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## A systematic review of the worldwide prevalence of polio reported in 31 studies.

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3 A systematic review of the worldwide prevalence of polio reported in 31 studies.

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## ABSTRACT

**Background** Recent accurate prevalence rates estimating the number of polio survivors at any given time are not available. We aim to systematically review literature concerning the prevalence of polio worldwide.

**Methods** Electronic databases were searched up to May 2016 for peer reviewed studies including population-based approach with a defined denominator and some form of diagnostic or clinical verification of polio. Exclusion criteria were any prevalence data were unable to be extracted or calculated and studies reporting on incidence only. The quality of each included study was assessed using an existing tool modified for use in prevalence studies. Average crude prevalence rates were used to calculate worldwide estimates.

**Results** Thirty-one studies met criteria with 90% of studies conducted in low-to-lower middle income countries. Significant variability in the prevalence of polio was revealed, in low-to-lower middle income (15 per 100,000 in Nigeria to 1,733 in India) and upper-middle to high-income countries (24 (Japan) to 380 per 100,000 (Brazil). The total combined prevalence of polio for those studies at low to moderate risk of bias ranged from 165 (high-income countries) to 425 (low-to-lower middle income countries) per 100,000 person years. Lameness surveys of children predominated, with wide variation in case definition and assessment criteria.

**Conclusions** These results highlight the need for future epidemiological studies of polio to examine nationally representative samples, including all ages and more focus on high-income countries. Such efforts will improve capacity to provide reliable and more robust worldwide prevalence estimates.

### Strengths and limitations of this study

- As the battle to eliminate new cases of polio continues, there are no accurate prevalence rates of polio available.
- This is the first study to systematically review studies undertaken examining the worldwide prevalence of polio.
- Results from this review show that studies to date, primarily based on lameness surveys, demonstrate significant variability in the prevalence of polio across low to high income countries. Few studies have examined nationally representative samples thus limiting the reliability of current prevalence estimates.
- While efforts were undertaken to identify and access all relevant article, it is likely that some studies were not identified by the search strategy.
- The estimates provided in this review are likely an underestimation of the prevalence of polio, but findings support the need for future studies examining nationally representative samples that are designed to reduce bias noted in this review.

## INTRODUCTION

Poliomyelitis (polio) is a highly infectious, incurable viral disease that appeared in endemic form in 1900-1950[1], caused by a wild or live vaccine-derived virus. Polio invades the central nervous system[2,3] and sequelae may include permanent physical disability (60-90%), and respiratory, heart and musculoskeletal diseases.[4] Up to 40% of survivors will experience post-polio syndrome, being new or worsening disabling symptoms 30-40 years after the original infection.[5] Efforts to eradicate polio with mass vaccination programs have led to reductions in confirmed cases[1], from an annual rate of approximately 50,000 in 1980 to <1,000 in 2001.[6] Estimates suggest that 12-20 million individuals are living with polio sequelae worldwide.[7] However, recent accurate prevalence rates estimating the number of polio survivors at any given time are not available.[8] Published international prevalence studies are problematic as they have (i) tended to focus on the initial disease and needs in the immediate aftermath, (ii) been inconsistent in the definition of “polio survivor,” with it often unclear whether this refers all those infected or only those sustaining some form of residual disability, (iii) predominantly focused on health status rather than the everyday effects on people’s lives, their needs, and those of their carers, iv) produced inconsistent findings on long-term outcomes, perhaps due to cultural differences, and/or been (vii) limited to lameness surveys of children in mostly low-to-lower middle income countries.[9] Cases of polio may also be missed as those affected may not attend hospital due to quarantine requirements. Worldwide, only 11% of those paralyzed by polio are thought to be captured by national surveillance systems.[10] Hence, regional prevalence estimates are often crude and

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3 fragmentary. This systematic literature review aims to synthesise current knowledge on the  
4 prevalence of polio worldwide using all available population-based prevalence studies.  
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## 8 9 **METHODS**

10 This review is reported according to the PRISMA Statement.

### 11 12 **Search strategy**

13 We searched Medline, CINAHL, Psychology and behavioral sciences, ProQuest, Scopus, and  
14 Web of Science from inception to May 2016 for relevant studies. A search strategy was  
15 developed for Medline using ‘post-polio syndrome’, ‘poliovirus’, ‘polio’, ‘postpolio’,  
16 ‘poliomyelitis’, ‘postpoliomyelitis’, ‘PPMA’, ‘PPMD’, ‘LEOP’, or ‘late effects of polio’ and  
17 ‘epidemiol’, ‘rate’, ‘proportion’, or ‘prevalence\*’, and was then adapted for other database  
18 searches. Hand searching of included articles was also undertaken.  
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### 28 29 **Inclusion/exclusion criteria**

30 Inclusion criteria were: peer reviewed; written in English; reporting of prevalence of polio;  
31 use of a population-based, epidemiology approach with a defined denominator; and some  
32 form of diagnostic or clinical verification of polio. Only those studies reporting on cases  
33 ascertained from a general population sample (i.e., not restricted by gender or ethnicity) were  
34 included to enable comparison between populations, and with other conditions, and to enhance  
35 representativeness of the findings. Studies in which any prevalence data were unable to be  
36 extracted or calculated, or studies reporting incidence data only were excluded. Duplicate  
37 publications reporting on the same research data were also removed.  
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### 48 49 **Quality appraisal**

50 Each study was assessed for methodological quality and risk of bias using a 10-item  
51 assessment tool (external (4 items) and internal (6 items) validity) specifically designed for  
52 population-based prevalence studies. Further, a summary assessment evaluates the overall risk  
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3 of study bias based on the 10 items.[1] A summary assessment deeming a study to be at low  
4 risk of bias suggests that ‘further research is very unlikely to change our confidence in the  
5 estimate’. A moderate risk of bias rating suggests that ‘further research is likely to have an  
6 important impact on our confidence in the estimate and may change the estimate’. The  
7 limitations of studies considered to be at high risk of bias suggest that ‘further research is  
8 very likely to have an important impact on our confidence in the estimate and is likely to  
9 change the estimate’. For this review, a study was considered to have a high risk of bias if the  
10 target population was not closely representative of the national population, if there was no use  
11 of random selection, and if the study had a more than a minimal risk of non-response bias.  
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### 22 **Data extraction and synthesis**

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24 Two authors (KJ, SB) independently reviewed abstracts for possible inclusion. In cases of  
25 non-consensus, a third independent review was obtained from a third author (VF). Any on-  
26 going discrepancies were resolved via discussion. In cases of incomprehensive study  
27 methodology, authors were approached to determine a study's potential inclusion. Where  
28 possible, copies of full articles were obtained for studies meeting the inclusion criteria.  
29 Reviewers extracted standard information per study on study characteristics, target  
30 population, research design, and verification of polio diagnosis. Only those studies  
31 considered to be at low to moderate risk of bias were included in the calculation of  
32 prevalence estimates. An average prevalence of polio is reported for each study as the  
33 number of cases per 100,000 people of all ages or a particular age range, depending on the  
34 data available. Rates were checked for accuracy where possible, depending on the data  
35 provided. Due to a lack of availability of standardized rates, prevalence rates are reported as  
36 crude estimate (i.e. unadjusted rates). Studies reporting adjusted values only have not been  
37 included the average prevalence calculation. In instances where a range of prevalence rates  
38 have been reported in a study and no overall rate reported (e.g. across ethnic groups or  
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3 different geographical regions or years), we have used the average of this range for the  
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5 purposes of calculating an overall average prevalence. The research protocol was not subject  
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7 to ethical approval as no such approval was required according to local regulations.  
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## 10 11 **RESULTS**

### 12 13 **Included studies**

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15 An overview of the study selection process is outlined in Figure 1. The initial search yielded  
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17 1,239 citations. Following scanning of the titles for appropriateness for inclusion, those not  
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19 meeting criteria and duplicate citations were removed (973). Where available, the abstracts of  
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21 the remaining 266 potentially relevant titles identified across all sources (EBSCO n = 25;  
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23 ProQuest n = 17; Scopus n = 88; Web of Science n = 136) were obtained. Following the  
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25 availability and review of 206 abstracts, 117 full articles were independently evaluated for  
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27 inclusion by two reviewers (SB, KJ). This process led to the elimination of 86 studies that did  
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29 not meet the required inclusion criteria. The remaining 31 articles met inclusion criteria and  
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31 were included in the review.  
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35 [INSERT FIGURE 1 ABOUT HERE]  
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39 The 31 eligible population-based studies reported data from 14 different countries.  
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41 Data on polio prevalence in low-to-lower middle income countries were reported in 28 (90%)  
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43 studies in 11 countries: India (14 studies); Nigeria (2 studies); Ethiopia (4 studies), and one  
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45 study in each of the following locations: Indonesia, Ghana, Bangladesh, Niger, Cameroon,  
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47 Sudan, Yemen, and Papua New Guinea. Population-based data on polio prevalence in upper-  
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49 middle to high-income countries were available from 3 studies in 3 countries: Japan; Sweden;  
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51 and Brazil.  
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### Study characteristics

