Quality of reporting on birth defects in birth certificates: case study from a Brazilian reference hospital

Qualidade da notificação de defeitos congênitos nas declarações de nascido vivo: estudo de caso em hospital de referência no Brasil

Daniela Varela Luquetti [1]
Rosalina Jorge Koifman [1]

[1] Escola Nacional de Saúde Pública Sergio Arouca, Fundação Oswaldo Cruz, Rio de Janeiro, Brasil.

Correspondence

D. V. Luquetti
Pós-Graduação em Saúde Pública e Meio-Ambiente, Escola Nacional de Saúde Pública Sergio Arouca, Fundação Oswaldo Cruz. Rua Leopoldo Bulhões 1480, Rio de Janeiro, RJ 21041-210, Brasil. luquetti@hotmail.com

Abstract

The aim of this study was to evaluate the coverage, validity and reliability of Brazil's Information System on Live Births (SINASC) for birth defects in a hospital in the city of Campinas (São Paulo State). The study population consisted of 2,823 newborn infants delivered in 2004 at the Women's Integrated Health Care Center (CAISM). A birth defect registry (ECLAMC) was used as the gold-standard. All birth defect cases reported at CAISM in 2004 (92 cases) were selected from SINASC data files. All 168 birth defect cases from the same city and year registered at ECLAMC were also retrieved. An underreporting of 46.8% was observed for all birth defects, and 36.4% when considering only the major birth defects. The ascertained sensitivity and specificity were, respectively, 54.2% and 99.8%. The reliability of three and four-digit ICD-10 coding for birth defects was 0.77 and 0.55 respectively (kappa statistic). These results suggest that information provided by birth certificates in Campinas still presents limitations when seeking to ascertain accurate estimates of the prevalence of birth defects, hence indicating the need for improvements in the SINASC database to enable it to portray birth defect prevalence at birth in this city.

Congenital Abnormalities; Birth Certificates; Information Systems

Introduction

Birth defects have an important public health impact in developing countries. Worldwide 7.6 million children are born each year with a severe genetic disorder or birth defect, close to 95% of them in the developing world, where 80% of the global population lives, contributing significantly to infant mortality [1].

In Brazil, the proportion of birth defects in infant mortality, previously obscured by infectious diseases, assumed great relevance with the epidemiologic transition. In 2005, chapter XVII of the International Classification of Diseases 10th revision (ICD-10), namely *Congenital Malformations, Deformations and Chromosomal Abnormalities*, represented the third leading cause of infant mortality (16.3%) in the first year of life, making evident a clear rise in the proportion of this particular form of mortality in the last 20 years: 7.1% in 1985-1987 and 11.2% in the period 1995-1997 (Departamento de Informática do SUS; http://www.datasus.gov.br, accessed on 18/Jul/2008).

In the late 1960s, after the thalidomide episode, the first registries for birth defects were created. The primary reason for this was improved surveillance that would prevent the repetition of a similar tragedy [2]. One of the purposes of surveillance is to detect changes in the prevalence of a specific defect or pattern of defects that might indicate the presence of a new causative factor.

However, the information has also been increasingly used for various other reasons, including epidemiologic studies for the etiology of birth defects, studies of their societal impact, census of disabled people for planning medical services and social welfare provision, assisting the development of clinical genetic services for care and prevention of birth defects, and evaluations of the effectiveness of preventive measures such as the enrichment of flour with folic acid, and the rubella vaccination [3].

In Brazil, the Information System on Live Births (SINASC), launched in 1990, has as instrument for data collection, the Birth Certificate, which includes information about maternal, gestation and newborn characteristics. In 1999, a new field named: "*Congenital Malformations and/or Chromosomal Anomaly*" was introduced. Thus, the basis for the observation of the prevalence at birth of birth defects was established by specific type, geographic unity and period, in a population-based surveillance system.

Another information system for birth defects in Brazil is the Latin-American Collaborative Study of Congenital Malformations (ECLAMC), operating since 1967, with a network of 130 hospitals, 36 in Brazil. This system, in the absence of others, serves as the only source of information for the surveillance of this form of morbidity and represents a good quality epidemiological database. Nevertheless, the coverage is only 2% of all Brazilian births and the information generated is hospital-based [4].

The information about pregnancies and newborns generated by SINASC has been the object of several validation studies [5,6,7,8]. Reliability of birth defects data retrieved from birth certificate data in Rio de Janeiro city was recently evaluated comparing the reports of birth defects from birth certificates with maternal and newborn medical records [9]. However, birth defects data still need extensive studies about the quality of the information including validity of data as well as coding reliability in other Brazilian municipalities.

This study evaluated the quality of the SINASC information regarding birth defects in Campinas city (Southeast of Brazil), from the Center for Women's Integrated Health Care (CAISM) at the State University of Campinas (UNICAMP), aiming to evaluate coverage, validity of diagnoses and reliability of birth defects coding. It constitutes the first step of a wider project of evaluation of the Birth Certificate, in hospitals distributed in the Northeast, Southeast and South of Brazil.

Methodology

This hospital-based study was performed using two data sources, SINASC and ECLAMC. Both systems had independent functioning working teams until 2004 and there was no internal practice for sharing information throughout the birth defect diagnostic and registry processes.

SINASC

The field for birth defects is composed of an open-ended question ("*Detected any congenital malformation and/or chromosomal anomaly?*" "*Yes, No or Ignored*") and a field for the description of the birth defect that, at a central level, is coded according to ICD-10. The Birth Certificate can be filled out by any person trained for this end.

