, exteriorized uterus, fetoscopic meningocele closure has fewer adverse neonatal outcomes than open hysterotomy

closure. *Am J Obstet Gynecol* 2021. S0002-9378(21)00534-2. Online ahead of print.

6. Joyeux L, De Bie F, Danzer E, Russo FM, Javaux A, Peralta CFA, et al. Learning curves of open and endoscopic fetal spina bifida closure: systematic review and meta-analysis. *Ultrasound Obstet Gynecol* 2020; **55**: 730–9.

## Methodological quality of systematic reviews on interventions for children with cerebral palsy: the evidence pyramid paradox

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**This commentary is on the original article by Kolaski et al. on pages 1316–1326 of this issue.**

The paper by Kolaski et al.<sup>1</sup> poses a series of questions related to evidence generation and appraisal that, albeit not new, cannot be ignored any further. These questions involve scientists, journals, and developers of clinical practice guidelines. However, the most critical, potentially devastating impact is on patients for whom research in medicine is undertaken.

Generating trustworthy evidence is the only way forward for medicine. This was made very clear during the COVID-19 pandemic, which included issues around rehabilitation.<sup>2,3</sup> However, research papers cannot be taken at face value: they must be appraised. The most naïve approach is to infer quality by using the primordial pyramid of evidence: the better the design, the lesser the risk of bias, the more reliable the results. Consequently, randomized controlled trials and meta-analyses are at the top. Nevertheless, the revised evidence pyramid includes blurred boundaries between the different layers:<sup>4</sup> for example, a bad quality randomized controlled trial is worse than a good quality cohort study. We would argue that a bad quality meta-analysis is even worse. It can spread false results under the misleading appearance of a reliable study.

The bleak picture on the quality of systematic reviews provided by Kolaski et al.<sup>1</sup> may be partly attributed to a zealous application of the AMSTAR-2 criteria. Nevertheless, the message is clear: authors of systematic reviews must do better. In the modern ‘publish or perish’ era,<sup>5</sup> researchers must rapidly produce results beneficial for their

career, funders, and/or employers. Systematic reviews could be regarded as less demanding than clinical trials, mainly when they are not well done. Controlling the methodological quality of these and other papers resides on the shoulders of journal editors and reviewers. Reviewers are the most important, yet weakest link of the peer review chain. Reviewers are made up of researchers and clinicians with similar constraints as the authors: working as unrewarded volunteers and seldom explicitly trained on the job. In clinical fields, clinician-researchers must manage the time pressure of combining patient care with research activities. Therefore, this link of the chain can easily break, and the quality of reviews suffers. Although editors provide review tools to guide peer reviewers (<https://www.equator-network.org/>), these cannot compensate for lack of time, thus impairing the system.

An even more devastating issue is the unreliability of clinical practice guidelines that use flawed systematic reviews. In the knowledge translation process, clinical practice guidelines are essential to inform clinicians, health decision-makers, and patients. Therefore, the methodology used to conduct clinical practice guidelines must be more rigorous than in primary and secondary research papers. Clinical practice guidelines are expensive to produce, are resource-intensive, and require the involvement of renowned scientists and research teams who must search the evidence and even more importantly, appraise it properly.

Finally, Kolaski et al.’s paper<sup>1</sup> confirms that adhering to the Cochrane methodology to conduct systematic reviews leads to high-quality reviews. However, chairing the Cochrane Rehabilitation Field, we see from within that too many authors do not commit to a Cochrane Review due to the effort and time required to respect the high methodological standards that guarantee the final quality. More rigorous literature searches, standardized application of risk of bias tools, and analyses and reporting of results that incorporate quality appraisal may be more time-consuming and labour intensive, but will improve future systematic reviews and ultimately have a positive impact on general health.

