

Research Article

Trends in Cerebral Palsy Infant Mortality from 1981 to 2011 in Spain

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Abstract

Background: Cerebral palsy (CP) is due to a defect or lesion of the immature brain and it is the most common form of chronic motor disability in children. In general, the health care system and, especially, perinatal and neonatal care, are considered to influence the prognosis of CP. The aim of this study was to analyze the trend in CP infant mortality in Spain in a 31-years period (1981-2011).

Methods: Data on CP deaths of children up to 1 year old were drawn from the National Statistics Institute of Spain, using the International Classification of Diseases. Crude CP infant mortality rates per 100000 live births were calculated, overall and by gender. Joinpoint regression models were used to describe changes in trend and an annual percentage change was calculated for each trend.

Results: The CP overall infant mortality rate was 0.94 in 1981 and 0.42 in 2011. By gender, infant mortality rates were 1.41 and 0.00 per 100000 male life births and 0.39 and 0.87 per 100000 female life births, in 1981 and 2011, respectively. The overall and male infant mortality rate showed a statistically significant decrease from 1981 to 2011. In female, the infant mortality rate decreased, although it was not statistically significant.

Conclusions: We support the impression that improvements in CP prognosis, in particular the decrease in infant mortality rates observed in this study, are more likely to reflect medical practices than changes affecting all the population.

INTRODUCTION

The term cerebral palsy (CP) encompasses a group of disorders characterized by chronic disorders of movement and posture due to a defect or lesion of the immature brain [1]. In fact, CP is the most common form of chronic motor disability in children [2]. Although CP occurrence can be influenced by several factors, most studies agree on the prevalence of CP at approximately 2 to 3 per 1000 live births, which has been remarkably stable during the past four decades [3-5]. An average prevalence of 2.08 and 2-2.5 per 1000 live births has been estimated in Europe [6] and in the United States [7], respectively. In Spain, a prevalence of 0.5 per 1000 live births was reported, although authors indicated that the figure underestimated the true prevalence [8]. In contrast with the stable CP prevalence, most studies carried out in other countries agree that CP mortality is decreasing and survival is increasing in the last decades [4,5,9,10].

Approximately, half of the CP cases diagnosed are born

with very low birth weight (< 1500 g) [11]. CP prevalence rates increase as gestational age decreases [4] and mortality is mainly due to pneumonia [12]. The aetiology of cerebral palsy is multifactorial, perinatal factors being the most important ones (80-90%) [13]. Among them, congenital malformations stand out, in particular, cerebral malformations with altered migration and cell division. Other well-known factors are intracranial haemorrhage, especially in preterm birth, multiple births (rising due to fertility treatments) and intrauterine infections [2,14,15]. Nevertheless, the causative link between several factors and incidence of CP has been questioned in some studies [5] and factors causing neonatal mortality not seem to be the same which produce mortality subsequently [9]. In general, the health care system and, especially, perinatal and neonatal care, are considered to influence the prognosis of CP [4,16].

The aim of this study was to analyze the trend in CP infant mortality in Spain in a 31-years period (1981-2011).

MATERIAL AND METHODS

Data on CP deaths of children up to 1 year old were drawn from the National Statistics Institute (NSI) (*Instituto Nacional de Estadística*), which collects data from all death certificates in Spain. The underlying cause of death was coded using the International Classification of Diseases, ninth revision (ICD-9), from 1981 to 1998, and tenth revision (ICD-10), from 1999 onwards. CP deaths were defined as those with an underlying cause of death coded as 343 (343.0 to 343.9) in ICD-9 and G80 (G80.0 to G80.9) in ICD-10.

Population data of live births in Spain during the study period were obtained from NSI census figures, broken down by gender. For each gender, crude CP infant mortality rates per 100000 live births were calculated. Joinpoint regression models were used to describe changes in trend. In joinpoint analysis, the best-fitting points (known as “joinpoint”), where the rate changes significantly (upwards or downwards), is chosen [17]. The analysis starts with the minimum number of joinpoints, and tests whether one or more joinpoints are statistically significant and must be added to the model (up to four joinpoints). In the final model, each joinpoint indicates a statistically significant change in trend, and an annual percentage change (APC) is calculated for each of these trends by means of generalized linear models, assuming a Poisson distribution.