ECLAMC

The ECLAMC is a hospital-based study, of active search, using a case-control approach. The next non-malformed baby of the same sex born in the same hospital is selected as a control subject for each case. The same reporting form is used for cases and controls. All major and minor anomalies diagnosed at birth in infants weighting 500g or more are registered according to a manual of procedures. Each defect is coded at the central level using a standard 6-digit code. Information about maternal age, parity, sex, birth weight, and twinning of all births of the hospital are collected monthly.

CAISM has been an ECLAMC participant hospital since 1987. For the present study, the information of this system was considered accurate and, therefore, it was used as a gold-standard for the validation of the information of birth defects present in the Birth Certificate.

The study population consisted of 2,823 newborns, born between January 1st and December 31st 2004 in CAISM. The Birth Certificates of all 92 cases of birth defect among babies born at CAISM during that year were selected from the SINASC database at the Health Secretariat of Campinas. Likewise, all Birth Certificates (paper form) from CAISM were reviewed for the birth defect field. In this search two other cases, not registered in the database, were identified. For the same period, all 168 notifications of newborns with birth defects in the ECLAMC database were selected. These cases were considered the gold-standard for the birth defect prevalence analysis in this hospital.

All newborns of CAISM and Campinas were selected to evaluate the epidemiological profile according to the variables: maternal age, mater-

nal occupation (classified by the Brazilian Occupational Classification of the Ministry of Work), maternal education (in years of study), gestational age (in weeks), twinning, sex, birth weight, Apgar scores and diagnosis of birth defects according to the ICD-10.

Birth defects were stratified into two groups: major and minor using the ECLAMC and Centers for Disease Control and Prevention criteria and, the major defects were further disaggregated into single or multiple. Afterwards, using the same database, the characteristics of the birth defects cases from CAISM and Campinas were described.

For determining validity, identification data for the ECLAMC cases (birth date, maternal name and age, newborn weight and sex) were registered in a standardized instrument and cross-checked with Birth Certificate data. The information regarding the birth defect field, the description of the birth defect and reporting date was collected from the Birth Certificate. Birth defects from ECLAMC forms were recoded to the ICD-10. The validity evaluation was made by sensitivity, specificity, positive predictive value and negative predictive value. For all analysis, 95% confidence intervals were calculated.

For the 77 cases underreported at SINASC the medical records were reviewed to assess the evaluation of the reported diagnoses, the occasion of reporting and where it was recorded in the medical record.

In order to verify the coding reliability of the birth defects diagnoses of the Birth Certificate made at the Health Secretariat of Campinas, recoding was performed by an ECLAMC trained professional. The reliability, between the coders, for three- and four-digits of the ICD-10, was calculated by the percentage of observed level of agreement and Cohen's kappa statistic [10]. For the interpretation of kappa, Landis & Koch proposed ranges of values were thus considered: greater than 0.75, 0.40-0.75, and less than 0.40; representing respectively, excellent, fair to good, and poor agreement beyond chance [11]. The cases identified in the manual search were excluded from the analysis for absence of codification from the Health Secretariat. For all analysis, 95% confidence intervals were calculated.

Comparison of the category variables was performed by the Pearson's χ^2-test or Fisher exact test. Continuous variables were compared using the Student's *t*-test. In all statistical tests, the alpha error was set at 5%.

The information collected was stored in a computerized database and analyzed by means of the SPSS software, version 13.0 (SPSS Inc., Chicago, U.S.A.).

The project was approved by the Ethics in Research Committee at the National School of Public Health, Oswaldo Cruz Foundation.

Results

Of the 19,282 newborns in Campinas in 2004 (residents and non-residents in the municipality), 2,823 (14.6%) were born in CAISM, and from these, 94 (3.3%) cases of birth defects were recorded in SINASC.

The mean maternal age in CAISM was 25.6 years (standard deviation – SD = 6.6) and 26.8 (SD = 6.3) in Campinas (p < 0.001). The mean birth weight in CAISM was 3,019.1g (SD = 706.2) and 3,115.2g (SD = 573.1) in Campinas (p < 0.001).

In CAISM a higher proportion of housewives, mothers with lower educational level, multiple gestations, prematurity, lower Apgar scores in the 1st and 5th minute and birth defects were observed in relation to Campinas, when all live births were analyzed; all these differences were statistically significant (p < 0.001).

For the birth cases from CAISM and Campinas a similar distribution was observed for the variables: maternal age, education level, and occupation, gestational age and Apgar scores in the 1st minute. The type of gestation and Apgar scores in the 5th minute did not show significant statistical differences (Table 1).

The distribution of the characteristics of the 77 underreported cases compared with those reported in SINASC did not show differences, except for low Apgar scores in the 1st minute (p = 0.0451) and in the 5th minute (p = 0.0252) where the reported cases presented lower scores. A higher proportion of prematurity among the reported cases was also observed, but with borderline statistical significance (p = 0.05) (Table 1).

In 2004, 94 (3.3%) birth defect cases were registered in SINASC between the newborns of CAISM, for the same year the ECLAMC registered 168 (5.9%). Among the birth defect cases identified by SINASC three were not registered by ECLAMC. After a careful analysis one case was confirmed, considered then a loss for ECLAMC.

The validation of the 94 cases registered in SINASC revealed a sensitivity of 54.2% (95%CI: 46.6-61.7), specificity of 99.9% (95%CI: 99.7-99.9), positive predictive value of 96.8% (95%CI: 91.6-99.2) and negative predictive value of 97.3% (95%CI: 96.6-97.8) (Table 2).

