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ORIGINAL ARTICLE

Prevalence of Cerebral Palsy in Indian Children: A Systematic Review and Meta-Analysis

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Abstract

Objective To determine the pooled-prevalence of cerebral palsy in Indian children.

Methods The authors searched the published literature from different databases (PubMed, Ovid SP and EMBASE) and also tried to acquire information from the unpublished literature about the prevalence of cerebral palsy. They screened prospective/retrospective, cross-sectional, and cohort studies of children with cerebral palsy in the Indian population. Data were extracted from the included studies, and quality assessment was performed. Data were analysed using STATA MP12 (Texas, College Station).

Results Of the 862 publications searched, eight studies were qualified and included for quantitative analysis. The overall pooled prevalence of cerebral palsy per 1000 children surveyed was 2.95 (95% CI 2.03–3.88). Sub-group analysis for rural, urban and mixed rural-urban study population demonstrated the pooled prevalence as 1.83 (95% CI 0.41–3.25), 2.29 (95% CI 1.43–3.16) and 4.37 (95% CI 2.24–6.51) respectively.

Conclusions This systematic review observed a paucity of high-quality, prevalence studies of cerebral palsy in India, which is a limitation to estimate the inferences for a national estimate. The observed prevalence of cerebral palsy in India is near similar to global estimates. There is a need to re-allocate resources and revisit the implementation of the existing policies for the prevention and management of cerebral palsy, taking into account the current disease burden.

Keywords Cerebral palsy · Children · Systematic review · Prevalence · India

Introduction

The term 'Cerebral palsy' is defined as a group of permanent disorders of the development of movement and posture, that cause activity limitation, and are attributed to non-progressive insults to the developing fetal or infant brain. The motor impairment of cerebral palsy is often accompanied by sensory disturbances, perception, intellectual disability, communication, behavior, by epilepsy and by secondary musculoskeletal problems [1]. Worldwide, it is one of the most common causes of disability in children.

Globally, studies have reported the prevalence range of cerebral palsy from 1.5 to 4 per 1000 live births or children [2–5]. In the past decade, three pertinent reviews about the prevalence of cerebral palsy have been published [6–8]. First, Hirtz et al. reported an overall prevalence estimate of 2.4 per 1000 live births in the United States [6]. Second, Himpens et al. reported the prevalence of cerebral palsy with relation to gestational age and demonstrated a significant decrease in the prevalence of cerebral palsy with increasing gestational age [7]. Third, Oskoui et al. reported that the overall prevalence of cerebral palsy has remained unchanged in recent years despite improved survival of at-risk preterm infants [8].

India is the second most populated country in the world with more than a billion people. This still growing population imposes a significant burden on the healthcare system. According to the National Family Health Survey (NFHS) 2015–16, 79% of childbirths took place in a health facility,

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while the rest were possibly conducted at homes by untrained birth attendants [9]. When translated into actual figures, the number of unsupervised obstetric deliveries in India is still huge. Such deliveries have a very high rate of obstetric complications and as a result, perinatal asphyxia. Furthermore, with improvement in neonatal care services in India, there is increased survival of very low birth weight and premature babies. Both perinatal asphyxia and prematurity constitute a major risk factor for cerebral palsy.

The available studies are not representative of the national prevalence estimates of cerebral palsy in Indian children. Hence, there is an urgent need to collate the prevalence rates of cerebral palsy from regional studies in Indian children to facilitate health policies formulation and to seek specific allocation of resources for early diagnosis and management of this disease. The present systematic review was aimed to determine the pooled prevalence of cerebral palsy as no such reviews have been conducted.

Material and Methods

The authors searched the published literature from different databases (PubMed, Ovid SP and EMBASE) and also tried to acquire information from the unpublished literature. The searches were current as of December 2018, and articles with information on the prevalence of cerebral palsy in Indian children were identified. The search strategy included the following search terms: ((((((“Cerebral Palsy”[Mesh] OR “Cerebral Palsy, Ataxic, Autosomal Recessive” [Supplementary Concept] OR “Cerebral palsy, spastic, diplegic” [Supplementary Concept] OR “Cerebral Palsy, Spastic Quadriplegic, 2” [Supplementary Concept] OR “Cerebral Palsy, Spastic Quadriplegic, 1” [Supplementary Concept] AND ((((infant) OR pediatrics) OR children) OR child))) AND India)) AND prevalence.