Table 1 provides a summary of the characteristics of those articles included in the review, including details of polio verification, population status, study design, and risk of bias. The included studies reported on polio prevalence data collected between 1974 and 2004. In terms of methodologies implemented across the included studies, diagnosis of polio was verified in 28 (90%) cases via the use of clinical investigations (i.e., examination by a physician or similar). Two of these studies also used laboratory investigations (i.e., virological confirmation) to confirm a history of polio. Twenty-nine (94%) of studies presented data collected by lameness surveys. These included surveys of schools (5), villages (1), families (1), house-to-house surveys (16), or a mixture there of (4), and postal questionnaires (1). One lameness survey examined a national population. The remaining two studies (6%) used multiple sources of case ascertainment. Studies most commonly examined urban/rural or semi-rural populations (12), with 8 eight studies limited to rural populations. Only one study reported a specific focus on an urban population. Of the 31 studies included, 26 (84%) presented data based on children and young persons aged <20 years. In terms of risk of bias, the majority of studies (77%) were at moderate (14 studies, 45%) to high (10 studies, 32%) risk of bias (Figure 2). Seven studies (23%) were considered to be at low risk of bias.

[INSERT TABLE 1 ABOUT HERE]

[INSERT FIGURE 2 ABOUT HERE]

### Polio prevalence

In the general population, crude average prevalence in across all included studies ranged from 15 per 100,000 in Nigeria to 1,733 in Ajmer City, India (Figure 3). Among all low-to-lower middle income countries, crude rates of polio prevalence ranged from 15 per 100,000 (Igbo-

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3 Ora, Nigeria) to 1,733 per 100,000 (Ajmer City, Rajasthan, India). Among all high-income  
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5 counties, crude rates of polio prevalence ranged from 24 (PPS) per 100,000 (Japan) to 380  
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7 per 100,000 (Brazil). For those studies considered to be at low to moderate risk of bias,  
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9 prevalence estimates ranged from 92 in Sweden to 730 in Ethiopia. The total combined  
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11 prevalence of polio for those studies at low to moderate risk of bias ranged from 165 (high-  
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13 income countries) to 425 (low-to-lower middle income countries) per 100,000 person years.  
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17 [INSERT FIGURE 3 ABOUT HERE]  
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## 20 21 **DISCUSSION**

22  
23 This study reviews all the available data from population-based polio prevalence studies.  
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25 Findings reveal significant discrepancies in average crude unadjusted prevalence rates, both  
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27 between and within countries. Across all included studies and within low-to-lower middle  
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29 income countries, prevalence rates ranged from 15 per 100,000 person-years in Igbo-Ora  
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31 Nigeria to 1,733 in Ajmer City, India. Within high income countries, rates ranged from 24 in  
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33 Japan to 380 in Brazil. Variations in prevalence between studies could be due to  
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35 methodological variations and shortcomings.  
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39 Prevalence studies to date have predominantly used lameness surveys and varying  
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41 clinical examinations or definitions to confirm a history of polio. Both of these approaches  
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43 are problematic. Lameness surveys are limited in that they only capture those individuals  
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45 with residual physical disabilities. Cases of people living with the effects of polio may be  
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47 missed in the absence of any physical abnormality. There has also been a predominant focus  
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49 on surveys of children living with the physical consequences of polio. Findings from such  
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51 surveys are further limited by the considerable variation in the range of age groups that have  
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53 been surveyed (i.e., 5-15 years, 0-6 months). Lameness surveys limited to the study of  
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55 children with physical ailments are likely to represent an underestimate of the true prevalence  
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3 of those living post-polio. Few studies examined the prevalence of polio across all age  
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5 groups, nor have samples that are representative of the respective national population been  
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7 studied to inform appropriate gender, age and ethnicity estimates.  
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10 Worldwide variations in prevalence ratings could also be attributed to the diversity  
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12 (and at times a lack of clarity) of applied case definitions and assessment processes to  
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14 determine a history of polio. For example, while the majority of studies used lameness  
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16 surveys, some studies were limited to the examination of lower extremity disabilities. Other  
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18 studies included the examination of upper extremity disabilities in their efforts to identify  
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20 those affected by polio. Three studies were limited to the examination of post-polio  
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22 syndrome. Such inconsistencies are of concern given incomplete case ascertainment or  
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24 disease misclassification can significantly skew the reported prevalence. Even assuming that  
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26 all cases were ascertained in a given study, data would still omit those survivors of polio who  
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28 are free from any observable, physical ailments. The estimates provided should be considered  
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30 to be an underestimation of the prevalence of polio worldwide. Our findings suggest the  
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32 average crude worldwide prevalence of 295/100,000 person years. However, given estimates  
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34 that 10 to 40% of polio survivors are free from permanent physical disability[4], it is worth  
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36 noting that the actual worldwide prevalence may be higher given the predominant focus to  
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38 date on children and lame populations.  
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42 Furthermore, the majority of research to date has been conducted within low-to-lower  
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44 middle income countries. Few studies have examined the prevalence of polio survivors in  
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46 high income countries, including those declared to be polio-free. For example, no prevalence  
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48 population-based studies were found based on data from Australasia, United Kingdom, or the  
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50 United States of America. Hence, there is currently little suitable evidence available to inform  
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52 the calculation of prevalence estimates for polio in high income countries.  
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3 This review has provided an overview of the worldwide prevalence of polio informed  
4 by population-based studies to date. While all efforts were undertaken to identify and access  
5 all articles relevant to the review, it is important to acknowledge the likelihood that some  
6 studies (i.e., unpublished or inaccessible studies) were not identified by the search strategy  
7 and therefore excluded. Alongside methodological concerns already discussed above, the  
8 international prevalence estimates from this review should be considered to be a likely  
9 underestimate of the true prevalence on an international scale. Well-designed epidemiological  
10 studies are now required to more accurately determine the prevalence of polio.  
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20 Future epidemiological polio studies must be designed to reduce bias noted in this  
21 review, including the use of random or cluster sampling, the examination of populations that  
22 are representative of the national population where possible, and the application of clear case  
23 definitions and diagnosis. In addition to recommending that future studies to adhere to the  
24 Strengthening the Reporting of Observational Studies in Epidemiology (STROBE)  
25 Statement[42], we offer specific recommendations for the pursuit of future studies examining  
26 prevalence of polio in Table 2.  
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35 [INSERT TABLE 2 ABOUT HERE]  
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Table 2. Suggested recommendations for future epidemiological studies of polio.

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**External validity**

Examine nationally representative populations to enhance the generalizability of findings

Further prevalence studies are required in high-income countries

Examine all age groups (i.e., adults and children)

Estimate prevalence by age, sex, residency (urban/rural) and ethnic-specific rates

Undertake a census OR use some form of random selection (i.e., cluster sampling)

Extending findings of lameness surveys by also capturing lame-free cases of polio (i.e., using multiple sources of case ascertainment including review of medical records)

Use an established risk of bias tool specifically designed for use in population-based studies

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**Internal validity**

Standard case definition and clinical evaluation

Describe any efforts to address potential sources of bias

Appropriate numerator (s) and denominator (s)

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**Conclusions**

In conclusion, the review reported prevalence of polio worldwide from all identified studies.

The majority of research to date has been limited to the examination of children and adolescents in low to lower middle income countries (predominantly India) who reside in geographical regions that are not representative of the national population (in terms of age,

sex, ethnic distributions, for example). Further research of polio prevalence is required using a population-based approach, examining nationally representative samples of all ages, particularly in high income countries, including those declared to be polio-free. Such efforts will reduce risks for sampling and measurement bias and improve capacity to provide reliable and more robust worldwide prevalence estimates.

**Competing Interests** None declared.

**Disclosure of conflict of interest** There are no conflicts of interest.

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### Legends

Figure 1. Flow diagram of included/excluded studies.

Figure 2. Risk of bias summary of included population-based studies in reverse chronological order.

Figure 3. Polio prevalence by country income level.

#### *What is already known on this subject?*

The World Health Assembly commitment to the worldwide eradication of polio has achieved great success in the Americas, Europe and the Western Pacific. Yet, elimination of new polio cases is not complete. While estimates suggest that up to 20 million people live with the disabling consequences of polio, no accurate prevalence rates are available.

