Research

Using the two-source capture–recapture method to estimate the incidence of acute flaccid paralysis in Victoria, Australia

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Objective To estimate the incidence and the completeness of ascertainment of acute flaccid paralysis (AFP) in Victoria, Australia, in 1998–2000 and to determine its common causes among children aged under 15 years.

Methods The two-source capture–recapture method was used to estimate the incidence of cases of AFP and to evaluate case ascertainment in the routine surveillance system. The primary and secondary data sources were notifications from this system and inpatient hospital records, respectively.

Findings The routine surveillance system indicated that there were 14 cases and the hospital record review identified 19 additional cases. According to the two-source capture–recapture method, there would have been 40 cases during this period (95% confidence interval (CI) = 29–51), representing an average annual incidence of 1.4 per 100 000 children aged under 15 years (95% CI = 1.1–1.7). Thus case ascertainment based on routine surveillance was estimated to be 35% complete. Guillain–Barré syndrome was the commonest single cause of AFP.

Conclusions Routine surveillance for AFP in Victoria was insensitive. A literature review indicated that the capture–recapture estimates obtained in this study were plausible. The present results help to define a target notification rate for surveillance in settings where poliomyelitis is not endemic.

Keywords Paralysis/diagnosis/epidemiology; Muscle hypotonia/diagnosis/epidemiology; Incidence; Epidemiologic surveillance/methods; Epidemiologic methods; Australia (*source: MeSH, NLM*).

Mots clés Paralysie/diagnostic/épidémiologie; Hypotonie musculaire/diagnostic/épidémiologie; Incidence; Surveillance épidémiologique/méthodes; Méthode épidémiologique; Australie (*source: MeSH, INSERM*).

Palabras clave Parálisis/diagnóstico/epidemiología; Hipotonia muscular/diagnóstico/epidemiología; Incidencia; Vigilancia epidemiológica/métodos; Métodos epidemiológicos; Australia (*fuente: DeCS, BIREME*).

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Introduction

In 1988 the World Health Assembly launched an initiative aimed at eradicating poliomyelitis by 2000. The strategy subsequently adopted by WHO relies on immunization and surveillance. Countries where the disease is endemic seek to interrupt transmission by using oral poliovirus vaccine in a combination of high routine immunization coverage of infants and supplementary immunization of children under 5 years of age, delivered through national and subnational immunization days. Mopping-up campaigns may also be necessary. Countries where poliomyelitis is not endemic rely exclusively on high routine immunization coverage of infants.

All countries are encouraged to conduct surveillance for cases of acute flaccid paralysis (AFP). AFP surveillance is a sensitive marker of poliovirus circulation and, for children under 15 years of age, is the recommended method for the detection of poliomyelitis caused by wild poliovirus. WHO recommends that surveillance systems should be sufficiently sensitive to identify an annual rate of non-poliomyelitis AFP of 1 per 100 000 children aged under 15 years. Each case should be reported and investigated within 48 hours of identification (*1*).

AFP is a working classification used to describe patients presenting with acute onset of paralysis in one or more limbs or acute onset of bulbar paralysis. There are many possible causes, but Australian data suggest that 63–72% of cases are attributable to either Guillain–Barré syndrome or transverse myelitis (*2*). Thorough clinical investigation, adequate virological examination of stool specimens and follow-up at 60 days after onset are required in order to determine the cause of paralysis.

In Australia, active AFP surveillance was introduced in 1995 as a joint initiative of the Australian Paediatric Surveillance Unit and the Commonwealth Department of Health (*2*). This surveillance relies on a network of paediatricians who voluntarily notify cases at the time of presentation. Paediatricians also make monthly reports of rare conditions seen, including cases of AFP. The notified cases are entered on a national register.

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In 1995–99, Australia did not attain the recommended target rate for AFP notifications of 1 per 100 000 children aged under 15 years. In order to satisfy WHO requirements for poliomyelitis-free certification in Australia, retrospective hospital record reviews were conducted in three states and one territory of the country with a view to identifying additional cases (*3*). Australia and, indeed, the entire Western Pacific Region, were certified free of poliomyelitis in 2000 (*4*).