Both adjusted and smoothed rates have been added to the trend figures for facilitating their reading. The smoothed values were used to interpret the visual representations of the data, not to fit the joinpoint analysis. The T4253H compound data smoothing procedure has been carried out using the SPSS statistical package.

RESULTS AND DISCUSSION

During the period 1981-2011, 118 children (64 male and 54 female) died in their first year of live because of CP in Spain. The CP overall infant mortality rate was 0.94 (95% confidence interval: 0.12-1.76) in 1981 and 0.42 (95 CI: 0.00-1.01) in 2011. By gender, infant mortality rates were 1.41 (95 CI: 0.03-2.85) and 0.00 (95 CI: 0.00-1.78) per 100000 male life births and 0.39 (95 CI: 0.00-1.16) and 0.87 (95 CI: 0.00-2.09) per 100000 female life births, in 1981 and 2011, respectively (table 1 and figure 1). There are not recent studies reporting CP infant mortality rates in Spain, so this study provides data to assess the CP impact on live births.

Joinpoint analysis of CP infant mortality rates revealed that there was not a statistically significant change in the trend during the study period. The overall and male infant mortality rate showed a statistically significant decrease from 1981 to 2011, with APC of -2.62% and -1.85%, respectively. In female, the infant mortality rate decreased with an APC of -0.48%, although it was not statistically significant.

The decrease in CP infant mortality observed in our work agrees with previous studies on outcome and trends in CP which stated a reduction of mortality and an increase of survival during the last 10 to 20 years [4,9,10]. The CP overall infant mortality rate was reduced to half in the period 1981-2011 in Spain, in agreement with other reports [10].

Table 1: CP infant mortality rates in Spain, 1981 to 2011.

Year	Deaths			Infant mortality rate*		
	Male	Female	Overall	Male	Females	Overall
1981	4	1	5	1.44	0.39	0.94
1982	2	2	4	0.74	0.81	0.78
1983	4	3	7	1.59	1.28	1.44
1984	6	1	7	2.44	0.44	1.48
1985	4	1	5	1.69	0.45	1.10
1986	4	4	8	1.76	1.89	1.82
1987	2	4	6	0.90	1.95	1.41
1988	2	1	3	0.92	0.49	0.72
1989	2	3	5	0.95	1.52	1.22
1990	2	0	2	0.96	0.00	0.50
1991	1	1	2	0.49	0.52	0.51
1992	2	2	4	0.98	1.04	1.01
1993	3	2	5	1.50	1.07	1.30
1994	3	4	7	1.57	2.23	1.89
1995	0	3	3	0.00	1.70	0.83
1996	1	1	2	0.54	0.57	0.55
1997	0	0	0	0.00	0.00	0.00
1998	0	1	1	0.00	0.57	0.27
1999	1	2	3	0.51	1.08	0.79
2000	3	0	3	1.46	0.00	0.75
2001	2	1	3	0.96	0.51	0.74
2002	0	3	3	0.00	1.48	0.72
2003	2	3	5	0.88	1.40	1.13
2004	2	3	5	0.85	1.37	1.10
2005	2	0	2	0.83	0.00	0.43
2006	0	2	2	0.00	0.86	0.41
2007	4	0	4	1.58	0.00	0.81
2008	0	2	2	0.00	0.80	0.38
2009	2	0	2	0.78	0.00	0.40
2010	2	2	4	0.80	0.85	0.82
2011	0	2	2	0.00	0.87	0.42

* Rate per 100000 live births.

Some authors have observed that factors associated with neonatal mortality were different than those implicated in the outcome at two years of age [9]. The former was mainly dependant on gestational age, birth weight and the use of antenatal corticosteroids whilst the later was predicted by bronchopulmonary dysplasia, retinopathy of prematurity and major brain lesions. Improvements over time in gross motor function, indicating less disability, and a concomitant decrease in the proportion of children with quadriplegia and an increase in the proportion with diplegia have been recently observed in children born at term [4]. The authors related those healthier infants suffering less extensive brain damage with improvements in perinatal and neonatal care. Children suffering severe disabilities have also shown an increase in their life expectancy in the past years, which was suggested to be attributable to multiple and overlapping reasons [10].