#### *What this study adds?*

This is the first study to systematically review studies examining the worldwide prevalence of polio. Our findings show that studies to date, primarily based on lameness surveys, demonstrate significant variability in the prevalence of polio across low to high income

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3 countries. Few studies have examined nationally representative samples thus limiting the  
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5 reliability of current worldwide prevalence estimates.  
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10 Contributorship statement

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12 GJ conceived on the study. SB conducted searches and extracted the data. KJ and SB  
13  
14 compiled and analyzed data. VF, AT and GJ advised throughout the study. KJ prepared the  
15  
16 initial draft of the manuscript. All authors provided conceptual input and critical review of  
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18 drafts.  
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22 Data sharing statement

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25 No additional data available.  
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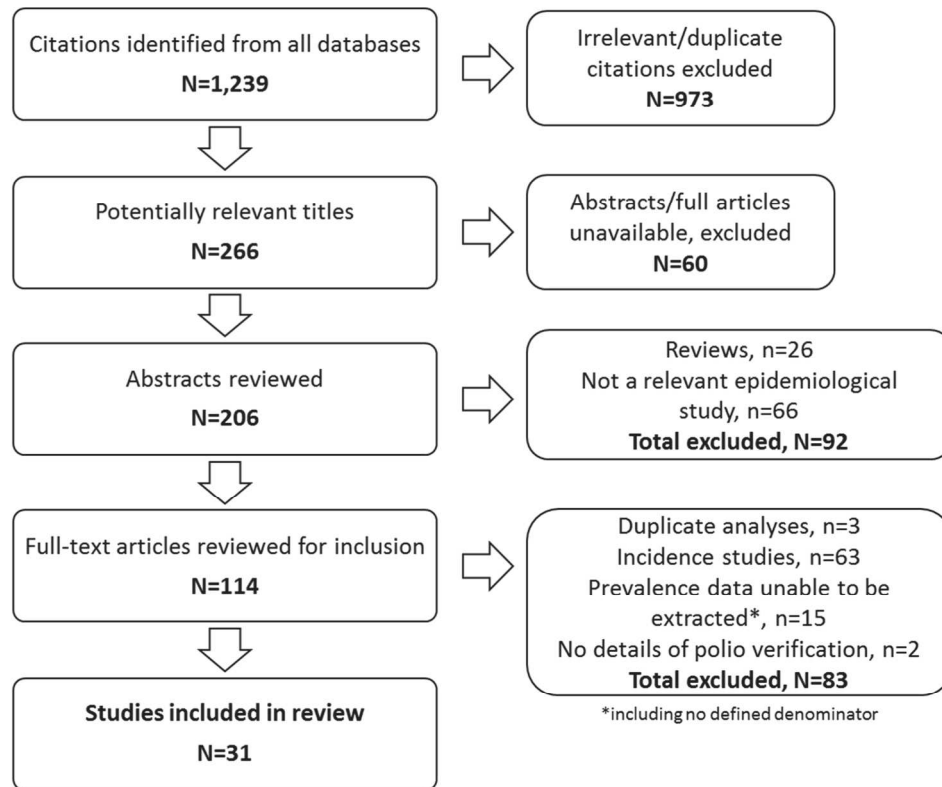


Figure 1. Flow diagram of included/excluded studies.

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	Target population close representation of national	Sampling frame true representation of target population	Random selection used	Minimal risk of non-response bias	Data collected from subjects (not a proxy)	Acceptable case definition	Reliable and validity measurement of parameter of	Consistent mode of data collection	Appropriate prevalence period	Appropriate numerators and denominators	Overall risk
Takemura 2004	X	✓	✓	X	✓	✓	X	✓	✓	✓	Moderate
Tessema 2001	X	✓	✓	X	✓	✓	✓	✓	✓	✓	Low
Anand 1998	X	X	X	X	✓	✓	✓	✓	✓	✓	High
Srinivasa 1997	X	✓	X	X	✓	✓	✓	✓	✓	✓	Moderate
Ahlstrom 1993	✓	✓	✓	X	X	✓	✓	✓	✓	✓	Low
Soudarssane 1993	X	✓	✓	X	✓	✓	✓	✓	✓	✓	Low
Kumar 1991	X	X	X	X	X	X	✓	✓	✓	✓	High
Balraj 1990	X	✓	✓	X	✓	✓	X	✓	✓	✓	Moderate
Mehra 1990	X	✓	X	X	✓	✓	✓	✓	✓	✓	Moderate
Parakoyi 1990	X	✓	✓	X	✓	✓	✓	✓	✓	✓	Low
Tekle-Haimanot 1990	X	✓	✓	✓	✓	X	X	✓	✓	✓	Moderate
Khajuria 1989	X	X	X	X	X	✓	X	X	✓	✓	High
Maru 1988	X	✓	✓	X	✓	✓	✓	✓	✓	✓	Low
Osuntokun 1987	X	✓	✓	X	✓	X	X	✓	✓	✓	Moderate
Tidke 1986	X	X	X	X	✓	✓	✓	✓	✓	✓	High
Broca 1985	X	✓	✓	X	✓	✓	X	✓	✓	✓	Moderate
Basu 1984	✓	✓	✓	X	✓	X	✓	✓	✓	✓	Low
Olive 1984	X	✓	✓	X	✓	X	✓	✓	✓	✓	Moderate
Srilatha 1984	X	✓	X	X	✓	X	✓	✓	✓	✓	High
Hajar 1983	✓	✓	✓	X	✓	✓	✓	✓	✓	✓	Low
Heymann 1983	X	✓	✓	X	✓	✓	✓	X	✓	✓	Moderate
Sabin 1983	X	✓	X	X	✓	✓	✓	✓	✓	✓	Moderate
Dekker 1982	X	X	X	X	✓	X	X	✓	✓	✓	High
Hall 1982	X	✓	X	X	X	✓	X	X	✓	✓	High
Rotti 1982	X	✓	X	X	✓	✓	✓	✓	✓	✓	Moderate
Thuriaux 1982	X	✓	✓	X	✓	✓	X	✓	✓	✓	Moderate
Snyder 1981	X	✓	X	X	✓	✓	✓	✓	✓	✓	Moderate
Ulfah 1981	X	X	X	X	✓	✓	✓	X	✓	✓	High
Nicholas 1977	X	X	X	X	X	✓	✓	X	✓	✓	High
Hardas 1974	X	✓	✓	X	✓	X	✓	✓	✓	✓	Moderate
Jhala 1973	X	X	X	X	✓	✓	✓	✓	✓	✓	High

Figure 2. Risk of bias summary of included population-based studies in reverse chronological order.

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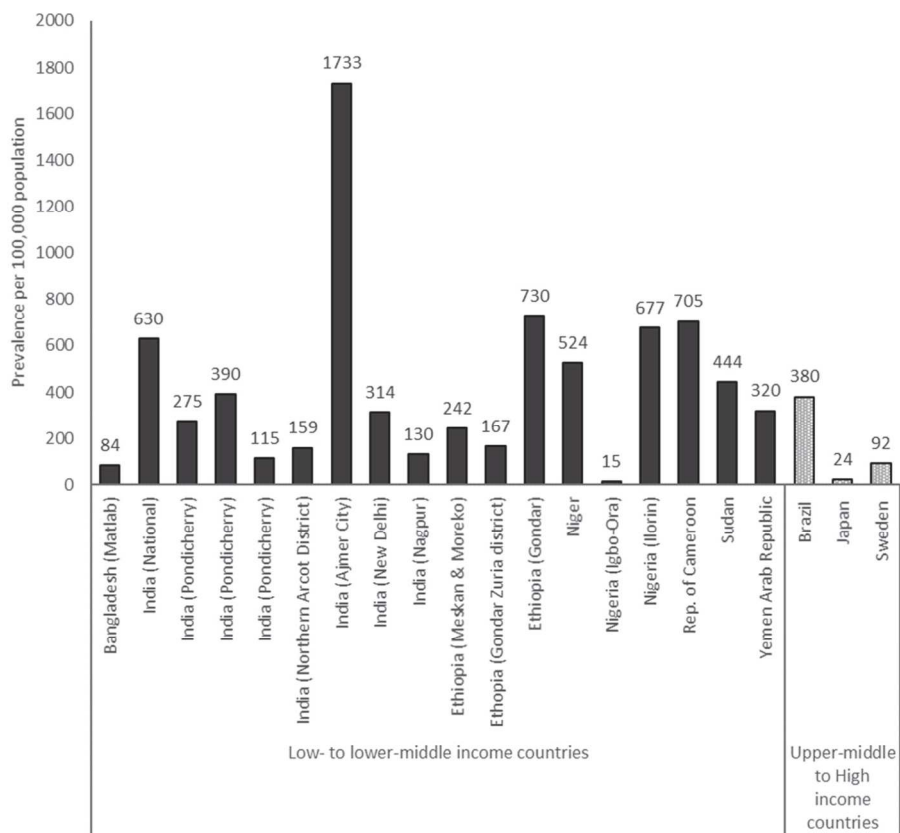


Figure 3. Polio prevalence by country income level

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Table 1. Details of included population-based studies in reverse chronological order.