A previous study in the State of Victoria, Australia, in which the two-source capture–recapture method was used, indicated that the average annual incidence of AFP was about 1.7 per 100 000 among children aged under 15 years in 1995–97 (*5*). The current retrospective study was also conducted in Victoria, where the projected population was 4 707 590 in 1999, of whom 948 124 were estimated to be aged under 15 years (*6*). We aimed to assess the completeness of AFP ascertainment in Victoria during 1998–2000, to estimate the incidence of the condition, and to determine its common causes among children aged under 15 years.

Methods

The two-source capture–recapture method was employed to estimate the incidence of AFP cases and to evaluate case ascertainment with respect to data in the national register (*7*). Estimation methods based on capture–recapture arose from census studies on animal populations. They have been employed in epidemiological studies on the incidence and/or prevalence of given diseases or injuries as well as in evaluating the completeness of case ascertainment (*8*). Such methods make it possible to compare two or more independent lists of cases in order to estimate the total number of cases in a given population (*9*).

Data sources

The primary ascertainment source for this study was the national register of AFP cases in Victoria for the period 1998–2000. All notified cases are reviewed by a national poliomyelitis expert committee and classified as non-AFP cases or confirmed AFP cases. When sufficient information is available, AFP cases are further classified as non-poliomyelitis, poliomyelitis-compatible, or poliomyelitis.

The secondary ascertainment source comprised hospital case records identified by a discharge diagnosis consistent with AFP-compatible conditions as defined by WHO. The list of AFP-compatible conditions included approximately 70 of the disease codes in the International Classification of Diseases (ICD-9). Clinical practice suggested that patients aged under 15 years presenting with AFP would probably be referred to either the Royal Children's Hospital or the Monash Medical Centre in Melbourne. Cases treated at these two hospitals were therefore identified and reviewed in 1998–2000. Ethical approval for the study was obtained from both hospitals. Case identification involved the interrogation of computerized hospital discharge records relating to this age group for ICD codes matching the WHO list of AFP-compatible conditions. Patients' medical records were audited in order to identify cases compatible with a presentation of AFP. A medical review of each potential AFP case confirmed which cases were eligible for inclusion in the secondary ascertainment source list.

A database of identifying, demographic, hospitalization and clinical details was then established for all eligible AFP cases indicated by hospital discharge records. This database was compared with the national register in order to link cases common to both sources. The comparison variables included the following identifiers: first two letters of family and given names, date of birth, age, sex, usual residential postcode, diagnosis and notifying doctor or treating hospital. The linking of the two data sources was performed manually. The relatively small number of cases, the ages of the patients, and the number of comparison variables facilitated manual linkage of the data sets and ensured a high level of confidence in the reliability of linking.

Data analysis

The model used in this study compares cases from two sources in order to estimate the total number of cases, N, based on the expression:

$$N = ab/c,$$

where a is the total number of cases ascertained from the primary source, b is the total number ascertained from the secondary source and c is the number of cases common to both sources (see Box 1). A modified formula for small numbers (*10*) was used in the present study:

$$N = [(a+1)(b+1)/(c+1)] - 1.$$

We applied the two-source capture–recapture method to estimate the total number of AFP cases in Victoria during 1998–2000. Estimates were made for the three-year period and for data stratified by year, age group, and diagnosis.

Box 1. **Two-source capture–recapture method for estimating the incidence of acute flaccid paralysis**

		Cases ascertained by secondary data source (review of hospital discharge records)		
		Yes	No	Total
		c	$a-c$	
Cases ascertained by primary source (national AFP case register)	Yes	(cases ascertained by both sources)		a (cases ascertained by primary source)
	No	$b-c$	d (cases not ascertained by either source)	$(b-c)+d$
		Total = b (cases ascertained by secondary source)	$(a-c)+d$	N (estimated total incident cases)

It is assumed that the probability of ascertainment from both sources is equal. This is equivalent to assuming that the odds ratio = 1, i.e. $cd = (a-c)(b-c)$. This equation can be solved for d, the number of cases not ascertained by either source. It can then be shown that $N = ab/c$.