Prospective/retrospective, cross-sectional, and cohort studies of children with cerebral palsy in the Indian

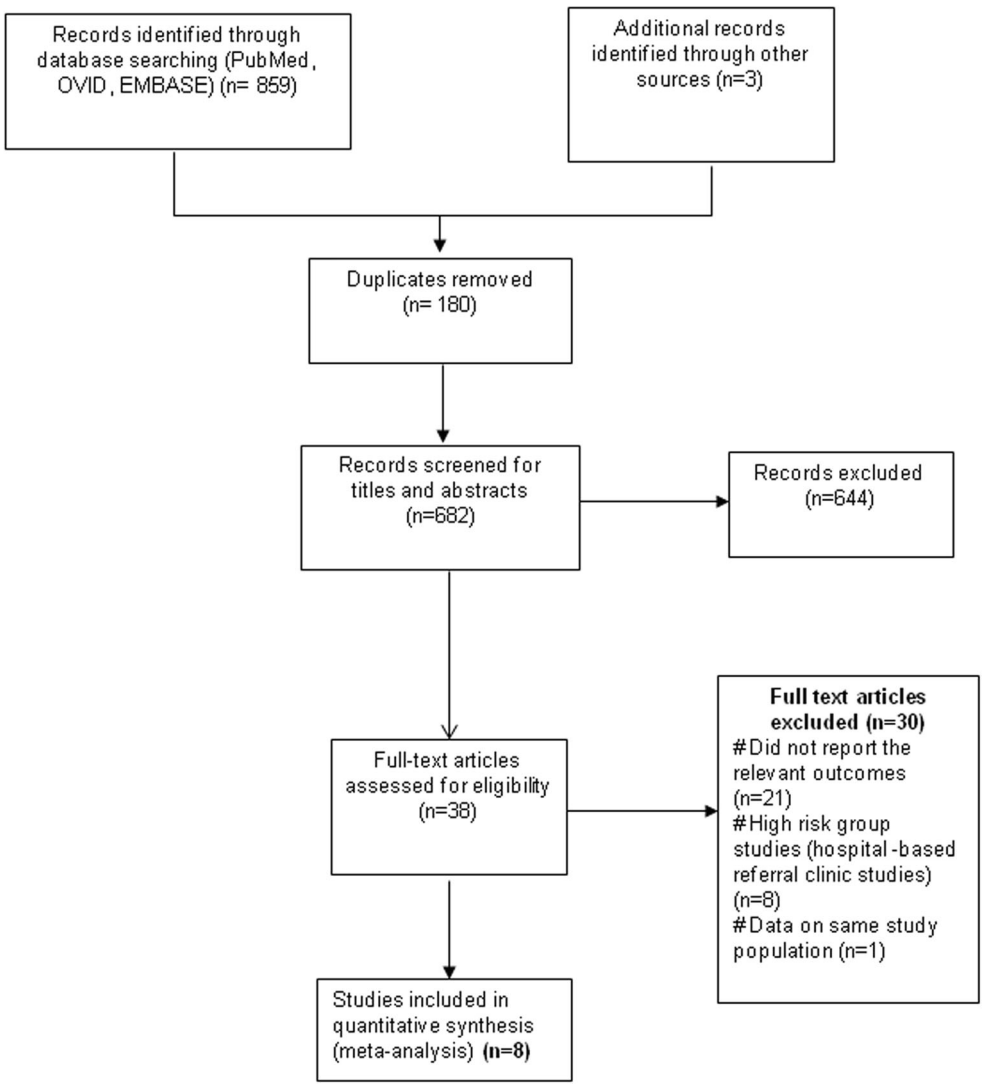

Fig. 1 The PRISMA flow diagram of literature search and for selection of studies

population were screened. The titles and abstracts of all searches were screened for obvious relevance by three authors (AC, MaS and JKS) through covidence (www.covidence.org), which is a core component of the Cochrane review production toolkit. Subsequently, AC and JKS retrieved the full-text of potential studies for comprehensive screening for eligibility. Inclusion criteria for studies were community-based prevalence studies of cerebral palsy in children aged 1–18 y in India. Discrepancies if any were resolved through discussion with the MS and her verdict was considered to be final. The reasons for exclusion of studies were mentioned in the PRISMA flow diagram (Fig. 1). Data extraction table was prepared to extract data from the included studies. Data were extracted from the included studies by four authors independently (AC, MaS, JKS and AA). After data extraction, the data analysis was done through STATA version 12.0. The authors checked for any error in data being entered in STATA MP12 software by directly comparing with the included studies.

AC and JKS assessed the quality of the included studies by using quality assessment tool developed from publications by Boyle MH and Loney PL et al. [10, 11]. The quality was assessed for the representativeness of the sample, assessment of neurological conditions and statistical analysis done. 'Trim and fill' method was used to determine and rectify for funnel plot asymmetry due to publication bias.

Three authors (NJ, AC & JKS) did the data analysis using STATA MP12 (Texas, College Station). Sub-group analysis was done based on study population belonging to rural, urban or mixed rural-urban setting. Heterogeneity was measured through the Galbraith plot.

Results