The evaluation of the SINASC 77 underreported cases revealed that 44 (57.1%) were cases classified as major birth defects, and of these, 37 (84%) were the only birth defects present in the newborn (Figure 1). When only the major birth

Table 1

Newborn characteristics and birth defects cases in Campinas, São Paulo State, Brazil, and the Center for Women's Integrated Health Care (CAISM) by selected variables from the Birth Certificate, 2004.

| Variables | CAISM (n = 2,823) n (%) | Newborns Campinas * (n = 16,459) n (%) | p-value | Newborns (birth defects in Campinas) | | p-value | Newborns (birth defects from CAISM) | | |
				CAISM (n = 94) n (%)	Campinas * (n = 143) n (%)		SINASC (n = 91) n (%)	Underreported (n = 77) n (%)	ECLAMC (n = 168) n (%)
Maternal occupation									
Housewife	2,615 (92.7)	7,618 (46.6)	< 0.001	88 (93.6)	60 (42.3)	< 0.001	85 (93.4)	73 (94.8)	159 (94.1)
Housekeeper	32 (1.1)	602 (3.7)		0 (0.0)	6 (4.2)		0 (0.0)	1 (1.3)	1 (0.6)
Other	170 (6.2)	8,141 (49.8)		6 (6.4)	76 (53.5)		6 (6.6)	3 (3.9)	9 (5.3)
Maternal education (years)									
1-3	17 (0.6)	213 (1.3)	< 0.001	0 (0.0)	2 (1.4)	< 0.001	0 (0.0)	1 (1.3)	1 (0.6)
4-7	332 (11.8)	4,505 (27.9)		14 (15.1)	39 (27.7)		14 (15.6)	12 (15.6)	24 (14.3)
8-11	2,361 (83.8)	8,212 (50.9)		76 (81.7)	68 (48.2)		73 (81.1)	61 (79.2)	136 (81.0)
12+	109 (3.9)	3,202 (19.8)		3 (3.2)	32 (22.7)		3 (3.3)	3 (3.9)	7 (4.2)
Gestational age (weeks)									
< 37	405 (14.5)	1,489 (9.1)	< 0.001	32 (35.2)	31 (21.8)	0.0371	32 (34.1)	16 (21.6)	50 (30.7)
37-41	2,375 (85.2)	14,736 (90.1)		59 (64.8)	111 (78.2)		59 (65.9)	58 (78.4)	113 (69.3)
> 42	6 (0.2)	133 (0.8)		0	0 (0.0)		0 (0.0)	0 (0.0)	0 (0.0)
Type of gestation									
Singleton	2,720 (96.4)	16,059 (97.7)	< 0.001	88 (93.6)	135 (95.1)	0.463	85 (93.4)	73 (94.8)	161 (95.3)
Multiple	103 (3.6)	386 (2.3)		6 (6.4)	7 (4.9)		6 (6.6)	4 (5.2)	8 (4.7)
Sex									
Male	1,467 (52.0)	8,370 (50.9)	0.3069	50 (53.2)	92 (64.3)	0.1256	49 (53.8)	44 (57.1)	93 (55.0)
Female	1,354 (48.0)	8,063 (49.0)		43 (45.7)	51 (35.7)		41 (45.1)	33 (42.9)	75 (44.4)
Intersex	2 (0.1)	26 (0.2)		1 (1.1)	0 (0.0)		1 (1.1)	0 (0.0)	1 (0.6)
Apgar 1									
Low (0-6)	373 (13.3)	1,321 (8.1)	< 0.001	34 (36.6)	31 (21.7)	0.0187	34 (37.8)	18 (23.4)	52 (31.0)
7-10	2,446 (86.7)	15,052 (91.9)		59 (63.4)	112 (78.3)		56 (62.2)	59 (76.6)	116 (69.0)
Apgar 5									
Low (0-6)	55 (2.0)	171 (1.0)	< 0.001	15 (16.3)	13 (9.1)	0.1444	15 (16.9)	4 (5.2)	21 (12.6)
7-10	2,762 (98.0)	16,208 (99.0)		77 (83.7)	130 (90.9)		74 (83.1)	69 (94.8)	146 (87.4)
Birth defect									
Yes	94 (3.3)	143 (0.9)	< 0.001	-	-	-	-	-	-
No	2,724 (96.7)	16,303 (99.1)		-	-	-	-	-	-

SINASC: Information System on Live Births; ECLAMC: Latin-American Collaborative Study of Congenital Malformations.

* Excluded CAISM.

defects registered by ECLAMC were considered, the SINASC sensitivity was altered to 63.6% (95%CI: 54.8-71.8) and the specificity to 99.9% (95%CI: 99.8-100.0). Figure 2 shows the Birth Certificate underreporting by specific birth defect.

All birth defect diagnoses, major and minor, were described in the medical report, and 62.3% of these were diagnosed in the delivery room, at birth. The interval between the description of the diagnosis in the medical report and the day the Birth Certificate was filled was evaluated to de-termine the maximum number of birth defects that could have been reported by the Birth Certificate. It was observed that 77.9% of the under-reported birth defect cases were described in the medical records when the Birth Certificate was filled. From those 26.5% were filled in the first day of life and 47.1% in the second day. For 89% of the reported cases, birth defects were described properly in the birth certificate.

The validity of the birth defect diagnosis was evaluated for all cases detected by both systems. In

Table 2

Comparison of birth defect cases registered by the Latin-American Collaborative Study for Congenital Malformations (ECLAMC) and the Information System on Live Births (SINASC) in newborns. Center for Women's Integrated Health Care (CAISM), Campinas, São Paulo State, Brazil, 2004.

Birth defects registered by SINASC	Birth defects registered by ECLAMC		
	Yes	No	Total
Yes *	91	3	94
No	77	2,652	2,722
Total	168	2,655	2,823

* Two cases were found only in the manual review of birth defect.