Results

The national register included information on 14 AFP cases resident in Victoria in 1998–2000, equivalent to an annual incidence of 0.5 per 100 000 children aged under 15 years. If ascertainment had been at the recommended WHO target level, a minimum of 28 cases would have been recorded. Patients discharged with AFP-compatible diagnoses identified from hospital information systems led to an audit of 350 medical records. Of these patients, 22 discharged from the Royal Children's Hospital, and seven discharged from the Monash Medical Centre had clinical presentations consistent with AFP.

The primary and secondary sources identified 14 and 29 cases, respectively, and 10 cases that were common to both. Two cases from the primary source (the national AFP register) were not identified from the secondary source (hospital discharge records) but were notified by clinicians at one of the hospitals. We attempted to identify these apparently missing patients in order to review their hospital records. However, an extensive search failed to locate them on hospital computerized patient information systems. The matter was also referred to the clinicians who notified the cases but no further information was obtained.

The two-source capture–recapture estimates of AFP case numbers for the overall data and for data stratified by year of diagnosis, age group, and diagnosis are presented in Table 1. A total of 40 AFP cases was estimated (95% confidence interval (CI)= 29–51). The estimated total number of expected AFP notifications based on stratified analysis was 38 by age (95% CI = 27–49), 39 by year of diagnosis (95% CI= 28–50), and 48 by diagnostic category (95% CI = 38–59).

The average annual incidence of AFP among children aged under 15 years was estimated to be 1.4 per 100 000 (95% CI = 1.1–1.7). The most common diagnoses were Guillain–Barré syndrome (13 cases) and acute disseminated encephalomyelitis (3 cases). Transverse myelitis accounted for two cases, whereas during the preceding 3 years a total of 7 of 28 cases had been attributed to this condition (*5*). On the basis of the stratified analysis, we estimated that in Victoria the combined annual incidence of Guillain–Barré syndrome and transverse myelitis among children aged under 15 years was 0.6 per 100 000.

Discussion

The estimated incidence of AFP of 1.4 per 100 000 children under 15 years of age is in excess of the WHO target of 1 per 100 000 in children aged under 15 years but is consistent with that of 1.7 per 100 000 obtained in a previous study in Victoria, covering the period 1995–97 (*5*). Had the two apparently missing cases been included as common to both source lists, thereby increasing the number of cases common to both sources from 10 to 12, our findings would not have been substantially altered: the estimated total number of AFP cases would have been 36 (95% CI = 30–41) and the incidence would have been 1.3 per 100 000 (95% CI = 1.1–1.5).

The previous study in Victoria estimated that routine surveillance had achieved 20% completeness of ascertainment during 1995–97 (*5*). We expected an increase in AFP ascertainment in Victoria because of improved routine surveillance and case identification by contributing clinicians. The present study confirmed that ascertainment had improved: completeness was estimated to be 35% overall and approximately 40% during 1999 and 2000.

Table 1. **Crude and stratified analysis of AFP ascertainment: cases ascertained and estimated total cases**

	Total from national register (primary source) (*a*)[a]	Total from RCH and MMC[b] (secondary source) (*b*)[a]	Total from both sources (*c*)[a]	Estimated total number of cases *N*[a]	95% confidence interval for *N*	Estimated completeness of ascertainment
Crude analysis	14	29	10	40	29–51	35%
Stratified analyses						
By year						
1998	4	11	3	14	9–18	29%
1999	6	8	3	15	8–22	41%
2000	4	10	4	10	–	40%
	14	29	10	39	28–50	36%
By age (years)						
0–4	5	15	5	15	–	33%
5–9	7	10	4	17	10–23	42%
10–14	2	4	1	6	3–10	31%
	14	29	10	38	27–49	37%
By diagnosis						
Guillain–Barré syndrome and transverse myelitis	8	16	8	16	–	50%
Other	6	13	2	32	10–53	19%
	14	29	10	48	38–59	29%

[a] See Box 1 and formulae in text for explanation of symbols.
[b] RCH = Royal Children's Hospital; MMC = Monash Medical Centre.
[c] CI = confidence interval.