## REFERENCES

1. Kolaski K, Logan Romeiser, Goss KD, Butler C. Quality appraisal of systematic reviews of interventions for children with cerebral palsy reveals critically low confidence. *Dev Med Child Neurol* 2021; **63**: 1316–26.
2. Ceravolo MG, de Sire A, Andrenelli E, Negrini F, Negrini S. Systematic rapid “living” review on rehabilitation needs due to COVID-19: update to March 31st, 2020. *Eur J Phys Rehabil Med* 2020; **56**: 347–53.
3. Ceravolo MG, Arienti C, de Sire A, et al. Rehabilitation and COVID-19: the Cochrane Rehabilitation 2020 rapid living systematic review. *Eur J Phys Rehabil Med* 2020; **56**: 642–51.
4. Murad MH, Asi N, Alsawas M, Alahdab F. New evidence pyramid. *Evid Based Med* 2016; **21**: 125–7.
5. Relman AS. Publish or perish—or both. *N Engl J Med* 1977; **297**: 724–5.

# Making cerebral palsy visible across the globe

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In 2014, the epidemiologist Maureen Durkin wrote that cerebral palsy (CP) is ‘... an invisible public health problem in most of the world’.<sup>1</sup> Nearly all our information about CP – its root causes, manifestation, and consequences – come from studies performed in just a handful of countries. Ongoing surveillance of CP is established in much of Europe and Australia, but even in some high-income countries (HICs) such as the USA, CP surveillance is limited and occasional.

Durkin could find just seven population-based studies of CP from low- to middle-income countries (LMICs) published between 1987 and 2012. Since then, some useful papers have been published, notably from Uganda,<sup>2</sup> where a population-based surveillance study was performed with as much rigor as any study performed in an HIC. That investigation began to lift the veil from CP in LMICs, finding that CP with a background of preterm birth was rare. The authors speculated (and the commenters agreed),<sup>3</sup> that the rarity of CP in infants born preterm was due to the high mortality of severely ill infants in locations where newborn infant intensive care is unavailable.

This finding is partially replicated in the report by Jahan et al.,<sup>4</sup> which adds to the modest literature on CP in

LMICs by summarizing some early findings from CP registries in Bangladesh, Nepal, Indonesia, and Ghana. The three cohorts that are population-based (all but Ghana) also found fewer than expected cases of CP in infants born preterm.

Jahan et al. suggest some other themes that differentiate CP in LMICs from CP in HICs. First, educational and rehabilitative services are much less available to children with CP than would be ideal. Second, poverty plays an even stronger role as a social antecedent of CP than it does in HICs, in part because of a high prevalence of home deliveries without trained birth attendants. Third, cases are skewed towards high severity, no doubt reflecting the difficulty of ascertaining milder cases.

Even with fewer milder cases, and fewer surviving infants born preterm, CP prevalence is likely to be higher in LMICs than in HICs. This preliminary report does not give prevalence figures, but a study from Bangladesh showed a prevalence of 3.5 per 1000 live births<sup>5</sup> and the Uganda study found a prevalence of 2.7 per 1000, both figures higher than in most HICs. The Uganda finding of a high proportion of post-neonatal cases – likely due to cerebral malaria – was replicated only in the Indonesian cohort where viral infections were thought responsible.

A high percentage of CP in LMICs could be remedied with effective public health and primary care services. Bilirubin and anoxic encephalopathies could be prevented or managed. As Durkin pointed out in 2014, ‘... human development and the brain development of infants and children in LMICs are being compromised globally each day because of exposures and causes we know how to prevent’.<sup>1</sup>

## REFERENCES

1. Durkin MS. A global perspective. In: Dan B, Mayston M, Paneth N, Rosenbloom L, editors. *Cerebral Palsy. Science and Clinical Practice*. London: Mac Keith Press, 2014.
2. Kakooza-Mwesige A, Andrews C, Peterson S, Wabwire Mangen F, Eliasson AC, Forsberg H. Prevalence of cerebral palsy in Uganda: a population-based study. *Lancet Glob Health* 2017; **5**: e1275–82.
3. Dan B, Paneth N. Making sense of cerebral palsy prevalence in low-income countries. *Lancet Glob Health* 2017; **5**: e1174–5.
4. Jahan I, Muhit M, Hardianto D, et al. Epidemiology of cerebral palsy in low and middle-income countries: preliminary findings from an international multicentre cerebral palsy register. *Dev Med Child Neurol* 2021; **63**: 1327–36.
5. Khandaker G, Muhit M, Karim T, et al. Epidemiology of cerebral palsy in Bangladesh: a population-based surveillance study. *Dev Med Child Neurol* 2019; **61**: 601–9.