Of the 862 publications searched, 180 were removed as being duplicates. Titles and abstracts of 682 publications were screened and 644 publications were excluded as being obviously irrelevant. Thirty eight full-text studies were assessed for eligibility. Finally, eight studies were included in quantitative analysis (Fig. 1) [12–19].

The demographic characteristics of the included studies are provided in (Table 1). The included studies have used varied screening and diagnostic tools such as INCLEN Diagnostic Tool for Neuro-Motor Impairments,Trivandrum Developmental Screening Chart (TDSC), Denver Developmental Screening Test (DDST), pre-tested Performa for Disabled Children, Lucknow Neurodevelopmental Screen (LNDS) and WHO questionnaire (Table 2).

In the present systematic review, the overall pooled prevalence of cerebral palsy per 1000 children surveyed is 2.95

Table 1 Demographic characteristics of the included studies

S. No	Study ID	Year of publication	Age range in years	Rural/Urban	State	Number of children with cerebral palsy	Type of cerebral palsy	Total number of children surveyed
1.	Arora et al. [12]	2018	2–9	Rural-Urban	Palwal, Haryana; Dhenkanal, Odisha; Hyderabad, Telangana; and North Goa	29	Spastic ($n = 29$)	3964
2.	Kumar et al. [13]	2016	9–15	Rural-Urban	Lucknow, Uttar Pradesh	20	Not defined	6431
3.	Nair et al. [14]	2014	<6	Rural	Thiruvananthapuram, Kerala	112	Not defined	101,438
4.	Kumar et al. [15]	2013	6 mo – 2 y	Rural-Urban	Lucknow, UP	13	Not defined	4801
5.	Raina et al. [16]	2011	< 10	Urban	R.S. Pura Srinagar, Jammu and Kashmir	11	Spastic (quadriparesis - 6, diplegia - 4) ($n = 10$), Dyskinetic-1	3966
6.	Nair et al. [17]	2009	<5	Rural	Anganwadi ICDS block Pattanakad, Thiruvantampuram, Kerala	32	Spastic ($n = 24$), Hypotonic ($n = 7$), Mild ($n = 1$)	12,520
7.	Banerjee et al. [18]	2009	≤ 18	Urban	Kolkata, West Bengal	48	Spastic (diplegia - 35, quadriplegia - 2) ($n = 37$), Hemiplegia ($n = 6$), Dystonic ($n = 3$), Hypotonic ($n = 2$)	16,979
8.	Mathur et al. [19]	1995	< 6	Urban	Gorakhpur, Uttar Pradesh	2	Not defined	1545