The stratified estimates are included in order to explore the possibility of variable "catchability" in case ascertainment as indicated by the source lists (*11*, *12*). Diagnosis and age may have affected the access of patients to the two specialist paediatric hospitals. Older children may have been treated in hospitals primarily intended for adult patients, while suspected specific conditions in children, such as Guillain–Barré syndrome, may have been referred to the specialist hospitals for further investigation and care. The crude estimate is to be preferred, given the relative consistency of the findings based on stratified and unstratified analyses (*13*).

As from 2000 the Australian Government has contracted the Victorian Infectious Diseases Reference Laboratory to coordinate AFP surveillance throughout the country. On the basis of all cases ascertained by routine surveillance, the incidence of AFP among children aged under 15 years in Australia during 2000 was estimated to be 1.2 per 100 000. A value of 1.1 per 100 000 was obtained if account was taken of only those cases notified with sufficient information for final classification by the national poliomyelitis expert committee (*14*). These rates are consistent with the estimates derived for Victoria in the present study. Comparison with reported AFP incidences in other Australian states also suggests that the estimated values for Victoria are plausible. Thus Queensland and New South Wales reported non-poliomyelitis AFP incidences of 1.5 and 1.4 per 100 000, respectively, in 2001 (National AFP Register, unpublished data, 2002).

Limitations of capture–recapture methods

It should be noted that capture–recapture methods rely on the assumptions that the ascertainment sources are independent, that all cases have an equal probability of inclusion, and that cases are correctly identified, i.e. diagnosed, and linked (*11*). Bias would result if any of these assumptions were invalid. In the present study it is likely that the two sources were independent because the routine surveillance system relied on notification by paediatricians, regardless of the practice setting. The hospital discharge data were largely the responsibility of junior hospital medical and health information staff. If the sources were not independent but positively dependent the true number of cases would be greater than the estimated number (*12*). The consistency of the estimates across the crude and stratified data makes it possible to have confidence in the independence of the two sources (*15*, *16*). There was also consistency between the estimates in the present study and those of the previous study in Victoria (*5*), although the same method was used in both.

The present study was unable to determine whether cases had the same chance of being captured by either of the two sources. We are confident that the two sources recruited cases from the same closed population, although there may have been some movement of patients across the state boundary. We are also confident in the accuracy of our identification of cases common to both sources, given the small numbers and the good discriminative constellation of identifying variables. There is perhaps less certainty regarding the correct identification of cases by diagnosis. The definition of AFP is not rigorous (*17*), and clinicians may assign a definitive diagnosis unrelated to poliomyelitis based on clinical presentation and diagnostic investigations. This may mean that some AFP cases are not notified.

Comparison with AFP rates in other settings

Notwithstanding these issues, the consistency of our estimates with those attained in the previous study in Victoria and with rates achieved by other Australian states and territories supports their plausibility. Similar or higher estimates of the incidence of AFP have been obtained from the routine AFP surveillance system in other countries. In Afghanistan and Pakistan, where poliomyelitis is still endemic, non-poliomyelitis AFP rates of 1.3 and 1.5 per 100 000, respectively, were reported between January 2000 and April 2002 (*18*). In 2002, some countries from each WHO region reported AFP rates above 1 per 100 000 and sometimes above 2 per 100 000, e.g. Belarus, Cambodia, El Salvador, Iraq, Nepal, and Nigeria reported rates of 2.9, 1.4, 2.5, 2.4, 1.2 and 2.5 per 100 000, respectively (*19*).

In developed countries, lower rates of AFP tend to be reported, but the threshold of 1 per 100 000 in children aged under 15 years can be reached or exceeded. In the USA, for example, hospital discharge records and death certificates indicated an AFP rate of 1.5 per 100 000 attributable to the Guillain-Barré syndrome alone over the period 1985–91 (*20*).

An AFP rate of 1 per 100 000 children aged under 15 years was found in a pilot study of patients admitted to adult and paediatric neurological units in Finland between July 1997 and June 1998 (*17*). In the Netherlands the paediatric surveillance system reported an AFP rate of 0.7 per 100 000 children in this age group between October 1992 and December 1994 (*21*). During this period, however, there was an outbreak of poliomyelitis and the surveillance system recorded only 7 of the 18 children affected. There was evidently a problem of incomplete case ascertainment, similar to that demonstrated in the current study.

