

Hospital incidence and costs of congenital hydrocephalus in Spain: a multicenter retrospective study

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Author contributions

JD contributed to the investigation by analyzing and interpreting the burden associated to congenital hydrocephalus in Spain and was a major contribution in the intellectual content revision. AM analyzed the current situation of congenital hydrocephalus in Spain, interpreted the statistical data and was a major contributor in writing the manuscript. All authors read and approved the final manuscript.

Acknowledgements

Not applicable.

Abstract

Background: Congenital hydrocephalus continues to represent a therapeutic challenge; however, few studies are available evaluating its hospital incidence and medical costs in Spain. This study aimed to review the profile of patients with this disorder admitted in Spanish hospitals and to estimate medical costs at the hospital level.

Methods: Records of hospital admissions of patients with congenital hydrocephalus between 2010 and 2019 were obtained from a Spanish hospital discharge database and analyzed in a retrospective multicenter study.

Results: A third of the patients included in the study were perinatal patients, however, the hospitalization rate in this group was higher to that in patients over 1 year of age. Perinatal patients required more ICU admissions and longer hospital stays, with more frequent surgical interventions and mechanical ventilation. The mean medical cost associated to congenital hydrocephalus was €9610 per admission, with significantly higher costs found in perinatal patients.

Conclusions: This study provides novel data on the hospital costs of congenital hydrocephalus in Spain. The hospital medical costs of this disorder have decreased over the past decade for perinatal patients but not in those aged 1 year and older, which should be considered in upcoming healthcare plans and resource allocation decisions.

Keywords: congenital hydrocephalus; cost analysis; direct medical costs; Spain.

1. Introduction

Congenital hydrocephalus is a developmental disorder of the brain characterized by the excessive accumulation of cerebrospinal fluid within the cerebral ventricles. The disorder is originated during the fetal development and is associated to postnatal neurological consequences that can include developmental delay and other symptoms [1].

The estimated prevalence of congenital hydrocephalus in Europe is 4.65 per 10,000 births [2]. Diagnosis in the prenatal period has been associated with worse outcomes, while the general mortality rate fluctuates between 5% and 15% [2,3].

Postnatal treatment is the most frequent approach, mainly with endoscopic third ventriculostomy (ETV) and ventriculoperitoneal shunt (VPS) insertion [4]. Additionally, fetal interventions are gaining interest in the past years [5]. Overall, shunt interventions have proven effective, yet, to improve patients quality of life other factors must be considered, including comorbidity and clinical background [4,6]. Moreover, a curative treatment able to prevent neurological damage is not available [7].

Few data is available regarding the incidence and management of this disorder in Spain, and the costs of the disease have not been evaluated, which represents a challenge for the improvement of healthcare planning and resource allocation for the disorder.

The objective of this study was to evaluate the number and characteristics of patients with congenital hydrocephalus in Spanish hospitals, and to analyze the medical costs associated to this disorder at the hospital level.

2. Methods

2.1. Study design

Records of outpatient visits and inpatient admissions were obtained from a Spanish National discharge database (covering 90% of hospitals in Spain), and analyzed in a

retrospective study. The data included in the database is codified at the hospital level via the International Statistical Classification of Diseases and Related Health Problems, 9th version (ICD-9) prior to 2016 and 10th version (ICD-10) after the year 2016 [8,9]. Data inclusion period was from 1 January 2016 to 31 December 2019. Centers are responsible for data codification, assessment and confidentiality. The database is validated internally and subjected to periodic audits; in this process, errors and unreliable data are eliminated.

2.2. Data extraction

The ICD-9 and ICD-10 codes used to identify records of congenital hydrocephalus were 742.3 and Q03, respectively. One single admission with a diagnosis of congenital hydrocephalus was considered sufficient for inclusion in the study. Parameters identifying healthcare centers or medical history were re-coded at the hospital center, and all records obtained were previously anonymized, in accordance with the principles of Good Clinical Practice and the Declaration of Helsinki. This research did not involve human participants and there was no access to identifying information; in this context the Spanish legislation does not require patient consent and ethics committee approval [10].

2.3. Study variables

The variables analyzed were: patients' age, sex, date of admission, type of admission, ICU stay, date of discharge, type of discharge, primary diagnosis, up to 20 secondary diagnoses registered during the admission, medical procedures and total admission cost. Admission costs correspond to 2019€ and are adjusted for inflation. The information regarding ICU stay was only available for 2018 and 2019.

2.4. Data analysis

Patients were indexed and classified based on the primary diagnosis and into two age groups: patients under 1 year of age and patients aged 1 year or older. The characteristics of patients were evaluated in the file corresponding to the first admission registered per patient, while admission details and costs were analyzed in all admission files.