First Author	Year	Country (Region)	Population status	Population base	Study design / Date	Full article	Verification of diagnosis	Risk rating
Takemura(11)	2004	Japan (Kitakyushu)	Registry of 13,000 physically disabled persons	342	Lameness survey (postal questionnaire)	Yes	Clinical investigation	Moderate
Tessema(12)	2001	Ethiopia* (Gondar Zuria district)	General population (urban/rural)	12,000 children aged 1-15 years	Lameness survey (house-to-house) Jul-Aug 1993	Yes	Clinical investigation	Low
Anand(13)	1998	India* (Ballabgarh, Haryana)	General population (rural)	28,464 children aged <15 years	Lameness survey (house-to-house) Dec 1996	Yes	Clinical and laboratory investigation	High
Srinivasa(14)	1997	India* (Pondicherry)	General population (urban/rural)	Approx. 11,000 children aged <60 months	Lameness survey (house-to-house) Jan-Feb of each year between 1989-91	Yes	Clinical investigation	Moderate
Ahlstrom(15)	1993	Sweden (Orebro)	General population (urban/rural)	269,341	Multiple sources (hospital, outpatient, institutional, insurance, medical records) 01 Jan 1988	Yes	Search of clinical records	Low
Soudarssane(16)	1993	India* (Pondicherry)	General population	47,960 children aged 0-6 years	Lameness survey (house-to-house) Apr-Jul 1989	Yes	Clinical investigation	Low
Kumar(17)	1991	India* (Ambala, Haryana State)	General population	15,761 children aged <15 years	Lameness survey (house-to-house) Jan 1989	Yes	Clinical investigation	High
Balraj(18)	1990	India* (Northern)	General population	42,045 children aged <5 years	Lameness survey (house-to-house)	Yes	Clinical investigation	Moderate

<b>First Author</b>	<b>Year</b>	<b>Country (Region)</b>	<b>Population status</b>	<b>Population base</b>	<b>Study design / Date</b>	<b>Full article</b>	<b>Verification of diagnosis</b>	<b>Risk rating</b>
		Arcot District, Tamil Nadu State)	(rural)		Feb-Oct 1988			
Mehra(19)	1990	India* (New Delhi)	General population (urban/rural)	7,318 children aged 5-15 years	Lameness survey (house-to-house) Jun-Aug 1986	Yes	Clinical investigation	Moderate
Parakoyi(20)	1990	Nigeria* (Ilorin, Kwara State)	General population (urban/rural)	4,576 children aged 5-9 years	Lameness survey (house-to-house) March 1988	Yes	Clinical investigation	Low
Tekle-Haimanot(21)	1990	Ethiopia* (Meskan & Moreko subdistricts)	General population (rural)	60,820 adults	Lameness survey (house-to-house) 1986-1988	Yes	Clinical investigation	Moderate
Khajuria(22)	1989	India* (Haryana)	General population (rural)	37,851 children aged 1-11 years	Lameness survey (village) 1985	Yes	Clinical investigation	High
Dekker(23)	1988	Ethiopia* (Addis Ababa)	General population	256,092 children aged <20 years	Lameness survey (school) Unknown date	Yes	Clinical investigation	High
Maru(24)	1988	Ethiopia* (Gondar)	General population (urban/rural)	17,941 children aged 5-9 years	Lameness survey (house-to-house) Feb-Jul 1983	Yes	Clinical investigation	Low
Osuntokun(25)	1987	Nigeria* (Igbo-Ora)	General population	18,954	Lameness survey (house-to-house) Unknown date	Yes	Clinical investigation	Moderate
Tidke(26)	1986	India* (Bombay)	General population	15,165 children aged <6 years	Lameness survey (house-to-house)	Yes	Clinical investigation	High

First Author	Year	Country (Region)	Population status	Population base	Study design / Date	Full article	Verification of diagnosis	Risk rating
			(slums)		Unknown date			
Broca(27)	1985	India* (Ajmer City, Rajasthan)	General population	6,000 children aged 5-15 years	Lameness survey (house-to-house) Aug 1981-Mar 1982	Yes	Clinical investigation	Moderate
Basu(28)	1984	India (National)*	General population (urban / rural)	715,039 children aged 5-9 years	Lameness survey (national) 1981-1982	Yes	Clinical investigation	Low
Heymann(29)	1983	Rep. of Cameroon* (Yaounde, Bamenda, Eseka)	General population (urban/rural)	37,130 children aged 5-11 years	Multiple sources (hospital and clinical registers, house-to-house and school lameness survey)	Yes	Clinical investigation	Moderate
Olive(30)	1984	Sudan* (Port Sudan and Juba, Khartoum)	General population (urban / semi-rural)	45,499 children aged 5-13 years	Lameness survey (house-to-house / school) July-Sept 1982	Yes	Clinical investigation	Moderate
Srilatha(31)	1984	South India* (North Arcot District, Tamil Nadu)	General population (rural)	14,643 children aged 5-17 years	Lameness survey (school) June-Dec 1979	Yes	Unknown	High
Hajar(32)	1983	Yemen Arab Republic*	General population (urban / rural)	12,443 children aged 5-13 years	Lameness survey (school and community) Nov 1980 – Jan 1981	Yes	Clinical investigation	Low
Sabin(33)	1983	Brazil (Federal District of Brazil)	General population	20,807 children aged 6-7 and 10-11 years	Lameness survey (school) 1980	Yes	Clinical investigation	Moderate

<b>First Author</b>	<b>Year</b>	<b>Country (Region)</b>	<b>Population status</b>	<b>Population base</b>	<b>Study design / Date</b>	<b>Full article</b>	<b>Verification of diagnosis</b>	<b>Risk rating</b>
Hall(34)	1982	Papua New Guinea* (Gulf Province)	General population	3,368 children (ages unspecified)	Lameness survey (school) 1979	Yes	Unknown	High
Rotti(35)	1982	South India* (Pondicherry)	General population (urban)	6,683 children aged 6-15 years	Lameness survey (house-to-house and school) July 1980-Sept 1981	Yes	Clinical investigation	Moderate
Thuriaux(36)	1982	Niger* (Niamey, Kollo, Tillaberry, Gotheye)	General population (rural/urban)	46,772 children aged 5-14 years	Lower limb motor disorders survey (school) Feb-May 1981	Yes	Clinical investigation	Moderate
Snyder(37)	1981	Bangladesh* (Matlab)	General population (rural)	25,000 children aged 5-14 years	Lameness survey (house-to-house) May-Jun 1979	Yes	Clinical investigation	Moderate
Ulfah(38)	1981	Indonesia* (Yogyakarta)	Philanthropic private agency	94,376 children and adolescents aged <20 years	Lameness survey (families)	Yes	Clinical and laboratory investigation	High
Jhala(39)	1979	India* (Patan Taluka, Gujarat)	General population (rural)	57,435 adults	Lameness survey (house-to-house)	Yes	Clinical investigation	High
Nicholas(40)	1977	Ghana* (Danfa)	General population (rural)	13,232 children aged 0-15 years	Lameness survey (school, village)	Yes	Clinical investigation	High
Hardas(41)	1974	India* (Nagpur)	General population (urban/rural)	36,826 children aged <12 years	Lameness survey (house-to-house)	Yes	Clinical investigation	Moderate
Low to lower middle income countries – Average prevalence <sup>+</sup> 425								

<b>First Author</b>	<b>Year</b>	<b>Country (Region)</b>	<b>Population status</b>	<b>Population base</b>	<b>Study design / Date</b>	<b>Full article</b>	<b>Verification of diagnosis</b>	<b>Risk rating</b>
Upper-middle to high income countries – Average prevalence†							165	
<b>International – Total average prevalence</b>							<b>295</b>	

\*Low-to-lower middle income countries.

PPS = Post-polio syndrome.

† Based on 18 studies with low to moderate risk of bias.

‡ Based on 3 studies with low to moderate risk of bias.



