

MALFUNCTION OF THE VALVULAR SHUNTING SYSTEM IN CHILDREN. EXPERIENCE IN THE NEUROSURGERY DEPARTMENT OF THE BACA ORTIZ PEDIATRIC HOSPITAL, QUITO-ECUADOR, 2016-2019

Disfunción del sistema de derivación valvular en niños. Experiencia en el servicio de neurocirugía del Hospital Pediátrico Baca Ortiz, Quito-Ecuador, 2016-2019

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ABSTRACT

Introduction: The prevalence of valve dysfunction (VD) in pediatric centers is high. A descriptive observational study was carried out, the objective of which was to find factors: epidemiological characteristics of hydrocephalus and other derivatives of the ventriculoperitoneal shunt system (VPSS) associated with VD.

Methods: All the diagnoses of VD for 3 years in the Neurosurgery Service of the Baca Ortiz Pediatric Hospital (BOPH) were collected; clinical-epidemiological variables associated with hydrocephalus and shunting were selected. VD was defined as the revision of the referral in patients using VPSS for malfunction. Multi-categorical variables and the prevalence of qualitative variables were analyzed using statistical analysis.

Results: A total of 376 children were initially subjected to VPSS placement between August 2016 and August 2019. 71 patients with VD were treated, of whom 60 were included in the study; of these 48 were treated at BOPH. Infectious dysfunction was found to be more frequent in children < 1-year, mechanical dysfunction in children > 5 years (64% and 38% respectively $p < 0.002$). Also, the permanence of the VPSS < 1 year was more frequent in infectious dysfunctions and the permanence of 1 to 5 years was more related to mechanical dysfunction (72 and 46% respectively $p 0.03$). The distal catheter dysfunction was more important in mechanical and mixed etiology (65.41 respectively $p < 0.001$) and that of the ventricular catheter in infectious etiology (81% $p < 0.001$). No independent association of VD was found with the etiology of hydrocephalus, the ventricular catheter site or with the Lansky scale.

Conclusions: Valvular dysfunction in pediatric hydrocephalic patients is an important complication that warrants further investigation.

Keywords: Hydrocephalus, Catheters, Neurosurgical Procedures, Hospitals, Pediatric. (Source: MeSH NLM)

RESUMEN

Introducción: La prevalencia de disfunción valvular (DV) en centros pediátricos es alta. Se realizó un estudio observacional descriptivo, cuyo objetivo fue encontrar factores: epidemiológicos propios de la hidrocefalia y otros derivados del sistema de derivación ventrículo peritoneal (SDVP) asociados a la DV.

Métodos: Se recopiló todos los diagnósticos de DV durante 3 años en el servicio de neurocirugía del Hospital Pediátrico Baca Ortiz (HPBO), se seleccionó variables clínico-epidemiológicas asociadas a la hidrocefalia y a la derivación. La DV se definió como la revisión de la derivación en pacientes usuarios de SDVP por mal funcionamiento. Se analizaron las variables multicategorías y la prevalencia de variables cualitativas mediante análisis estadístico.

Resultados: Un total de 376 niños fueron sometidos inicialmente a colocación de SDVP entre agosto de 2016 y agosto de 2019. Se trataron 71 pacientes con DV de los cuales 60 fueron incluidos en el estudio, de estos 48 fueron tratados en HPBO. Se encontró que la disfunción infecciosa fue más frecuente en menores de 1 año, la disfunción mecánica en mayores 5 años (64% y 38% respectivamente $p < 0.002$). También, la permanencia del SDVP < de 1 año fue más frecuente en disfunciones infecciosas y la permanencia de 1 a 5 años se relacionó más a disfunción mecánica (72 y 46% respectivamente $p 0.03$). La disfunción de catéter distal fue más importante en etiología mecánica y mixta (65,41 respectivamente $p < 0.001$) y la de catéter ventricular en etiología infecciosa (81% $p < 0.001$). No se encontró asociación independiente de la DV con la etiología de hidrocefalia, el lugar de catéter ventricular o con la escala de Lansky.

Conclusiones: La disfunción valvular en paciente pediátrico hidrocefálico es una complicación importante que amerita mayor investigación.

Palabras clave: Hidrocefalia, Catéteres, Procedimientos Neuroquirúrgicos, Hospitales Pediátricos. (Fuente: DeCS Bireme)

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Ventriculo-peritoneal shunt is the historically preferred procedure for hydrocephalus ¹. In children, the permanence of ventricular bypass devices, although they avoid the consequences of hydrocephalus on mortality and delayed psychomotor development, also represent an important comorbidity factor. ² They are causes of hospital readmission due to malfunction and infection, increase in-hospital costs ³ and have a considerable impact on the quality of life of the patient and their caregivers. ⁴ In Ecuador there are no documented reports on valve dysfunction in pediatric age.