Age-adjusted hospitalization rate was defined as the annual number of patients registered with congenital hydrocephalus within the total number of admission files in the database. Direct medical costs of specialized healthcare were obtained directly from the database, where they are automatically assigned according to the standardized average expenses of admissions and medical procedures determined by the Spanish Ministry of Health (these include the costs of examination, medication, surgery, diet, costs associated to personnel, medical equipment and resources).

Normality was tested with the Kolmogorov-Smirnov test. Frequencies and percentages are presented for dichotomous variables and mean and standard deviation (SD) or median and interquartile range (IQR) were calculated for continuous variables. Time trends were evaluated using the Spearman correlation test, And two-tailed non-parametric independent t-test (Mann-Whitney U test) or one-way analysis of variance (Kruskal-Wallis test) were used as appropriate. A $p < 0.05$ was considered statistically significant.

Statistical analyses were performed using Microsoft Excel© Professional Plus 2016 (Microsoft Corporation, Redmond, WA, USA) and StataSE 12 for Windows (StataCorp LP, 2011. Stata Statistical Software: Release 12. College Station, TX, USA).

3. Results

In total, 2087 admissions were identified between 2010 and 2019 with congenital hydrocephalus in the hospital discharge database; these admissions corresponded to

1716 individual patients (Table 1). Patients over 1 year of age displayed a median age of 36 years; age range in this group was 1-91 years. Patients were diagnosed with numerous secondary conditions, the most frequent being episodic and paroxysmal disorders (epilepsy, migraine, sleep apnea), essential primary hypertension, hyperlipidemias, congenital malformations of the circulatory system and musculoskeletal system (macrocephaly, unspecified malformation of skull and face bones), bacterial and viral infections, disorders of the newborn (low birth weight, preterm newborn), hemorrhagic disorders and diabetes mellitus. Patients were classified into two groups according to age, with significant differences in the presence of comorbid conditions between the two groups.

In terms of all-cause mortality, 16 deaths were registered during the hospital admission in perinatal patients, 13 in patients aged 1 year or older. Overall in-hospital mortality rate was 1.7%.

The annual hospitalization rate measured in this database was 0.5 per 10,000 patients, 5.2 per 10,000 when considering perinatal patients alone. No significant trends were identified in the hospitalization rate over time.

Forty-one percent of the registered admissions were urgent or non-scheduled, and median length of hospital stay was 5 days (Table 2). More frequent urgent and ICU admissions were registered in perinatal patients. In addition, hospital stays were significantly longer in perinatal patients ($p < 0.0001$); the same was found for ICU stays ($p = 0.0095$) (Table 4). Neurosurgery was the most common department registered at discharge, found in 52.4% of admissions, in perinatal patients the most common department was pediatrics.

Numerous medical procedures were registered during the hospital admission (Table 3). The most frequent procedures corresponded to imaging diagnostic procedures, cerebral bypass, cerebral drainage and the introduction of therapeutic substances.

Mean annual admission cost was €9610 (Table 4). Longer hospital stays and ICU admission were associated to higher admission costs. The total admission cost was significantly higher in perinatal patients ($p < 0.0001$), this difference was not found for ICU admissions.

Mean admission costs decreased significantly over the study period in the total study population ($p = 0.0002$) and in perinatal patients ($p = 0.0011$), in patients over 1 year of age no significant trends were identified in admission costs over time ($p = 0.1139$) (Figure 1).

4. Discussion

There is few data available describing the situation of congenital hydrocephalus in Spain. This study aimed to provide a general description of hospital incidence, patient profile, care received and medical costs. Median age was 9 years, 33.5% of the patients included in the study were perinatal patients. The comorbid conditions registered during the hospitalization were significantly different between age groups and may have implications for treatment outcomes and quality of life. The presence of several comorbid conditions at shunt implantation, including systemic infection and coagulopathy, have been associated with lower life expectancy and are contraindications for surgery [6].

The hospitalization rate was relatively high in perinatal patients in this database. In addition, patients under 1 year of age registered a higher proportion of urgent and ICU admissions and more extended hospital stays.

The use of imaging diagnostic procedures was to be expected in this population. Computed tomography (CT) and magnetic resonance imaging (MRI) are recommended in guidelines to assess brain morphology [11]. Cerebral bypass was the most frequent procedure in both of the age groups analyzed although it was only registered in 47.2% of admissions.

Few studies have attempted to quantify the hospital-related cost of congenital hydrocephalus. Several studies are available evaluating the cost-effectiveness of different surgical approaches in hydrocephalus, suggesting that timely and effective interventions reduce medical costs that otherwise can represent a significant burden for healthcare systems [12,13]. In addition, high readmission rates and shunt failure have been associated with higher costs [14].