Table 2 Screening tools and methodology of included studies

Study Id	Screening tools	Screening personnel	Clinical evaluation	Quality Score[a]
Arora et al. 2018 [12]	INCLEN Diagnostic Tool for Neuro-Motor Impairments	(a) The field team comprised of one physician and two social scientists (b) The diagnostic team comprised of one physician, one audiologist/speech therapist, and two psychologists	Physician assessment for neuromotor impairments including cerebral palsy	8
Kumar et al. 2016 [13]	Lucknow Neurodevelopmental Screen (LNDS)	Trained field investigators administered the LNDS in targeted households	Pediatric Neurologist and child psychologist clinically validated neurological disorders	7
Nair et al. CDC Kerala 16, 2014 [14]	Trivandrum Developmental Screening Chart (TDSC)	Trained Accredited Social Health Activist (ASHA) workers performed an initial screening	Clinical evaluation was done by specialists at Developmental Evaluation Camps	6
Kumar et al. 2013 [15]	LNDS & clinical validation	Trained field investigators administered the LNDS to targeted children in the house to house survey	Pediatric Neurologist and child psychologist clinically validated the neurological disorder	7
Raina et al. 2011 [16]	Modification of the WHO Questionnaire	Anganwadi workers carried out house to house survey and completed the screening questionnaire	Neurologist and an epidemiologist clinically examined the suspected cases	5
Nair et al. 2009 [17]	Trivandrum Developmental Screening Chart (TDSC), Denver Developmental Screening Test (DDST), Amiel Tison Passive Angle Method	Anganwadi workers and trained personnel did the development assessment	Detailed clinical examination and appropriate referral was done by a pediatrician and physiotherapist	6
Banerjee et al. 2009 [18]	Modification of the WHO questionnaire	Field worker performed door to door survey using a general screening questionnaire	Neurologists further clinically examined the cases at their homes and cases at dilemma were examined by a senior neurologist	6
Mathur et al. 1995 [19]	Pre-tested performa for disabled children	Anganwadi workers did the initial screening using pre-tested performa and were helped by pediatricians	Pediatricians examined all the cases and also confirmed the findings of Anganwadi workers	5

[a] Quality score ranges from 1 to 8 and a higher score indicates better-quality study

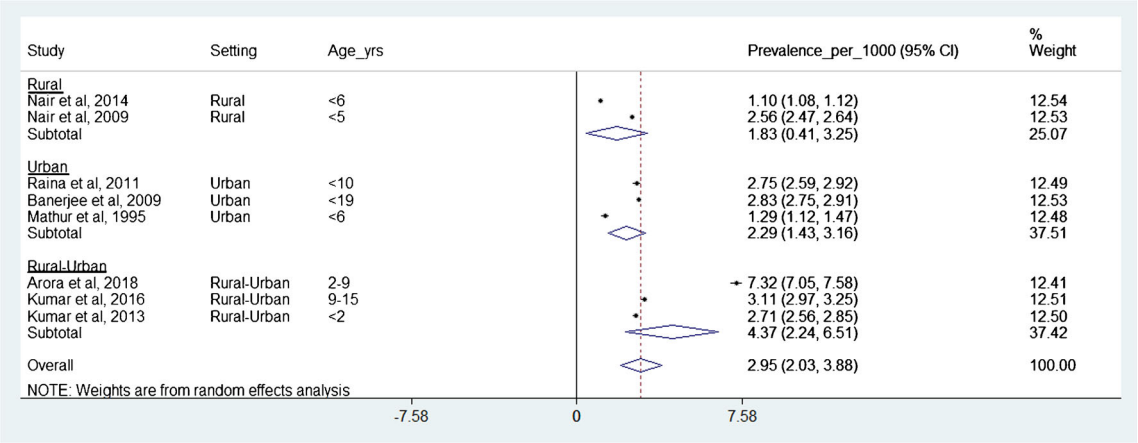

Fig. 2 Prevalence of cerebral palsy per 1000 children in urban and rural settings

(95%CI 2.03–3.88) (Fig. 2) [12–19]. Sub-group analysis was done based on the rural, urban and mixed rural-urban settings for the included studies (Fig. 2). The pooled prevalence derived from two studies conducted in rural settings is 1.83 (95% CI 0.41–3.25) [14, 17]. The pooled prevalence at urban settings is 2.29 (95% CI 1.43–3.16), based upon three studies [16, 18, 19]. In mixed rural-urban settings, the pooled prevalence of cerebral palsy per 1000 children surveyed is 4.37 (95% CI 7.05–7.58) [12, 13, 15] (Fig. 2, Table 2). A recent multicentric study by Arora et al. has reported the highest 7.32 (95% CI 7.05–7.58) prevalence among all the included studies [12].