Table 3, sensitivity, specificity, and predictive values of selected birth defects are showed. Sensitivity ranged from 28.6% (95%CI: 5.1-617.0), for Down syndrome, to 100% (95%CI: 65.2-100.0) for spina bifida. Specificity was 100% for all birth defects.

The 92 cases coded by the Health Secretariat were considered for the reliability analysis. The observed level of agreement between the birth defect diagnoses coded by SINASC and the re-coding done by the coder trained by ECLAMC revealed, for ICD-10 three and four digits, 79% (95%CI: 71.1-87.6) e 56% (95%CI: 46.4-66.7), respectively. Agreement adjusted for chance (kappa) was 0.77 (95%CI: 0.75-0.79) for three digits, and 0.55 (95%CI: 0.53-0.58) for four digits of the ICD-10.

Discussion

The Birth Certificate is an attractive source of information for the notification of birth defects in Brazil because it is universal, standardized and inexpensive. However, the underreporting observed through the comparison between SINASC and ECLAMC systems, in CAISM, was high (46.8%) and it maintained high levels (36.4%) even when the analysis was restricted to the defects classified as major.

In a study on the occurrence of birth defects in Rio de Janeiro city, using the SINASC database, a birth defects prevalence of 83 per 10,000 live births for the 2000-2004 period was reported [12]. This result also suggests an underreporting for birth defects from birth certificates, particularly when compared to birth defects prevalence at birth (170 per 10,000 live births), as reported in a previous cross-sectional study based on a sample of 9,386 postpartum women from Rio de Janeiro [13].

The underreporting for major birth defects, in studies performed in the USA with the Birth Cer-

tificate, which is similar to the Brazilian, varied from 55% to 88% [14,15,16,17,18,19,20,21]. In our study, even if it was a high percentage it was better than in the USA, although similar to a study made in Birmingham, England, where underreporting was 32% [22].

Studies performed with the North-American Birth Certificate have demonstrated low completeness, a high number of false positive and negative cases and, in the reported cases, birth defects were inadequately described [14,15,16,17,18,19,20,21]. Watkins et al. [21], in a comparative analysis between the Birth Certificate and data from the Metropolitan Atlanta Congenital Defects Program (MACPD), in 1989, observed a sensitivity of 28% and positive predictive value of 77% for birth defects diagnosed at birth. Honein et al. [15] performed an analysis with the same source of data for 1995 and the sensitivity was found to be 11.9%. Hexter et al. [23] in a similar study found 20% of sensitivity and 80% of positive predictive value, when comparing the Birth Certificate with the California Birth Defects Monitoring Program. Olsen et al. [18] reported a sensitivity of 12-13% and 45% of false-positive when comparing the Birth Certificate with the data of the Congenital Malformations Registry (CMR), in New York. It is important to point out that all values were obtained in these studies considering only major birth defects.

In our study, we identified specificity, and positive and negative predictive values higher to those observed by other authors in North-American studies, showing that the cases which SINASC succeeded in collecting did present at least one of the birth defects described in the Birth Certificate. On the other hand, a high percentage of newborns that were not considered as having birth defects in CAISM, in fact did not. For 89% of the reported cases the description was correct, with only 11% of classifying errors, especially