The medical costs associated congenital hydrocephalus in this study averaged €9610 per admission, while the Spanish ministry of health declared an overall medical cost per citizen of €2255 the year 2018 in the general population, €1421 spent in specialized care [15]. This data suggests that the medical costs associated to patients with congenital hydrocephalus are substantially higher to those registered in the general population, presumably due to the length of hospital stay and overall care needs. A 2020 study estimated a median admission cost in patients with idiopathic normal pressure hydrocephalus of €9554 (\$11 230) in the United States [16]. In Australia, the cost of initial surgical treatment of hydrocephalus was €8635 (\$14 000) per admission, while the cost of shunt revision was €6016 (\$9753) per admission [17].

Medical costs herein were significantly higher in perinatal patients and ICU admissions, increasing with length of hospital stay. Finally, a decrease in admission costs was observed over time in perinatal patients and the total study population, but not in patients aged 1 year or older.

A number of limitations may have influenced the results of this study. Data inclusion was limited to hospital facilities and to that registered in the database. The analysis of specific surgical procedures was restricted by the data codification system used in the database and ICU data was only available for 2018 and 2019. Additionally, the database provides representative hospitalization data, not allowing the analysis of long-term patient management.

5. Conclusions

This study provides an evaluation of the cost of congenital hydrocephalus in Spanish hospitals, aiming to support healthcare planning and resource allocation decisions in the country. The hospital medical costs associated to this disorder have decreased over the study period in perinatal patients but have remained stable in patients over 1 year of age. Additional research will be required to further understand the economic and social burden of congenital hydrocephalus in Spain.

6. Transparency section

6.1. Ethics approval and consent to participate

Ethics committee approval and consent were not required for this study.

6.2. Data availability statement

Data sharing is restricted due to legal stipulations, yet the data that support the findings of this study is fully available from the Spanish Ministry of Health via the Unit of Health Care Information and Statistics (Spanish Institute of Health Information) for researchers who meet the criteria for access to confidential data at: <https://www.msrebs.gob.es/estadEstudios/sanidadDatos/home.htm>.

6.3. Declaration of funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

6.4. Declaration of financial and other interest

The authors declare that they have no competing interests.

7. References

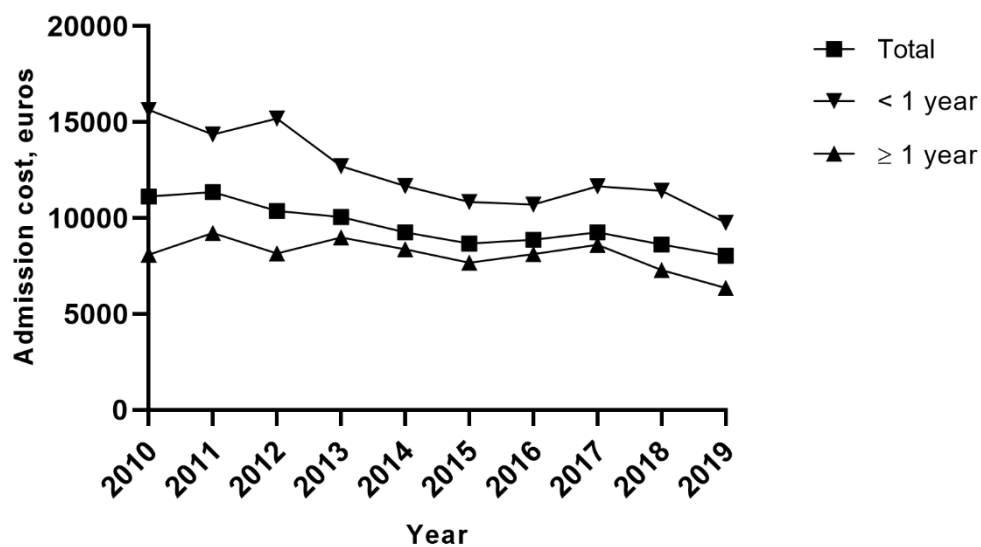
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8. Figures

Figure 1 Mean admission cost by age group in patients with congenital hydrocephalus.



9. Tables

Table 1 Characteristics of patients diagnosed with congenital hydrocephalus.

	Total	Perinatal patients (< 1 year of age)	Patients ≥ 1 year of age	p-value
Admissions, N	2087	663	1424	-
Patients, N	1716	574	1142	-
Median age, years (IQR)	9 (49)	0 (0)	36 (56)	<0.0001
Age range, years	0-91	0-0	1-91	-
Males, %	54.1	55.7	53.2	0.3251
Episodic and paroxysmal disorders, %	9.1	4.4	11.3	<0.0001
Essential primary hypertension, %	11.8	0.6	17.0	<0.0001
Hyperlipidemias, %	9.7	0.0	14.3	<0.0001

Congenital malformations and deformations of the musculoskeletal system, %	5.9	8.6	4.1	<0.0001
Bacterial, viral infection, %	5.7	9.2	4.0	<0.0001
Disorders of newborn related to length of gestation and fetal growth, %	7.0	22.0	0.1	<0.0001
Hemorrhagic and hematological disorders of newborn, %	5.7	17.5	0.1	<0.0001
Congenital malformations of the circulatory system, %	5.7	13.4	2.0	<0.0001
Diabetes mellitus, %	6.7	0.0	9.8	<0.0001

IQR: Interquartile range.