Quality assessment was done using a quality assessment tool. The quality score ranged from 1 to 8, and a higher score indicated a better-quality study. The quality scoring for most of the included studies was greater than 4 (Table 2). Publication bias of the included studies was assessed through filled funnel plot. There was a significant publication bias in the present systematic review as most of the included studies were on the upper area of the plot. This publication bias could not be present if five studies were present in the lower area of the plot (Fig. 3). Heterogeneity among the included studies was reported through the Galbraith plot. There is significant heterogeneity in the present systematic review as demonstrated by the distribution of included studies in the Galbraith plot (Fig. 4).

Discussion

The present study is a singular systematic review on the prevalence of cerebral palsy in India. The prevalence of cerebral

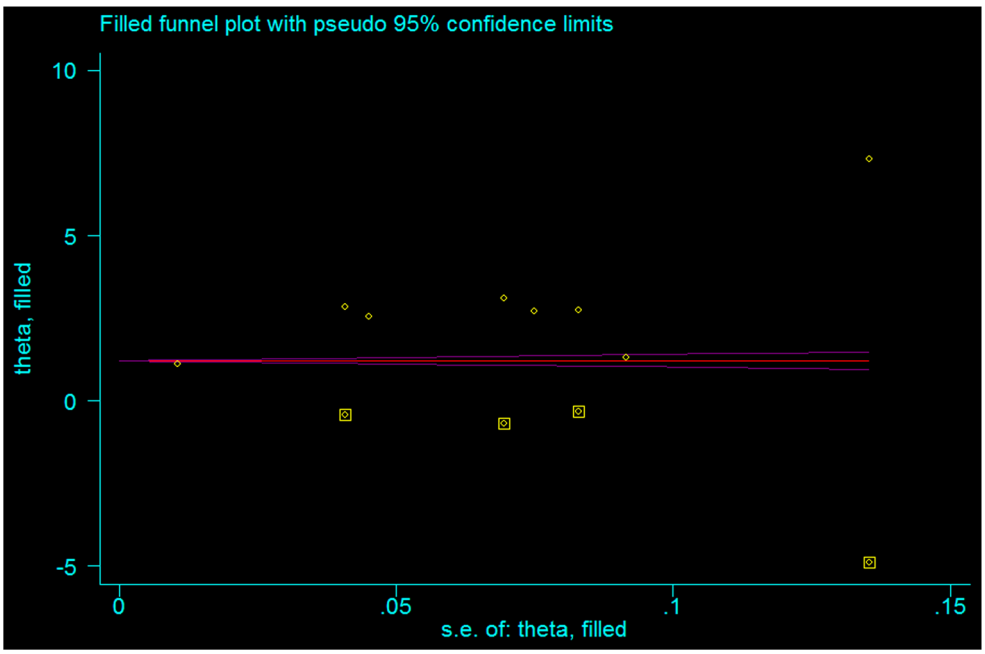

Fig. 3 Filled funnel plot with Trim and Fill method

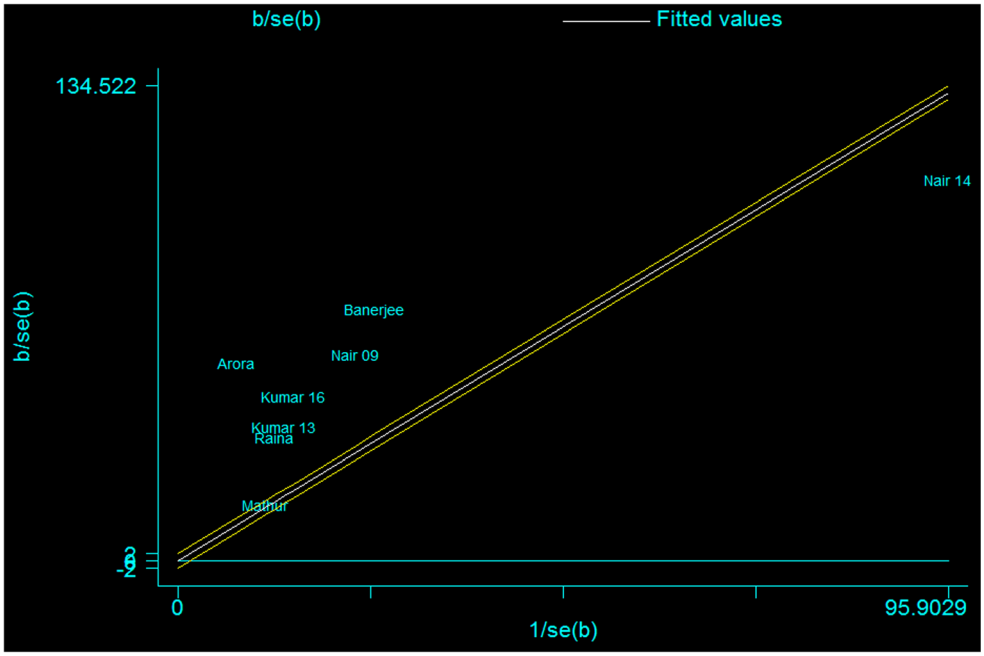

Fig. 4 Galbraith plot for analyzing heterogeneity

palsy in India is similar to global estimates. The present study also highlights a paucity of high-quality, population-based prevalence studies on cerebral palsy in India. Furthermore, there is a clinical heterogeneity across the studies based on the use of varied screening and diagnostic tools (Table 2). Random effects model for metanalysis was used as there was significant methodological heterogeneity between the included studies.

Four studies have classified cerebral palsy, based on the extent of neurological deficits, into monoplegia, hemiplegia, diplegia, triplegia and quadriplegia. However, the data could not be pooled for analysis due to significant heterogeneity and incomplete details (Table 1) [12, 14, 16, 18]. Of the included studies, Banerjee et al. reported that the majority of children with cerebral palsy had spastic diplegia. Preterm birth is an important risk factor for spastic diplegic cerebral palsy, while term birth asphyxia is a risk factor for spastic quadriplegic cerebral palsy [18]. Singhi et al., a study from a tertiary care hospital in North India, reported 1000 cases of cerebral palsy, and identified spastic quadriplegia (61%) as the most common type followed by diplegia (22%) [20].