Figure 1

Distribution of birth defect cases registered and underreported in Information System on Live Births (SINASC) according to classification in minor and major (1a), and, among major, single and multiple (1b).
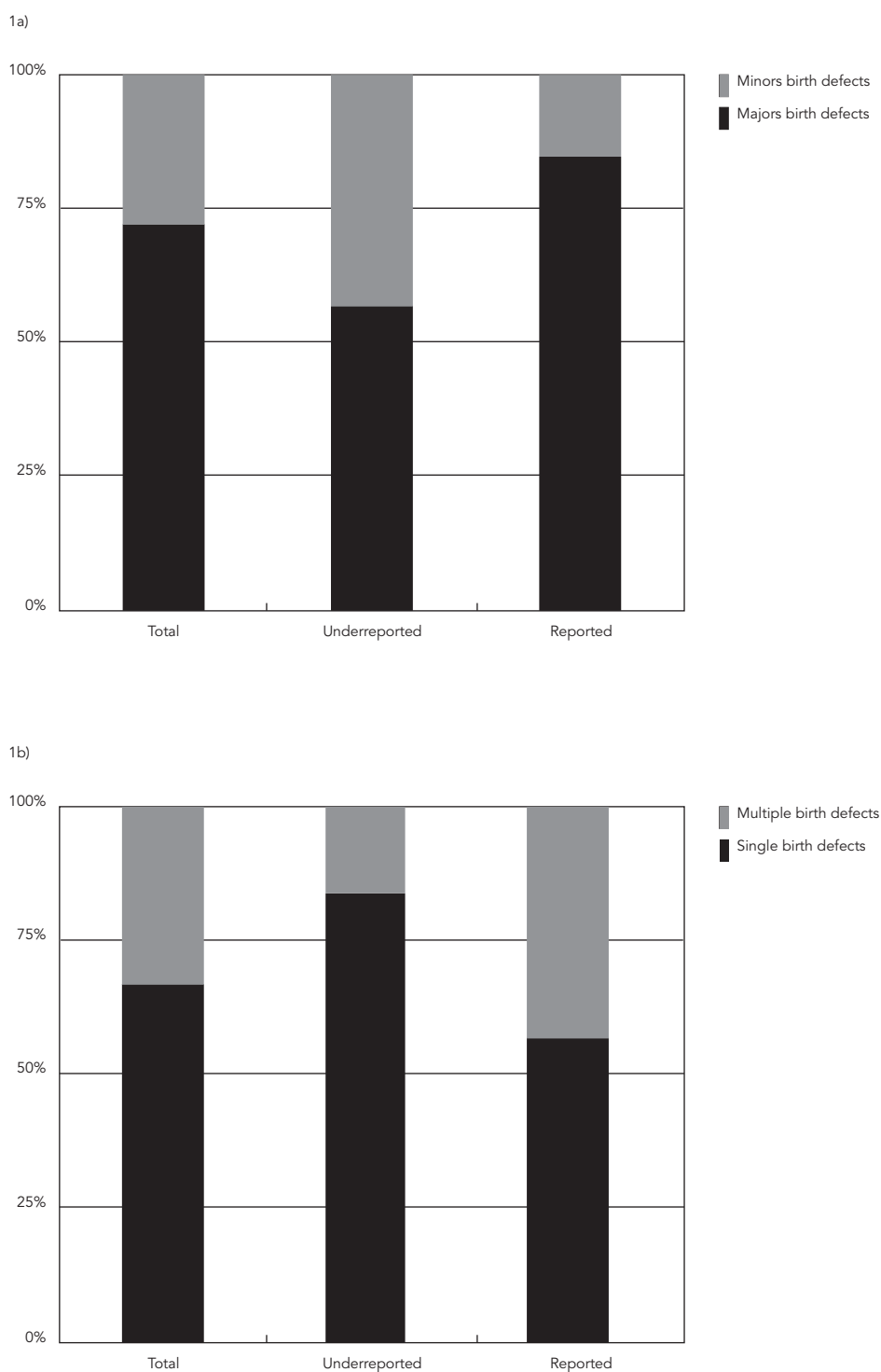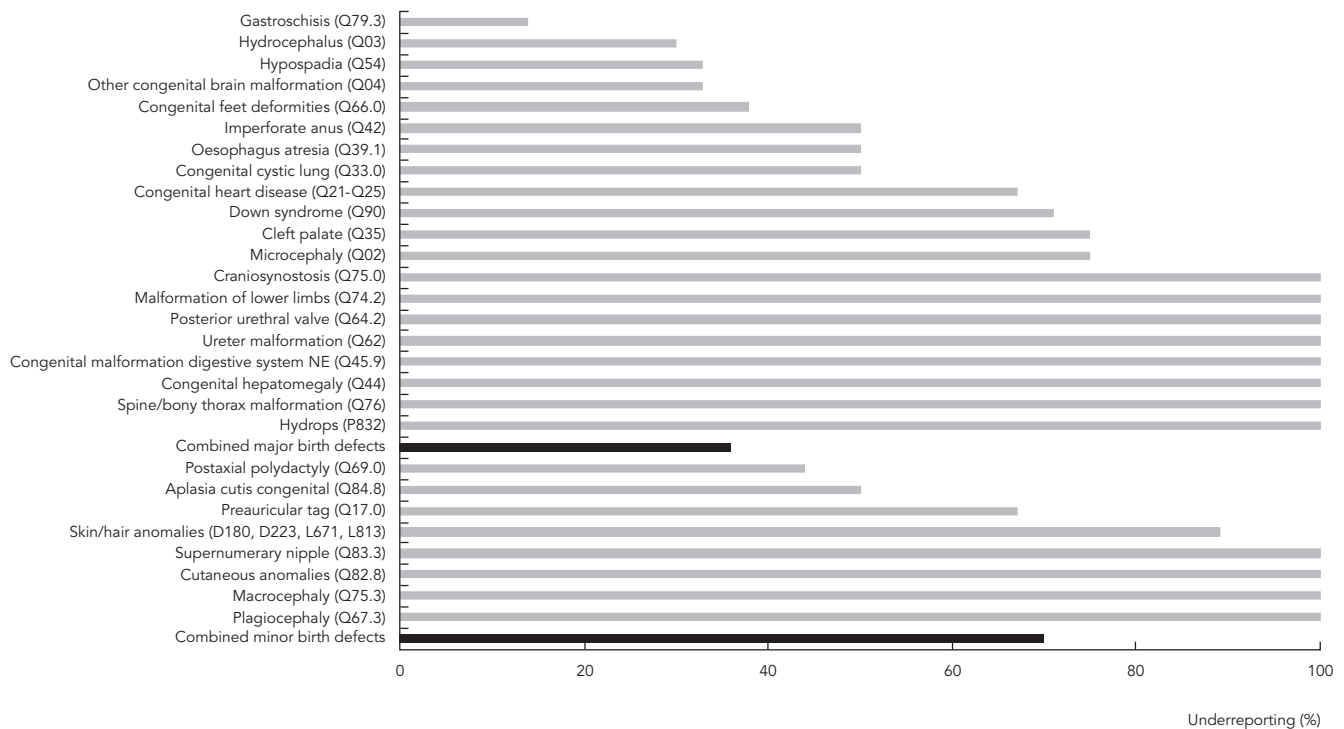
1a)

1b)

Figure 2

Percent underreported at the Center for Women's Integrated Health Care (CAISM) for birth defects by the birth certificate compared to Latin-American Collaborative Study of Congenital Malformations (ECLAMC), 2004.

for the central nervous system anomalies. This analysis considered the general diagnosis, however the diagnostic specificity should improve. For example, all hypospadias in Birth Certificates were not specified.

Varying levels of agreement according to birth defect diagnoses, when comparing the SINASC data to medical records in Rio de Janeiro city, were reported [9]. In such a study, prevalence-adjusted kappa varied according to the three- or four-digit ICD-10 code, with better results for the musculoskeletal, digestive, genitourinary systems and chromosomal anomalies [9].