Table 2 Characteristics of hospital admissions by age group in patients with congenital hydrocephalus.

	Total	Perinatal patients (< 1 year of age)	Patients ≥ 1 year of age	p-value
Urgent admissions, %	41.9	60.2	33.6	<0.0001
Admissions into ICU, %	24.1	81.0	19.6	0.0014
Department registered at discharge:	-	-	-	-
Neurosurgery, %	52.4	21.4	84.2	<0.0001
Pediatrics, %	18.4	32.7	14.9	<0.0001
Pediatric surgery, %	9.1	9.8	11.0	0.4399
Neonatology, %	8.7	27.3	0.0	<0.0001
Neurology, %	4.6	0.8	8.0	<0.0001
Neonatal intensive medicine, %	1.0	3.2	0.0	<0.0001

ICU: Intensive care unit.

Table 3 Procedures registered during the hospital admission by age group in patients with congenital hydrocephalus.

	Total	Perinatal patients (< 1 year of age)	Patients \geq 1 year of age	p-value
Cerebral bypass, %	47.2	54.1	44.0	<i><0.0001</i>
Drainage of brain, cerebral ventricle or intracranial space, %	17.4	17.8	17.2	0.7394
Introduction of therapeutic substance, %	26.1	52.0	14.0	<i><0.0001</i>
Introduction of nutritional substance, %	3.2	7.3	1.5	<i><0.0001</i>
Measurement of intracranial pressure or nervous conductivity/electrical activity, %	6.0	11.6	3.4	<i><0.0001</i>
Monitoring of	13.5	2.3	18.8	<i><0.0001</i>

intracranial pressure, %				
Monitoring of cardiac function, %	2.6	7.8	0.6	<0.0001
Respiratory ventilation, %	4.6	10.7	1.7	<0.0001
Computerized tomography (CT scan) of brain or skull or head, %	34.5	21.6	40.5	<0.0001
Magnetic resonance imaging (MRI) of brain or head, %	29.3	37.7	25.4	<0.0001
Ultrasonography of brain, %	15.7	47.2	1.0	<0.0001
Ultrasonography of heart, %	4.6	12.1	1.1	<0.0001
Ultrasonography of abdomen, %	5.6	13.4	2.0	<0.0001

Table 4 Mean annual admission costs by age group and admission type in patients with congenital hydrocephalus.

	Total	Perinatal patients (< 1 year of age)	Patients ≥ 1 year of age	p-value
Mean LOS, days (SD)	9.1 (16.0)	14.1 (23.8)	6.7 (9.7)	<0.0001
- Median (IQR)	5 (6)	7 (12)	4 (5)	-
Mean LOS in ICU, days (SD)	4.4 (8.5)	7.0 (11.6)	2.3 (3.8)	0.0095
- Median (IQR)	1 (3)	2 (8.5)	1 (1.5)	-
Mean admission cost, € (SD)	9610 (9513)	12633 (12951)	8202 (6957)	<0.0001
- Median (IQR)	8206 (8559)	9433 (9354)	7823 (7871)	-
Mean cost per day, € (SD)	2020 (1984)	1797 (2058)	2124 (1940)	<0.0001
- Median (IQR)	1516 (1938.8)	1292 (1800.2)	1666 (2073.7)	-
Mean admission cost in hospital admissions ≤ 4 days, € (SD)	6425 (4694)	5861 (5476)	6605 (4403)	<0.0001
- Median (IQR)	3911 (7335)	3663 (5036)	3912 (7444)	-

Mean admission cost in hospital admissions > 4 days, € (SD)	11678 (10220)	14447 (11909)	10082 (8724)	<0.0001
- Median (IQR)	10573 (7527)	10881 (6247)	10515 (8145)	-
Mean admission cost in ICU admissions, € (SD)	12077 (10380)	13328 (10556)	11077 (10244)	0.3027
- Median (IQR)	10573 (3628)	10111 (5409)	10573 (7280)	-
Mean cost per day in ICU, € (SD)	3522 (3517)	3279 (3932)	4360 (3079)	0.4685
- Median (IQR)	2076 (4923.7)	1442 (4618.6)	5422 (5875.8)	-

LOS: Length of stay. ICU: Intensive care unit. SD: Standard deviation. IQR: Interquartile range.