# PRISMA 2009 Checklist

Section/topic	#	Checklist item	Reported on page #
<b>TITLE</b>			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	1
<b>ABSTRACT</b>			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	3
<b>INTRODUCTION</b>			
Rationale	3	Describe the rationale for the review in the context of what is already known.	4
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	4
<b>METHODS</b>			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	n/a
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	5
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	5
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	5
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	5
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	6
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	6
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	5
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	6
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., $I^2$ ) for each meta-analysis.	6



# PRISMA 2009 Checklist

Page 1 of 2

Section/topic	#	Checklist item	Reported on page #
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	6
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	n/a
<b>RESULTS</b>			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	7
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	7
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	8
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	n/a
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	8
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	9
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	n/a
<b>DISCUSSION</b>			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	9
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	10
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	12
<b>FUNDING</b>			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	13

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(6): e1000097. doi:10.1371/journal.pmed1000097

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Page 2 of 2

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# BMJ Open

## A systematic review of the worldwide prevalence of survivors of poliomyelitis reported in 31 studies.

Journal:	<i>BMJ Open</i>
Manuscript ID	bmjopen-2016-015470.R1
Article Type:	Research
Date Submitted by the Author:	23-Feb-2017
Complete List of Authors:	Jones, Kelly; Auckland University of Technology Balalla, Shivanthi; Auckland University of Technology, National Institute for Stroke and Applied Neurosciences Theadom, Alice; Auckland University of Technology, National Institute for Stroke and Applied Neurosciences Jackman, Gordon; Polio NZ Inc. Feigin, Valery; Auckland University of Technology,
<b>Primary Subject Heading</b>:	Epidemiology
Secondary Subject Heading:	Global health
Keywords:	EPIDEMIOLOGY, PUBLIC HEALTH, STATISTICS & RESEARCH METHODS

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3 A systematic review of the worldwide prevalence of survivors of poliomyelitis reported in 31  
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5 studies.  
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## ABSTRACT

**Background** Accurate prevalence figures estimating the number of survivors of poliomyelitis (disease causing acute flaccid paralysis), following poliovirus infection are not available. We aim to undertake a systematic review of all literature concerning the prevalence of survivors of poliomyelitis.

**Methods** Electronic databases were searched from 1900 up to May 2016 for peer-reviewed studies including population-based approach with a defined denominator and some form of diagnostic or clinical verification of polio. Exclusion criteria were any prevalence data were unable to be extracted or calculated and studies reporting on incidence only. The quality of each included study was assessed using an existing tool modified for use in prevalence studies. Average crude prevalence rates were used to calculate worldwide estimates.

**Results** Thirty-one studies met criteria with 90% of studies conducted in low-to-lower middle-income countries. Significant variability in the prevalence of survivors of poliomyelitis was revealed, in low-to-lower-middle-income (15 per 100,000 in Nigeria to 1,733 in India) and upper-middle to high-income countries (24 (Japan) to 380 per 100,000 (Brazil)). The total combined prevalence of survivors of poliomyelitis for those studies at low to moderate risk of bias ranged from 165 (high-income countries) to 425 (low-to-lower middle-income countries) per 100,000 person years. Historical lameness surveys of children predominated, with wide variation in case definition and assessment criteria, and limited relevance to current prevalence given the lack of incidence of poliovirus infection in the ensuing years.

**Conclusions** These results highlight the need for future epidemiological studies of poliomyelitis to examine nationally representative samples, including all ages and more focus on high-income countries. Such efforts will improve capacity to provide reliable and more robust worldwide prevalence estimates.

### Strengths and limitations of this study

- This is the first and largest international systematic review, including 31 studies, of the prevalence of survivors of poliomyelitis.
- The study found significant variability in the reported prevalence of survivors across low to high-income countries.
- There are a lack of studies examining nationally representative samples.
- There are no accurate current data on the prevalence of survivors of poliomyelitis worldwide.

## INTRODUCTION

Poliovirus (polio) is a highly infectious, incurable viral disease caused by a wild or live vaccine-derived virus that remains endemic in Afghanistan, Pakistan and Nigeria [1]. Since the creation of the Global Polio Eradication Initiative in 1988, alongside mass vaccination programs aimed at eradicating polio, the number of new cases has been cut by 99% [2] from 350,000 cases to 74 reported cases in 2015 [3]. Polio, a human enterovirus [4], primarily affects children aged <5 years with infection most commonly spread by the faecal-oral route. Up to 75% of poliovirus infections in children are asymptomatic, while approximately 24% of cases may experience a low grade fever and sore throat [5]. Less than 1% of cases experience viral replication in the central nervous system causing temporary or permanent acute flaccid paralysis (AFP) (known as poliomyelitis).[6] While there is wide variability regarding the impact of poliovirus infection, estimates suggest that 12-20 million individuals are living with the consequences of the disease worldwide.[7] Up to 40% of all survivors of acute poliomyelitis will experience post-poliomyelitis syndrome (PPS) [8], being the delayed appearance of new or worsening disabling neuromuscular symptoms 30-40 years after the original poliomyelitis attack.[9] However, recent accurate prevalence rates estimating the number of survivors of poliomyelitis are not available.[10]

Published international prevalence studies are problematic. These studies have (i) tended to focus on the initial disease and needs in the immediate aftermath, (ii) been inconsistent in the definition of “polio survivor,” with it often unclear whether this refers all those infected or only those sustaining some form of residual disability, (iii) predominantly focused on health status rather than the everyday effects on people’s lives, their needs, and those of their carers, (iv) produced inconsistent findings on long-term outcomes, perhaps due to cultural differences, and/or have been (vii) limited to lameness surveys of children in mostly low-to-lower middle-income countries.[11] Hence, regional prevalence estimates are often crude and

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3 fragmentary. This systematic literature review aims to synthesise current knowledge on the  
4 prevalence of survivors of poliomyelitis worldwide using all available population-based polio  
5 prevalence studies.  
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## 10 11 **METHODS**

12  
13 This review is reported according to the PRISMA Statement.  
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### 15 **Search strategy**

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17 We searched Medline (1946 to May 2016), CINAHL (1937 to May 2016), Psychology and  
18 Behavioral Sciences Collection (1945 to May 2016), ProQuest (1971 to May 2016), Scopus  
19 (1970 to May 2016), and Web of Science (1900 to May 2016) databases from inception to  
20 May 2016 for relevant studies. A search strategy was developed for Medline using ‘post-  
21 polio syndrome’, ‘poliovirus’, ‘polio’, ‘postpolio’, ‘poliomyelitis’, ‘postpoliomyelitis’,  
22 ‘PPMA’, ‘PPMD’, ‘LEOP’, or ‘late effects of polio’ and ‘epidemiol’, ‘rate’, ‘proportion’, or  
23 ‘prevalence\*’, and was then repeated for other database searches. The complete search  
24 strategy is available online (see supplementary Table S1). Hand searching of included  
25 articles was also undertaken.  
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43 Selection Criteria.

### 44 **Inclusion/exclusion criteria**

45 Inclusion criteria were: peer reviewed; written in English; reporting of prevalence of  
46 poliomyelitis survivors; use of a population-based, epidemiology approach with a defined  
47 denominator; and some form of diagnostic or clinical verification of polio. Only those studies  
48 reporting on cases ascertained from a general population sample (i.e., not restricted by gender  
49 or ethnicity) were included to enable comparison between populations, and with other  
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3 conditions, and to enhance representativeness of the findings. Studies in which any prevalence  
4 data were unable to be extracted or calculated, or studies reporting incidence data only were  
5 excluded. Duplicate publications reporting on the same research data were also removed.  
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### 8 9 **Quality appraisal**

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11 Each study was assessed for methodological quality and risk of bias using a 10-item  
12 assessment tool (external (4 items) and internal (6 items) validity) specifically designed for  
13 population-based prevalence studies [12]. Further, a summary assessment evaluates the overall  
14 risk of study bias based on the 10 items [13]. A summary assessment deeming a study to be at  
15 low risk of bias suggests that 'further research is very unlikely to change our confidence in  
16 the estimate'. A moderate risk of bias rating suggests that 'further research is likely to have  
17 an important impact on our confidence in the estimate and may change the estimate'. The  
18 limitations of studies considered to be at high risk of bias suggest that 'further research is  
19 very likely to have an important impact on our confidence in the estimate and is likely to  
20 change the estimate'. For this review, a study was considered to have a high risk of bias if the  
21 target population was not closely representative of the national population, if there was no use  
22 of random selection, and if the study had a more than a minimal risk of non-response bias.  
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### 37 **Data extraction and synthesis**

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39 Two authors (KJ, SB) independently reviewed abstracts for possible inclusion. In cases of  
40 non-consensus, an additional independent review was obtained from a third author (VF). Any  
41 on-going discrepancies were resolved via discussion. In cases of incomprehensive study  
42 methodology, authors were approached to determine a study's potential inclusion. Where  
43 possible, copies of full articles were obtained for studies meeting the inclusion criteria.  
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45 Reviewers extracted standard information per study on study characteristics, target  
46 population, research design, and verification of poliovirus infection. Only those studies  
47 considered to be at low to moderate risk of bias were included in the calculation of  
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3 prevalence estimates. An average prevalence of poliomyelitis is reported for each study as the  
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5 number of cases per 100,000 people of all ages or a particular age range, depending on the  
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7 data available. Rates were checked for accuracy where possible, depending on the data  
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9 provided. Due to a lack of availability of standardized rates, prevalence rates are reported as  
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11 crude estimate (i.e. unadjusted rates). Studies reporting adjusted values only have not been  
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13 included the average prevalence calculation. In instances where a range of prevalence rates  
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15 have been reported in a study and no overall rate reported (e.g. across ethnic groups or  
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17 different geographical regions or years), we have used the average of this range for the  
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19 purposes of calculating an overall average prevalence. The research protocol was not subject  
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21 to ethical approval as no such approval was required according to local regulations.  
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## 26 **RESULTS**