In a comparative study between the CMR and the Birth Certificate, of the 2,824 captured cases by the two systems 85.2% had diagnostic categories that agreed completely, 9.1% partially agreed and 6% had completely discordant diagnoses. Newborns with a single birth defect had higher levels of agreement in diagnosis (89.7%) compared to newborns with multiple defects (27.5%). Between the defect categories, visible defects, such as oral clefts and gastroschisis had a higher chance of being correctly diagnosed in the two systems.

It was observed in this study that the cases with major and/or multiple birth defects were more reported in relation to minors and single. Similar results were observed in other studies [15,16,17]. Wang et al. [20] observed reporting of 88.7% in multiple cases and 69.5% in cases with single defects. Olsen et al. [18] however, analyzing the cases registered in the Birth Certificate, showed that these had a higher chance of having a single and minor anomaly, which was not verified in the CMR. In the study by Mackeprang et al. [16] the fact that major and easily diagnosed defects were more frequently reported was evaluated by the authors to be the result of selective judgment by the person responsible for completion.

The level of underreporting varies with specific anomalies. In Watkins et al.'s [21] study, it was 14% for anencephaly and 90% for rectal atresia. In the study by Olsen et al. [18] the reporting of cases also differed regarding the type of major birth defect; the Birth Certificate showed under-

Table 3

Sensitivity, specificity positive and negative predictive value (PPV and NPV) from Birth Certificate data on selected birth defects compared with data from ECLAMC. Center for Women's Integrated Health Care (CAISM), Campinas, São Paulo State, Brazil, 2004.

Birth defect	Sensitivity (%)	Specificity (%)	PPV (%)	NPV (%)
Hydrocephaly	69.2 (9/13)	100.0	100.0 (9/9)	99.9
95%CI	41.3-89.4	99.9-100.0	71.7-100.0	99.6-99.9
Spina bifida	100.0 (7/7)	100.0	100.0 (7/7)	100.0
95%CI	65.2-100.0	99.9-100.0	65.2-100.0	99.9-100.0
Cleft lip/palate	57.1 (4/7)	100.0	100.0 (4/4)	99.9
95%CI	21.6-87.7	99.9-100.0	47.3-100.0	99.6-99.9
Preauricular tag	33.3 (3/9)	100.0	100.0 (3/3)	99.9
95%CI	14.7-85.3	99.9-100.0	47.3-100.0	99.6-99.9
Congenital heart disease	40.0 (2/5)	100.0	100.0 (2/2)	99.9
95%CI	7.3-81.7	99.9-100.0	22.4-100.0	99.7-99.9
Gastroschisis	87.5 (7/8)	100.0	100.0 (7/7)	99.9
95%CI	52.0-99.4	99.9-100.0	65.2-100.0	99.8-99.9
Polydactyly	56.2 (9/16)	100.0	100.0 (9/9)	99.7
95%CI	31.9-78.5	99.9-100.0	71.7-100.0	99.5-99.9
Down syndrome	28.6 (2/7)	100.0	100.0 (2/2)	99.9
95%CI	5.1-67.0	99.9-100.0	22.4-100.0	99.6-99.9

reporting for the cardiac and digestive anomalies. In our study, cases of abdominal wall defects (15 cases), were well reported, with only one case (gastroschisis) being missed. The categories that were most underreported were hydrops (100%), microcephaly (75%), cleft palate (75%), congenital heart disease (66%) and Down syndrome (75%). It is important to emphasize that only in one case the diagnosis described in the medical record, at birth, was "Down syndrome", and in all the others, only the classic dysmorphisms of the syndrome were described. The underreporting of this syndrome and of others easily diagnosed could be the result of the health care team's desire for diagnostic confirmation and avoidance of stigmatization of the child.

Anomalies like cleft palate and congenital heart disease are birth defects frequently underreported because the diagnosis is usually done after the reporting period. The Brazilian hospitals receive a financial incentive if the Birth Certificate is filled out in the first 48 hours after birth, in CAISM 73.6% of the Birth Certificate were fulfilled by the second day of birth which could explain this underreporting. However, the SINASC system, in the municipal Health Secretary level, accepts diagnostic rectification after the completion of the Birth Certificate. Our results show that this communication path is not being used; as the underreported cases were collected by ECLAMC during the newborn admission.

Uncertain diagnosis at the time of Birth Certificate completion; and completion by persons not involved in the care of infants, some of whom lack the knowledge to complete it accurately, could be among the possible reasons for the birth defects underreporting in Birth Certificate [21,23]. For example, the person responsible may not know that "trisomy 21" is the same as "Down syndrome". Minton & Seegmiller [17] observed an improvement of the information when the newborn's physician was made responsible for the reporting and a specially trained person was designated for the completion. However, there is no agreement on best practice in this situation. A study by Hemminki et al. [24], with three registers (birth, hospital in-patient discharge and birth defects) in Finland showed discordant data from the American studies. The Birth Certificate presented three times more diagnoses of birth defect cases than the Register of Malformations. Anencephaly and cleft palate and lip were the only birth defects reported in a similar way in both registers. At that time, the Birth Certificate was completed by a clerk after abstracting information from the discharge summary and the Malformation Registry by physicians who were obliged by law to report birth defects.

The underreporting in prevalence can hide the degree to which birth defects are affecting a population; when this underestimated information is used in the evaluation of prevention strat-

egies it can lead to erroneous conclusions about the effectiveness of a program and can influence health policies, as well as resource allocations.

A study by Wang et al. [25], showed that using hospital discharge summaries is an efficient strategy to improve the coverage of birth defects register with a gain of 21% of all cases of the CMR. The SINASC linkage with the Brazilian Hospital Information and Mortality Systems (SIAH and SIM) could be an alternative.