A review of AFP surveillance rates from other countries supports the plausibility of the estimates obtained in our study. Many countries report AFP rates in excess of the rate we estimated based on the upper limit of the 95% CI. Lower published rates can be explained by incomplete ascertainment. These results can be used to define a target notification rate for future AFP surveillance in Victoria, other Australian states, and developed countries from which poliovirus has been eradicated. Recent instances of imported poliovirus (*22*) and outbreaks attributed to circulating vaccine-derived poliovirus (*23*, *24*) demonstrate the need for continued vigilance and surveillance systems of high quality, even in countries certified as being free of circulating wild poliovirus. ■

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Conflicts of interest: none declared.

Résumé

Utilisation de la méthode de capture-recapture avec deux sources de données pour estimer l'incidence de la paralysie flasque aiguë dans l'Etat de Victoria (Australie)

Objectif Estimer l'incidence de la paralysie flasque aiguë (PFA) et l'exhaustivité de la comptabilisation des cas dans l'Etat de Victoria (Australie) en 1998-2000 et déterminer quelles sont les causes fréquentes de cette affection chez les moins de 15 ans dans cet Etat.

Méthodes La méthode de capture-recapture avec deux sources de données a été utilisée pour estimer l'incidence des cas de PFA et évaluer la comptabilisation des cas par le système de surveilllance de routine. La source primaire de données était constituée par les notifications du système de surveillance et la source secondaire par les dossiers des malades hospitalisés.

Résultats Le système de surveillance de routine a indiqué 14 cas et les dossiers hospitaliers 19 autres cas. Selon la méthode de capture-recapture avec deux sources de données, il y aurait eu 40 cas pendant cette période (intervalle de confiance (IC) à 95 % : 29-51), soit une incidence moyenne annuelle de 1,4 pour 100 000 enfants de moins de 15 ans (IC 95 % : 1,1-1,7). On estime que le taux de comptabilisation des cas par le système de surveillance de routine était de 35 %. Le syndrome de Guillain-Barré était la cause la plus fréquente de PFA.

Conclusion La surveillance de routine de la PFA dans l'Etat de Victoria manquait de sensibilité. Un examen des données publiées a indiqué que les estimations obtenues dans cette étude par capture-recapture étaient plausibles. Les présents résultats aident à définir le taux de notification que doit atteindre un système de surveillance là où la poliomyélite n'est pas endémique.

Resumen

Uso del método de captura–recaptura con dos fuentes para calcular la incidencia de parálisis fláccida aguda en Victoria, Australia

Objetivo Estimar la incidencia y el grado de exhaustividad de la verificación de la parálisis fláccida aguda (PFA) en Victoria (Australia) en 1998–2000 y establecer sus causas comunes entre los menores de 15 años en ese Estado.

Métodos Se utilizó el método de captura-recaptura con dos fuentes para estimar la incidencia de casos de PFA y para evaluar la verificación de casos efectuada en el marco del sistema de vigilancia sistemática. Las fuentes de datos primaria y secundaria fueron las notificaciones efectuadas a partir de ese sistema y las historias clínicas de los pacientes hospitalizados, respectivamente.

Resultados El sistema de vigilancia sistemática reveló 14 casos, y revisando las historias clínicas se identificaron 19 casos más. Según el método de captura-recaptura a partir de dos fuentes, durante el periodo considerado se habrían producido 40 casos (intervalo de confianza del 95% (IC) = 29–51), lo que representa una incidencia anual media de 1,4 por 100 000 niños menores de 15 años (IC95% = 1,1–1,7). Así pues, se deduce que la verificación de casos basada en la vigilancia sistemática fue del 35%. El síndrome de Guillain–Barré fue la causa única más común de PFA.

Conclusión La vigilancia sistemática de la PFA en Victoria carecía de sensibilidad. Una revisión de la literatura mostró que las estimaciones obtenidas en el estudio mediante el sistema de captura–recaptura eran verosímiles. Los presentes resultados pueden ayudar a definir como meta una determinada tasa de notificación para los sistemas de vigilancia en los entornos donde la poliomielitis no es endémica.

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