### 27 **Included studies**

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29 Figure 1 presents an overview of the study selection process. The initial search yielded 1,239  
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31 citations. Following scanning of the titles for appropriateness for inclusion, those not meeting  
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33 criteria and duplicate citations were removed (973). Where available, the abstracts of the  
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35 remaining 266 potentially relevant titles identified across all sources (EBSCO n = 25;  
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37 ProQuest n = 17; Scopus n = 88; Web of Science n = 136) were obtained. Following the  
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39 availability and review of 206 abstracts, 117 full articles were independently evaluated for  
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41 inclusion by two reviewers (SB, KJ). This process led to the elimination of 86 studies that did  
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43 not meet the required inclusion criteria. The remaining 31 articles met inclusion criteria and  
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45 were included in the review.  
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50 [INSERT FIGURE 1 ABOUT HERE]  
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3 The 31 eligible population-based studies reported data from 14 different countries.  
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5 Data on polio prevalence in low-to-lower middle-income countries were reported in 28 (90%)  
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7 studies in 11 countries: India (14 studies); Nigeria (2 studies); Ethiopia (4 studies), and one  
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9 study in each of the following locations: Indonesia, Ghana, Bangladesh, Niger, Cameroon,  
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11 Sudan, Yemen, and Papua New Guinea. Population-based data on polio prevalence in upper-  
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13 middle to high-income countries were available from 3 studies in 3 countries: Japan; Sweden;  
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15 and Brazil.  
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### 20 **Study characteristics**

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23 Table 1 provides a summary of the characteristics of those articles included in the review,  
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25 including details of polio verification, population status, study design, and risk of bias. The  
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27 included studies reported on polio prevalence data collected between 1974 and 2004. In terms  
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29 of methodologies implemented across the included studies, poliovirus infection was verified  
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31 in 28 (90%) cases via the use of clinical investigations (i.e., examination by a physician or  
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33 similar). Two of these studies also used laboratory investigations (i.e., virological  
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35 confirmation) to confirm a history of polio. Twenty-nine (94%) of studies presented data  
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37 collected by lameness surveys. These included surveys of schools (5), villages (1), families  
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39 (1), house-to-house surveys (16), or a mixture there of (4), and postal questionnaires (1). One  
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41 lameness survey examined a national population. The remaining two studies (6%) used  
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43 multiple sources of case ascertainment. Studies most commonly examined urban/rural or  
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45 semi-rural populations (12), with 8 eight studies limited to rural populations. Only one study  
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47 reported a specific focus on an urban population. Of the 31 studies included, 26 (84%)  
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49 presented data based on children and young persons aged <20 years. In terms of risk of bias,  
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51 the majority of studies (77%) were at moderate (14 studies, 45%) to high (10 studies, 32%)  
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53 risk of bias (Figure 2). Seven studies (23%) were considered to be at low risk of bias.  
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[INSERT TABLE 1 ABOUT HERE]

[INSERT FIGURE 2 ABOUT HERE]

### **Poliomyelitis prevalence**

In the general population, crude average prevalence across all included studies ranged from 15 per 100,000 in Nigeria to 1,733 in Ajmer City, India (Figure 3). Among all low-to-lower middle-income countries, crude rates of poliomyelitis prevalence ranged from 15 per 100,000 (Igbo-Ora, Nigeria) to 1,733 per 100,000 (Ajmer City, Rajasthan, India). Among all high-income countries, crude rates of polio prevalence ranged from 24 (PPS) per 100,000 (Japan) to 380 per 100,000 (Brazil). For those studies considered to be at low to moderate risk of bias, prevalence estimates ranged from 92 in Sweden to 730 in Ethiopia. The total combined prevalence of polio for those studies at low to moderate risk of bias ranged from 165 (high-income countries) to 425 (low-to-lower middle-income countries) per 100,000 person years.

[INSERT FIGURE 3 ABOUT HERE]

### **DISCUSSION**

This study reviews all the available data from population-based poliomyelitis prevalence studies. Findings reveal significant discrepancies in average crude unadjusted prevalence rates, both between and within countries. Across all included studies and within low-to-lower middle-income countries, prevalence rates ranged from 15 per 100,000 person-years in Igbo-Ora Nigeria to 1,733 in Ajmer City, India. Within high-income countries, rates ranged from 24 in Japan to 380 in Brazil.

Worldwide variations in prevalence ratings could be attributed to the diversity (and at times a lack of clarity) of applied case definitions and assessment processes to determine a

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3 history of poliomyelitis. For example, while the majority of studies used lameness surveys,  
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5 some studies were limited to the examination of lower extremity disabilities. Other studies  
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7 included the examination of upper extremity disabilities in their efforts to identify those  
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9 affected by poliovirus leading to poliomyelitis. Three studies were limited to the examination  
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11 of PPS only. Such inconsistencies are of concern given incomplete case ascertainment or  
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13 disease misclassification can significantly skew the reported prevalence. Even assuming that  
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15 all cases were ascertained in a given study, data would still omit those survivors of  
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17 poliomyelitis who are now free from any observable, physical ailments. However, perhaps  
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19 more problematic are the risks for over-reporting due to the inclusion of cases of non-polio  
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21 AFP.  
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25         Alongside methodological variations and shortcomings discussed above, rather than  
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27 informing estimates of the prevalence of survivors of poliomyelitis worldwide, limitations in  
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29 the literature render this review largely of the historical prevalence of residual AFP that may  
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31 be due to poliomyelitis. AFP is a clinical syndrome with a broad array of possible etiologies  
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33 (i.e., spinal cord compression, trauma, exposure to chemicals, recent illness) that serves as a  
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35 proxy for poliomyelitis [14]. Figures from AFP surveillance surveys, an essential strategy of  
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37 the Global Polio Eradication Initiative [13], suggest that non-polio AFP affects 1-3 cases per  
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39 100,000 children aged <15 years per year [15]. Subsequently lameness surveys, most  
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41 common in this review, risk overstating the prevalence of survivors of polio. Such risks are  
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43 especially high in areas such as Afghanistan, India and Nigeria who have the highest  
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45 annualized non-polio AFP rate compared with the number of poliovirus cases [16].  
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47 Furthermore, few studies examined the prevalence of survivors of poliomyelitis across all age  
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49 groups, nor have samples that are representative of the respective national population been  
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51 studied to inform appropriate gender, age and ethnicity estimates. Findings from lameness  
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3 surveys in the current review also have considerable variation in the range of age groups  
4 surveyed (i.e., 5-15 years, 0-6 years).  
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7 Our findings suggest the average crude worldwide prevalence of 295/100,000 person  
8 years. However, many of the studies included in this review were undertaken in geographical  
9 areas where rates of non-polio AFP are high, the dated nature of studies (many being  
10 published more than 30 years ago) and since aging population, and the 99% reduction in the  
11 more recent incidence of poliovirus infection, it must be noted that the actual worldwide  
12 prevalence is likely much lower.  
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20 This review has provided an overview of studies to date that have endeavored to  
21 examine the prevalence of survivors of poliomyelitis. While all efforts were made to identify  
22 and access all articles relevant to the review, it is important to acknowledge the likelihood  
23 that some studies (i.e., unpublished or inaccessible studies) were not identified by the search  
24 strategy and therefore excluded. Well-designed epidemiological studies are clearly required  
25 to accurately determine the current prevalence of poliomyelitis survivors, living either with or  
26 without AFP.  
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35 Future epidemiological polio studies can reduce bias noted in this review by including  
36 the use of random or cluster sampling, the examination of populations that are representative  
37 of the national population where possible, and the application of clear case definitions and  
38 diagnosis. In addition to recommending that future studies to adhere to the Strengthening the  
39 Reporting of Observational Studies in Epidemiology (STROBE) Statement [17], we offer  
40 specific recommendations for the pursuit of future studies examining prevalence of survivors  
41 of poliomyelitis in Table 2.  
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Table 2. Suggested recommendations for future epidemiological studies of poliomyelitis.