The strategies to reduce reoperation due to malfunction of the ventriculo-peritoneal shunt (VPS) or valve dysfunction are numerous, such as: use of devices with new mechanisms that favor the flow of cerebrospinal fluid, devices with impregnation of antibiotics, as well as the use of endoscopy and stereotaxy; however, none has been accurately effective in solving the malfunction. The bibliography describes a great variability of malfunctioning situations and multiple symptoms, the common denominator being the high prevalence in pediatric centers. ²

Valve dysfunction is described as a dynamic entity, often not evident and late diagnosed, sometimes being the cause of prolonged hospitalization. ³ The most representative studies aimed at finding predictive and risk factors are: Shunt Design Trial (1998) ⁵, the Endoscopic Shunt Insertion Trial (2003) ⁶, which describe that the type of device is not related to the presence of dysfunction.

The Shunt malfunction in pediatric hydrocephalus (2015) ⁷ study is multicenter-prospective and demonstrates that early age, early endoscopic use, and cardiac comorbidity are risk factors for malfunction. The rest of the most relevant retrospective studies are heterogeneous and highlight etiology and age as the factors most associated with malfunction ^{8,9}; The experiences of other centers (non-multicenter, non-randomized studies) describe risk factors, most of them conclude that the risk factors are related to the experience and management of each center, being difficult to extrapolate. ⁸⁻¹⁰

Next, we describe the characteristics of patients with malfunction of the ventriculoperitoneal shunt system over 3 years of experience in the Pediatric Neurosurgery Service of the Hospital Baca Ortiz, Pediatric Reference Center in Ecuador. The objective is to determine the types of valve dysfunction and its most frequent variables.

METHODS

Population studied

The inclusion criteria of this descriptive observational clinical study were patients using valve bypass systems with a diagnosis of: "dysfunction", "malfunction" and "failure" of the system in children younger than 15 years and 11 months at Baca Ortiz Pediatric Hospital (BOPH), over a 3-year period, from August 2016 to August 2019. The excluded patients were those who had a diagnosis of hydrocephalus and were not users of a valve shunt. Likewise, patients

whose data in the medical record were confusing or insufficient were eliminated.

Data collection and management

The data collection was developed through a form with instructions prepared by the authors, after training medical professionals who compiled clinical records and operating protocols. The data were tabulated in a protected database with restricted access in the BOPH Neurosurgery Service system.

Primary study results

The need for hospitalization and surgical intervention, which included a revision of the referral system or its withdrawal, was considered as a malfunction of the VPS. A mechanical dysfunction was considered if an alteration in the hardware, an alteration in the position or integrity of the path of the VPS was described in the operative protocol findings.

Infectious type dysfunction was considered if the following were recorded in the clinical history and laboratory results: identification of organisms in Gram stain or CSF culture, abdominal wound or injury, exposure through discontinuity of the skin of any bypass system segment, positive blood culture in the presence of ventricular-atrial system; and mixed dysfunction was considered if it presented the criteria for both mechanical and infectious dysfunction, as well as the presence of an abdominal pseudocyst.

Demographic variables

They included sex, age (polytomized according to the authors' consensus), ethnicity, residence (defined by the political territorial division of Ecuador) and socioeconomic risk (defined by WHO and PAHO)

Clinical history variables

Nutritional history (all hospitalized patients have a nutritional diagnosis in the medical history), psychomotor development (all patients have an assessment and neurodevelopmental test in the medical history), clinical history (consensus of authors according to the most common comorbidities), hospital stay at the time of the placement of the referral system that presented malfunction, a history of infections related to a previous surgical act (data presented in the clinical and surgical history of the medical history) and the Lansky Scale (validated scale that assesses the percentage of global functionality in pediatric patients).

Design variables

Shunt failure / dysfunction / malfunction was defined as symptoms and signs caused by disturbance in the normal flow of cerebrospinal fluid in the shunt device evidenced in reports of surgical protocols.

The previously defined important risk factors, as well as the significant factors in the univariate analyzes, were analyzed looking for independent associations by means of a statistical analysis for the multi-categorical variables and as a difference in prevalence for qualitative variables.

RESULTS

Of the 376 cerebrospinal fluid bypass system placement surgeries, performed from August 2016 to August 2019, 71 cases presented dysfunction. Of these, 5 cases did not meet the inclusion criteria and 6 cases were eliminated for not meeting the conditions proposed for the study, leaving 60 cases finally. Of these, 48 cases were treated at the BOPH and 12 cases at another hospital institution.