Reliability analysis of birth defects data in the Birth Certificate after recoding revealed an excellent level of agreement for three-digit codes of the ICD-10, however for four-digit codes, agreement was fair. Some possible causes could be a lack of knowledge of some birth defects, inadequate handling of specific codes that classify them and choosing codes of less important anomalies in malformative sequences.

The reliability of the coding process is an essential component of the quality of the data that supplies the birth defect surveillance system, allowing the aggregation of similar cases, storage and recovering of information [26]. Rassmussen & Moore [26] suggested that birth defects should be coded by a professional with knowledge in them and the coding system. Studies have shown false alarms associated with the interpretation of birth defects surveillance data, resulting of inappropriate coding [27,28]. The periodic evaluation of the reliability of coding should be done in a surveillance system.

The SINASC manual of procedures do not establish specific rules for the notification of birth defects [29]. The accuracy of the information generated in Brazilian cities will rise if there is uniformity regarding the definition of a case, description, and the coding process, i.e., a procedures manual for "describers" and "coders". Until 2004, only one birth defect per newborn could be registered in the SINASC database. Considering that many teratogens and chromosomal anomalies are associated with multiple birth defects, the recent introduction of more coding fields per newborn could improve the detection of possible teratogens.

Each surveillance system is created within a unique political, social, geographic, economic and historical context. It also reflects the particular interests and training of the creators of each system. There is no single ideal, universally applicable model, however the diversity of the structure of the programs results in a variability of the register systems and, inevitably in the quality of the data [2]. Considering the resource limitations for surveillance activities, systems implemented that presents limitations should not be abandoned but improved. The Birth Certificate can provide, at least, low-end estimates of the prevalence of birth defects [21].

Folic acid fortification, in the peri-conceptional period, is associated with risk reduction for neural tube defects [30]. A regulatory law for mandatory folic acid enrichment of wheat and corn flour has been enforced in Brazil since June 2004 [31]. The impact of this health policy could be evaluated, at a national level, using the birth certificate data as a population-based database, especially for neural tube defects that are generally easily identified at birth. However, any conclusion should be taken with caution, considering the potential current underreporting of birth defects cases in the period prior to the flour fortification.

The evaluation of the reliability of information, using the medical record as a comparison, evaluates the accuracy of transferring the information from the medical record to the Birth Certificate, but the determination of epidemiological data validity derived from vital statistics also requires comparison with an independent, uniform, and complete source of information. The data found corroborates the indication of ECLAMC as a gold-standard considering that, in an exhaustive revision of the medical records and Birth Certificate, only one case was not registered in ECLAMC.

One of the limitations of this study was the analysis that was performed in a birth defects reference university hospital. Hence, any external validity of the observed results relative to the SINASC database must be evaluated with caution. Nonetheless, a marked underreporting of birth defects, in all Brazilian regions, can be estimated trough the National Health Information System. Considering that CAISM is a university hospital, we may think that the results presented here could correspond to one of the best scenarios, which, if true, will be indicative of the urgent need of SINASC improving.

Further studies should be performed to assess validity for specific birth defects, as the sensitivity, specificity, and predictive values obtained in this study were based on few cases.

In conclusion, the analysis of the quality of birth defects data from Birth Certificates in CAISM showed low sensitivity and moderate reliability. These observations suggest that Birth Certificate information provided by birth certificates still presents limitations when it comes to accurately ascertaining estimates for birth defect prevalence in Campinas.

Resumo

O presente estudo objetivou avaliar a cobertura, validade e confiabilidade do Sistema de Informações sobre Nascidos Vivos (SINASC) para anomalias congênitas, em hospital de Campinas, São Paulo, Brasil. A população de estudo consistiu nos 2.843 nascidos vivos do Centro de Atenção Integrada à Saúde da Mulher (CAISM), em 2004. Na base de dados SINASC, foram selecionados os 92 casos com diagnóstico de anomalia congênita no CAISM. Para o mesmo período, na base de dados ECLAMC, foram selecionados os 168 registros de nascidos vivos com anomalia congênita considerados como padrão-ouro, também no CAISM. Observou-se subnotificação de 46,8% para o conjunto dos casos de anomalias congênitas e de 36,4% quando a análise foi restringida para as anomalias congênitas maiores. A sensibilidade foi de 54,2%, e a especificidade, de 99,8%. Na análise da confiabilidade da codificação das anomalias congênitas, observou-se, para três e quatro dígitos da CID-10, um coeficiente kappa de 0,77 e 0,55, respectivamente. Esses resultados sugerem que o SINASC ainda apresenta limitações como fonte para a determinação da prevalência das anomalias congênitas e indicam que esse sistema demanda aprimoramento para retratar a situação desses casos em Campinas.

Anormalidades Congênitas; Declaração de Nascimento; Sistemas de Informação

Contributors

D. V. Luquetti participated in the planning of the study, data collection, statistical analysis, interpretation of results, and drafting of the article. R. J. Koifman participated in the planning of the study, statistical analysis, interpretation of results, and drafting of the article.

Acknowledgments

References

1. World Health Organization. Human genetics, 1999: services for the prevention and management of genetic disorders and birth defects in developing countries. Geneva: World Health Organization; 1999.

2. Castilla EE, Peters PW. Public health impact of birth defects monitoring systems: national and international efforts. Birth Defects Orig Artic Ser 1992; 28:27-36.

3. Reefhuis J, de Jong-van den Berg LT, Cornel MC. The use of birth defect registries for etiological research: a review. Community Genet 2002; 5:13-32.

4. Castilla EE, Orioli IM. ECLAMC: the Latin-American collaborative study of congenital malformations. Community Genet 2004; 7:76-94.