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**External validity**

Examine nationally representative populations to enhance the generalizability of findings

Further prevalence studies are required in high-income countries

Examine all age groups (i.e., adults and children)

Estimate prevalence by age, sex, residency (urban/rural) and ethnic-specific rates

Undertake a census OR use some form of random selection (i.e., cluster sampling)

Extending findings of lameness surveys by also capturing lame-free cases of poliomyelitis (i.e., using multiple sources of case ascertainment including review of medical records)

Use an established risk of bias tool specifically designed for use in population-based studies

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**Internal validity**

Standard case definition and clinical evaluation

Describe any efforts to address potential sources of bias

Appropriate numerator (s) and denominator (s)

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**Conclusions**

In conclusion, this review reported prevalence of poliomyelitis survivors worldwide from all identified studies. The majority of research to date has been limited to the examination of children and adolescents in low to lower middle-income countries (predominantly India) who reside in geographical regions that are not representative of the national population (in terms of age, sex, ethnic distributions, for example) and face high rates of non-polio AFP. Further research of the prevalence of survivors of poliomyelitis required using a population-based

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3 approach, examining nationally representative samples of all ages, particularly in high  
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5 income countries including those declared to be polio-free. Such efforts will reduce risks for  
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7 sampling and measurement bias and improve capacity to provide reliable and more robust  
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9 worldwide prevalence estimates.  
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11  
12 **Competing Interests** None declared.  
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14 **Disclosure of conflict of interest** There are no conflicts of interest.  
15

16 **Funding** This work was funded by The Sir Thomas and Lady Duncan Trust Fund.  
17

### 18 **Legends**

19  
20  
21 Figure 1. Flow diagram of included/excluded studies.  
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23 Figure 2. Risk of bias summary of included population-based studies in reverse chronological  
24 order.  
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26  
27 Figure 3. Poliomyelitis prevalence by country income level.  
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### 31 **Contributorship statement**

32  
33 GJ conceived on the study. SB conducted searches and extracted the data. KJ and SB  
34  
35 compiled and analyzed data. VF, AT and GJ advised throughout the study. KJ prepared the  
36  
37 initial draft of the manuscript. All authors provided conceptual input and critical review of  
38  
39 drafts.  
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### 43 **Data sharing statement**

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46 No additional data available.  
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Table 1. Details of included population-based studies in reverse chronological order.

<b>First Author</b>	<b>Year</b>	<b>Country (Region)</b>	<b>Population status</b>	<b>Population base</b>	<b>Study design / Date</b>	<b>Full article</b>	<b>Verification of diagnosis</b>	<b>Risk rating</b>
Takemura[18]	2004	Japan (Kitakyushu)	Registry of 13,000 physically disabled persons	342	Lameness survey (postal questionnaire)	Yes	Clinical investigation	Moderate
Tessema[19]	2001	Ethiopia* (Gondar Zuria district)	General population (urban/rural)	12,000 children aged 1-15 years	Lameness survey (house-to-house) Jul-Aug 1993	Yes	Clinical investigation	Low
Anand[20]	1998	India* (Ballabgarh, Haryana)	General population (rural)	28,464 children aged <15 years	Lameness survey (house-to-house) Dec 1996	Yes	Clinical and laboratory investigation	High
Srinivasa[21]	1997	India* (Pondicherry)	General population (urban/rural)	Approx. 11,000 children aged <60 months	Lameness survey (house-to-house) Jan-Feb of each year between 1989-91	Yes	Clinical investigation	Moderate
Ahlstrom[22]	1993	Sweden (Orebro)	General population (urban/rural)	269,341	Multiple sources (hospital, outpatient, institutional, insurance, medical records) 01 Jan 1988	Yes	Search of clinical records	Low
Soudarssane[23]	1993	India* (Pondicherry)	General population	47,960 children aged 0-6 years	Lameness survey (house-to-house) Apr-Jul 1989	Yes	Clinical investigation	Low
Kumar[24]	1991	India* (Ambala, Haryana State)	General population	15,761 children aged <15 years	Lameness survey (house-to-house) Jan 1989	Yes	Clinical investigation	High
Balraj[25]	1990	India* (Northern)	General population	42,045 children aged <5 years	Lameness survey (house-to-house)	Yes	Clinical investigation	Moderate

First Author	Year	Country (Region)	Population status	Population base	Study design / Date	Full article	Verification of diagnosis	Risk rating
		Arcot District, Tamil Nadu State)	(rural)		Feb-Oct 1988			
Mehra[26]	1990	India* (New Delhi)	General population (urban/rural)	7,318 children aged 5-15 years	Lameness survey (house-to-house) Jun-Aug 1986	Yes	Clinical investigation	Moderate
Parakoyi[27]	1990	Nigeria* (Ilorin, Kwara State)	General population (urban/rural)	4,576 children aged 5-9 years	Lameness survey (house-to-house) March 1988	Yes	Clinical investigation	Low
Tekle-Haimanot[28]	1990	Ethiopia* (Meskan & Moreko subdistricts)	General population (rural)	60,820 adults	Lameness survey (house-to-house) 1986-1988	Yes	Clinical investigation	Moderate
Khajuria[29]	1989	India* (Haryana)	General population (rural)	37,851 children aged 1-11 years	Lameness survey (village) 1985	Yes	Clinical investigation	High
Dekker[30]	1988	Ethiopia* (Addis Ababa)	General population	256,092 children aged <20 years	Lameness survey (school) Unknown date	Yes	Clinical investigation	High
Maru[31]	1988	Ethiopia* (Gondar)	General population (urban/rural)	17,941 children aged 5-9 years	Lameness survey (house-to-house) Feb-Jul 1983	Yes	Clinical investigation	Low
Osuntokun[32]	1987	Nigeria* (Igbo-Ora)	General population	18,954	Lameness survey (house-to-house) Unknown date	Yes	Clinical investigation	Moderate
Tidke[33]	1986	India* (Bombay)	General population	15,165 children aged <6 years	Lameness survey (house-to-house)	Yes	Clinical investigation	High



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First Author	Year	Country (Region)	Population status	Population base	Study design / Date	Full article	Verification of diagnosis	Risk rating
			(slums)		Unknown date			
Broca[34]	1985	India* (Ajmer City, Rajasthan)	General population	6,000 children aged 5-15 years	Lameness survey (house-to-house) Aug 1981-Mar 1982	Yes	Clinical investigation	Moderate
Basu[35]	1984	India (National)*	General population (urban / rural)	715,039 children aged 5-9 years	Lameness survey (national) 1981-1982	Yes	Clinical investigation	Low
Heymann[36]	1983	Rep. of Cameroon* (Yaounde, Bamenda, Eseka)	General population (urban/rural)	37,130 children aged 5-11 years	Multiple sources (hospital and clinical registers, house-to-house and school lameness survey)	Yes	Clinical investigation	Moderate
Olive[37]	1984	Sudan* (Port Sudan and Juba, Khartoum)	General population (urban / semi-rural)	45,499 children aged 5-13 years	Lameness survey (house-to-house / school) July-Sept 1982	Yes	Clinical investigation	Moderate
Srilatha[38]	1984	South India* (North Arcot District, Tamil Nadu)	General population (rural)	14,643 children aged 5-17 years	Lameness survey (school) June-Dec 1979	Yes	Unknown	High
Hajar[39]	1983	Yemen Arab Republic*	General population (urban / rural)	12,443 children aged 5-13 years	Lameness survey (school and community) Nov 1980 – Jan 1981	Yes	Clinical investigation	Low
Sabin[40]	1983	Brazil (Federal District of Brazil)	General population	20,807 children aged 6-7 and 10-11 years	Lameness survey (school) 1980	Yes	Clinical investigation	Moderate

First Author	Year	Country (Region)	Population status	Population base	Study design / Date	Full article	Verification of diagnosis	Risk rating
Hall[41]	1982	Papua New Guinea* (Gulf Province)	General population	3,368 children (ages unspecified)	Lameness survey (school) 1979	Yes	Unknown	High
Rotti[42]	1982	South India* (Pondicherry)	General population (urban)	6,683 children aged 6-15 years	Lameness survey (house-to-house and school) July 1980-Sept 1981	Yes	Clinical investigation	Moderate
Thuriaux[43]	1982	Niger* (Niamey, Kollo, Tillaberry, Gotheye)	General population (rural/urban)	46,772 children aged 5-14 years	Lower limb motor disorders survey (school) Feb-May 1981	Yes	Clinical investigation	Moderate
Snyder[44]	1981	Bangladesh* (Matlab)	General population (rural)	25,000 children aged 5-14 years	Lameness survey (house-to-house) May-Jun 1979	Yes	Clinical investigation	Moderate
Ulfah[45]	1981	Indonesia* (Yogyakarta)	Philanthropic private agency	94,376 children and adolescents aged <20 years	Lameness survey (families)	Yes	Clinical and laboratory investigation	High
Jhala[46]	1979	India* (Patan Taluka, Gujarat)	General population (rural)	57,435 adults	Lameness survey (house-to-house)	Yes	Clinical investigation	High
Nicholas[47]	1977	Ghana* (Danfa)	General population (rural)	13,232 children aged 0-15 years	Lameness survey (school, village)	Yes	Clinical investigation	High
Hardas[48]	1974	India* (Nagpur)	General population (urban/rural)	36,826 children aged <12 years	Lameness survey (house-to-house)	Yes	Clinical investigation	Moderate
Low to lower middle income countries – Average prevalence <sup>+</sup> 425								

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First Author	Year	Country (Region)	Population status	Population base	Study design / Date	Full article	Verification of diagnosis	Risk rating
Upper-middle to high income countries – Average prevalence†								165
<b>International – Total average prevalence</b>								<b>295</b>

\*Low to lower-middle income countries.  
 PPS = Post-polio syndrome.  
 † Based on 18 studies with low to moderate risk of bias.  
 ‡Based on 3 studies with low to moderate risk of bias.