In general, factors that have been associated with CP outcomes have been the gestational age, the birth weight, the use of antenatal and postnatal corticosteroids and surfactant, an early recognition and vigorous treatment of infections, the attention to and management of the nutritional requirements, the gross motor function levels and even socioeconomic factors [4,5,9,10,16,18].

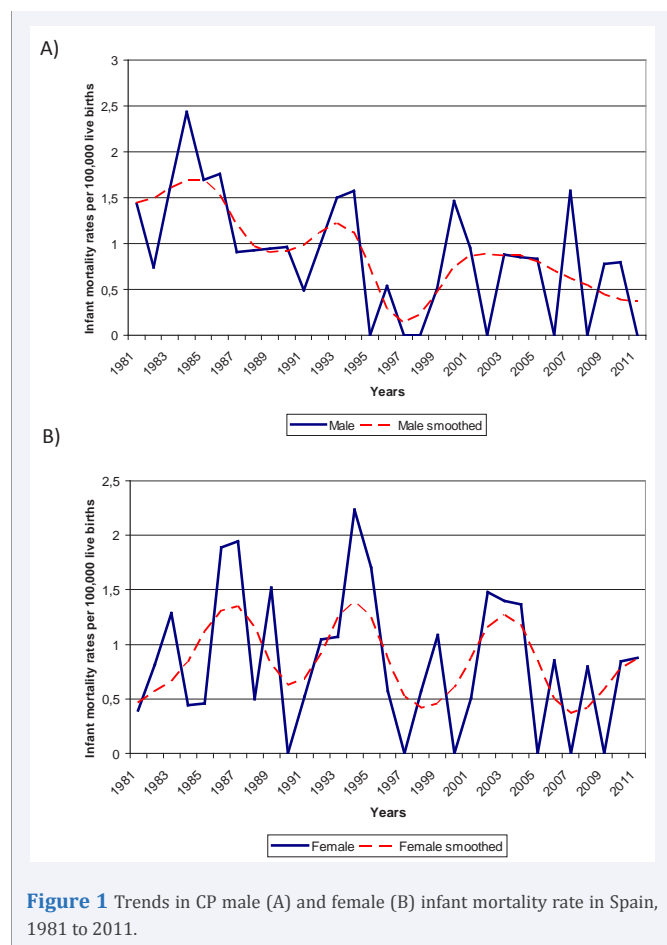


Figure 1 Trends in CP male (A) and female (B) infant mortality rate in Spain, 1981 to 2011.

As has been elsewhere recognized, cerebral palsy is not known to be preventable nowadays [2]. However, obstetricians and the health care system in general could be the key to improve life expectancy and survival of children suffering CP by offering the right treatments and avoiding exposure to suggested risk factors. There are several aspects that have changed in Spain during the study period and that may have played an important role in the decrease of the infant mortality rate observed in our study. Some of them could be the universal and free health care system or the increase of obstetrical resources.

CONCLUSIONS

The overall and male infant mortality rate showed a statistically significant decrease from 1981 to 2011. In female, the infant mortality rate decreased, although it was not statistically significant.

Some limitations of this study have to be mentioned. Firstly, some studies observed that fewer than a third of deaths are attributed to cerebral palsy as underlying cause of death on death certificates and that there was no mention of cerebral palsy on 46% of certificates [19]. Consequently, our results of infant mortality rates are likely considerably underestimated. Secondly, the data source used in this study only provided the underlying cause of death and thus, other concomitant causes of death are unknown. For that reason, all deaths analyzed in this study were directly related to CP but other diagnoses related were not

accessible in the data source. Finally, the validity of codes provided by the NSI has not been previously evaluated and therefore, we do not know the proportion of codes incorrectly assigned from data in the death certificates. Despite these limitations, data of the NSI based on death certificates remain a valuable source of information since we aimed to assess mortality trends and the underestimation is supposed to be constant over the time. On the other hand, the change in classification procedures from ICD-9 to ICD-10 in death certificates might have introduced some biases, although we suppose they are probably not significant since both ICD codes for CP are specific codes.

We support the impression that improvements in CP prognosis, in particular the decrease in infant mortality rates observed in this study, are more likely to reflect medical practices than changes affecting all the population.

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