5. Almeida MF, Alencar GP, França Jr. I, Novaes HMD, Siqueira AAF, Schoeps D, et al. Validade das informações das declarações de nascidos vivos com base em estudo de caso-controle. Cad Saúde Pública 2006; 22:643-52.

6. Mello Jorge MHP, Gotlieb SL, Soboll ML, Almeida MF, Latorre MR. An information system on live births and the use of its data in epidemiology and health statistics. Rev Saúde Pública 1993; 27(6 Suppl):1-46.

7. Romero DE, Cunha CB. Avaliação da qualidade das variáveis epidemiológicas e demográficas do Sistema de Informações sobre Nascidos Vivos, 2002. Cad Saúde Pública 2007; 23:701-14.

8. Theme Filha MM, Gama SG, Cunha CB, Leal MC. Confiabilidade do Sistema de Informações sobre Nascidos Vivos Hospitalares no Município do Rio de Janeiro, 1999-2001. Cad Saúde Pública 2004; 20 Suppl 1:S83-91.

9. Guerra FAR, Llerena Jr. JC, Gama SGN, Cunha CB, Theme Filha MM. Confiabilidade das informações das declarações de nascido vivo com registro de defeitos congênitos no Município do Rio de Janeiro, Brasil, 2004. Cad Saúde Pública 2008; 24:438-46.

10. Cohen J. A coefficient of agreement for nominal scales. Educ Psychol Meas 1960; 20:37-46.

11. Landis JR, Koch GG. Measurement of observer agreement for categorical data. Biometrics 1977; 33:159-74.

12. Guerra FAR, Llerena Jr. JC, Gama SGN, Cunha CB, Theme Filha MM. Defeitos congênitos no Município do Rio de Janeiro, Brasil: uma avaliação através do SINASC (2000-2004). Cad Saúde Pública 2008; 24:140-9.

13. Costa CMS, Gama SGN, Leal MC. Congenital malformations in Rio de Janeiro, Brazil: prevalence and associated factors. Cad Saúde Pública 2006; 22:2423-31.

14. Hexter AC, Harris JA. Bias in congenital malformations information from the birth certificate. Teratology 1991; 44:177-80.

15. Honein MA, Paulozzi LJ. Birth defects surveillance: assessing the "gold standard". Am J Public Health 1999; 89:1238-40.

16. Mackeprang M, Hay S, Lunde AS. Completeness and accuracy of reporting of malformations on birth certificates. HSMHA Health Rep 1972; 87:43-9.

17. Minton SD, Seegmiller RE. An improved system for reporting congenital malformations. JAMA 1986; 256:2976-9.

18. Olsen CL, Polan AK, Cross PK. Case ascertainment for state-based birth defects registries: characteristics of unreported infants ascertained through birth certificates and their impact on registry statistics in New York state. Paediatr Perinat Epidemiol 1996; 10:161-74.

19. Piper JM, Mitchel Jr. EF, Snowden M, Hall C, Adams M, Taylor P. Validation of 1989 Tennessee birth certificates using maternal and newborn hospital records. Am J Epidemiol 1993; 137:758-68.

20. Wang Y, Druschel CM, Cross PK, Hwang SA, Gensburg LJ. Problems in using birth certificate files in the capture-recapture model to estimate the completeness of case ascertainment in a population-based birth defects registry in New York State. Birth Defects Res A Clin Mol Teratol 2006; 76:772-7.

21. Watkins ML, Edmonds L, McClearn A, Mullins L, Mulinare J, Khoury M. The surveillance of birth defects: the usefulness of the revised US standard birth certificate. Am J Public Health 1996; 86: 731-4.

22. Knox E, Armstrong E, Lancashire R. The quality of notification of congenital malformations. J Epidemiol Community Health 1984; 38:296-305.

23. Hexter AC, Harris JA, Roeper P, Croen LA, Krueger P, Gant D. Evaluation of the hospital discharge diagnoses index and the birth certificate as sources of information on birth defects. Public Health Rep 1990; 105:296-307.

24. Hemminki E, Merilainen J, Teperi J. Reporting of malformations in routine health registers. Teratology 1993; 48:227-31.

25. Wang Y, Sharpe-Stimac M, Cross P, Druschel C, Hwang S. Improving case ascertainment of a population-based birth defects registry in New York State using hospital discharge data. Birth Defects Res A Clin Mol Teratol 2005; 73:663-8.

26. Rasmussen SA, Moore CA. Effective coding in birth defects surveillance. Teratology 2001; 64 Suppl 1:S3-7.

27. Cunniff C, Kirby RS, Senner JW, Canino C, Brewster MA, Butler B, et al. Deaths associated with renal agenesis: a population-based study of birth prevalence, case ascertainment, and etiologic heterogeneity. Teratology 1994; 50:200-4.

28. Reefhuis J, Walle H, Cornel MC. Artefactual increasing frequency of omphaloceles in the Northern Netherlands: lessons for systematic analysis of apparent epidemics. Int J Epidemiol 1999; 28:258-62.

29. Fundação Nacional de Saúde. Manual de intruções para o preenchimento da Declaração de Nascido Vivo. Brasília: Fundação Nacional de Saúde; 2001.

30. Lopez-Camelo JS, Orioli IM, Graca DM, Nazer-Herrera J, Rivera N, Ojeda ME, et al. Reduction of birth prevalence rates of neural tube defects after folic acid fortification in Chile. Am J Med Genet A 2005; 135:120-5.

31. Agência Nacional de Vigilância Sanitária. Resolução RDC nº. 344, de 13 de dezembro de 2002. Diário Oficial da União 2002; 18 dez.