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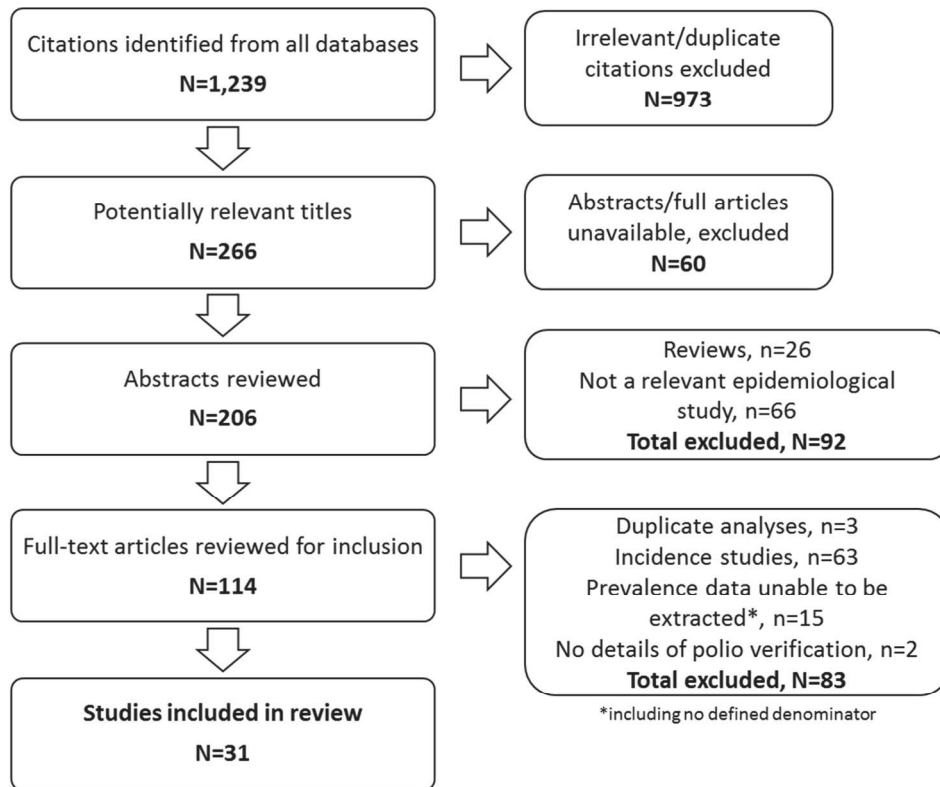


Figure 1. Flow diagram of included/excluded studies.

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	Target population close representation of national	Sampling frame true representation of target population	Random selection used	Minimal risk of non-response bias	Data collected from subjects (not a proxy)	Acceptable case definition	Reliable and validity measurement of parameter of	Consistent mode of data collection	Appropriate prevalence period	Appropriate numerators and denominators	Overall risk
Takemura 2004	X	✓	✓	X	✓	✓	X	✓	✓	✓	Moderate
Tessema 2001	X	✓	✓	X	✓	✓	✓	✓	✓	✓	Low
Anand 1998	X	X	X	X	✓	✓	✓	✓	✓	✓	High
Srinivasa 1997	X	✓	X	X	✓	✓	✓	✓	✓	✓	Moderate
Ahlstrom 1993	✓	✓	✓	X	X	✓	✓	✓	✓	✓	Low
Soudarssane 1993	X	✓	✓	X	✓	✓	✓	✓	✓	✓	Low
Kumar 1991	X	X	X	X	X	X	✓	✓	✓	✓	High
Balraj 1990	X	✓	✓	X	✓	✓	X	✓	✓	✓	Moderate
Mehra 1990	X	✓	X	X	✓	✓	✓	✓	✓	✓	Moderate
Parakoyi 1990	X	✓	✓	X	✓	✓	✓	✓	✓	✓	Low
Tekle-Haimanot 1990	X	✓	✓	✓	✓	X	X	✓	✓	✓	Moderate
Khajuria 1989	X	X	X	X	X	✓	X	X	✓	✓	High
Maru 1988	X	✓	✓	X	✓	✓	✓	✓	✓	✓	Low
Osuntokun 1987	X	✓	✓	X	✓	X	X	✓	✓	✓	Moderate
Tidke 1986	X	X	X	X	✓	✓	✓	✓	✓	✓	High
Broca 1985	X	✓	✓	X	✓	✓	X	✓	✓	✓	Moderate
Basu 1984	✓	✓	✓	X	✓	X	✓	✓	✓	✓	Low
Olive 1984	X	✓	✓	X	✓	X	✓	✓	✓	✓	Moderate
Srilatha 1984	X	✓	X	X	✓	X	✓	✓	✓	✓	High
Hajar 1983	✓	✓	✓	X	✓	✓	✓	✓	✓	✓	Low
Heymann 1983	X	✓	✓	X	✓	✓	✓	X	✓	✓	Moderate
Sabin 1983	X	✓	X	X	✓	✓	✓	✓	✓	✓	Moderate
Dekker 1982	X	X	X	X	✓	X	X	✓	✓	✓	High
Hall 1982	X	✓	X	X	X	✓	X	X	✓	✓	High
Rotti 1982	X	✓	X	X	✓	✓	✓	✓	✓	✓	Moderate
Thuriaux 1982	X	✓	✓	X	✓	✓	X	✓	✓	✓	Moderate
Snyder 1981	X	✓	X	X	✓	✓	✓	✓	✓	✓	Moderate
Ulfah 1981	X	X	X	X	✓	✓	✓	X	✓	✓	High
Nicholas 1977	X	X	X	X	X	✓	✓	X	✓	✓	High
Hardas 1974	X	✓	✓	X	✓	X	✓	✓	✓	✓	Moderate
Jhala 1973	X	X	X	X	✓	✓	✓	✓	✓	✓	High

Figure 2. Risk of bias summary of included population-based studies in reverse chronological order.

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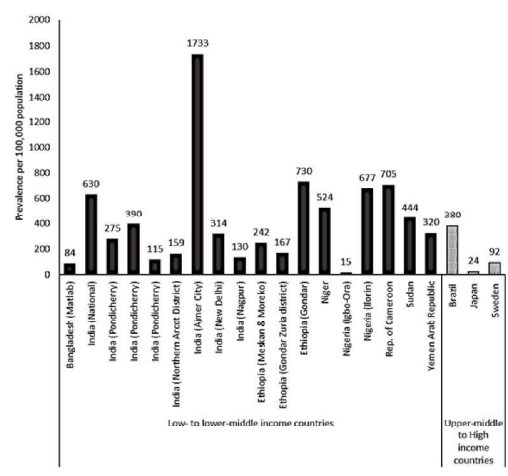


Figure 3. Poliomyelitis prevalence by country income level.

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Supplementary Table 1. Full search criteria for studies assessing prevalence of survivors of polio.

Database (number of hits)	Search Terms
	1.post-polio syndrome
	2.poliovirus
	3.polio
	4.postpolio
PROQUEST (17)	5.poliomyelitis
SCOPUS (88)	6.postpoliomyelitis
WEB OF SCIENCE (136)	7.PPMA
EBSCO (Medline, CINAHL, and Psychology & Behavioral Sciences) (25)	8.PPMD
PROQUEST (17)	9.LEOP
	10.late effects of polio
	11.1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10
	12.epidemol
	13.rate
	14.proportion
	15.prevalence*
	16.11 and 12 or 13 or 14 or 15

The original search date across all databases was 15<sup>th</sup> of May 2016.



# PRISMA 2009 Checklist

Section/topic	#	Checklist item	Reported on page #
<b>TITLE</b>			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	1
<b>ABSTRACT</b>			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	3
<b>INTRODUCTION</b>			
Rationale	3	Describe the rationale for the review in the context of what is already known.	4
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	4
<b>METHODS</b>			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	n/a
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	5
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	5
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	5
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	5
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	6
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	6
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	5
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	6
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., $I^2$ ) for each meta-analysis.	6



# PRISMA 2009 Checklist

Page 1 of 2

Section/topic	#	Checklist item	Reported on page #
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	6
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	n/a
<b>RESULTS</b>			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	7
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	7
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	8
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	n/a
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	8
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	9
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	n/a
<b>DISCUSSION</b>			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	9
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	10
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	12
<b>FUNDING</b>			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	13

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(6): e1000097. doi:10.1371/journal.pmed1000097

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