A stringent methodology and quality assessment of included studies are strengths of present study. However, the present systematic review had a few inadvertent limitations. Firstly, authors have not analyzed the risk factors for cerebral palsy (prematurity, low birth weight) due to the inadequate available information. Secondly, there was heterogeneity in the diagnostic tools used in the included studies. Thirdly, they could not perform a time-trend analysis due to the limited number of published studies.

With the limitations of the study, it is concluded that the overall pooled prevalence of cerebral palsy per 1000 children surveyed is 2.95 (95%CI 2.03–3.88). The paucity of high-quality, prevalence studies of cerebral palsy in India is a barrier to estimate the inferences for a national estimate. There is a further need to conduct large good quality community-based studies to explore risk factors and type of cerebral palsy at different age groups. Meanwhile, the present study data would be useful to re-allocate resources and revisit the implementation of the existing policies for the prevention and management of cerebral palsy.

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Authors' Contribution The titles and abstracts of all potential studies were screened by AC, JKS & MaS. Data were extracted from the included studies by AC, MaS, JKS & AA. AC & JKS assessed for quality of the included studies by using a quality assessment tool. NJ, AC & JKS did the data analysis using STATA MP12 (Texas, College Station). Discrepancies if any were resolved through discussion with MS. AC, MaS and JKS wrote the initial draft and the final draft was approved by all the authors. MS is the guarantor for this article.

Compliance with Ethical Standards

Conflict of Interest None.

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