Regarding age, age less than 1 year had a higher incidence of valve dysfunction, being 31.67%; Likewise, poor nutritional status and low socioeconomic level influenced their presentation, being 76.67% and 61.67% respectively (Table 1)

The studied patients presented a Lansky scale of 70 (38.33%), which is related to the presence of chronic neurological sequelae (40%) because of their different base pathologies. (Table 1)

The etiology of hydrocephalus in children operated on using an VPS was diverse, with the post-hemorrhagic etiology being 31.67%, post-infection 18.33%, congenital 20%, related to myelomeningocele 16.67%, and tumor obstruction 13.33%. (Table 2)

Regarding the time of use of the valve before dysfunction ("valve age"), this occurred in 53.33% of cases before the year of its placement, with mechanical dysfunction being the one with the greatest presentation with 43.33% , followed by infectious disease with 36.67% and mixed dysfunction with 20%. (Table 2)

Of the 60 valve dysfunctions, 50.67% had no previous failures in valve function; on the other hand, the difference between proximal vs. distal catheter malfunction was not relevant (45% vs. 46% respectively). Exposure of the valve system through the skin was a frequent cause of

TABLE 1: Variables related to valve dysfunction in pediatric patients operated in the Neurosurgery Service of the Baca Ortiz Pediatric Hospital, Quito - Ecuador, 2016-2019.

Variables	Frequency (%) N = 60
DEMOGRAPHIC VARIABLES	
Male	35 (58.3)
Age (years)	
Under 1 year old	19(31.67)
1-5	17(28.33)
6-10	15 (25)
11-15	9 (15)
Ethnic "Mestizo"	51(85)
Rural origin	30 (50)
Socio-Economic Risk Presence	37 (61.67)
CLINICAL BACKGROUND VARIABLES	
No presence of nutritional pathology	46 (76.67)
Disruption of psychomotor development	41 (68.33)
Clinical history	
Chronic multiple neurological sequelae	24 (40)
Epilepsy and epileptic syndromes	5 (8.33)
Chronic, recurrent infections	4 (6.67)
Polimaleformaciones	4 (6.67)
Cancers and tumors	3 (5)
Hormonal	1 (1.67)
No clinical history	19 (31.67)
Hospital stay	
Short	30(50)
Prolonged	21(35)
Medium	9(15)
History of deep infections at the surgical wound site	19 (31.67)
Lansky's Scale of Functionality	
50	6 (10)
60	13 (21.67)
70	23 (38.33)
80	10 (16.67)
90	6 (10)
100	2 (3.33)

Source: Base data from the Neurosurgery Service of Baca Ortiz Pediatric Hospital

hospitalization (28% of cases), followed by fractures, disconnections, perforations, and kinks in the system.

Likewise, valve migration through natural orifices occurred in 12% through the rectum, 4% in the scrotum due to inguinal hernias and 16% in other extraperitoneal migrations. Of all the cases cited, 35% required temporary placement of external ventricular drainage prior to placement of the definitive valve system. (Table 2)

In relation to valve dysfunctions of infectious origin, it was found that the germ with the highest presentation was Staphylococcus aureus (40.90%), followed by Escherichia coli (18.18%), Staphylococcus epidermidis and Candida albicans (13.63% each).

In mixed dysfunctions, Staphylococcus aureus was also the most frequent (41.66%), this type of dysfunction was associated with the presence of an abdominal pseudocyst in 11.67% of cases. In the isolation of germs in mixed dysfunction, a case of Candida albicans, one from Enterobacter, one from Pseudomonas aeruginosa, one from Staphylococcus epidermidis and one from Streptococcus milleri were considered in the “others” group. In dysfunction of infectious type, one case of Pantoea agglomerans and one case of Acinetobacter baumannii were reported. The antibiogram profile of all the isolated organisms was multisensitive in 80.77% of the cases. (Table 3)

TABLA 2: Variables related to hydrocephalus and valve placement in pediatric patients operated in the Neurosurgery Service at Baca Ortiz Pediatric Hospital, Quito - Ecuador, 2016-2019.

Variables	Frequency (%) N = 60
Hydrocephalus etiology	
Poshemorrágica	19 (31.67)
Posinfecciosa	11 (18.33)
Related to myelomeningocele	10 (16.67)
Congenital	12 (20)
Tumoral	8 (13.33)
Valve usage time	
Less than 1 year	32 (53.33)
1-5 years	21 (35)
6 to 10 years	7 (11.67)
Type of dysfunction	
Mechanical	26 (43.33)
Infecciosa	22 (36.67)
Mixedin	12 (20)
Number of dysfunctions	
No previous dysfunctions	34 (50.67)
Two dysfunctions	20 (33.33)
More than 3 dysfunctions	6 (10)
History of use of external drainage	
Si	21 (35)
Noo	39 (65)
Place of dysfunction	
Proximal catheter	27 (45)
Distal Catheter	28 (46.66)
Mixed	5 (8.33)
Ventricular system integrity damage	
Other obstructions	8 (32)
Exhibition	7 (28)
Fracture	4 (16)
Disconnection	3 (12)
Drilling	2 (8)
Acodamiento	1 (4)
Valve system placed in another hospital	
Si	12 (20)
Noo	48 (80)
Distal catheter migration	
Through the rectum	3 (12)
Through the scrotum	1 (4)
Extraperitoneal	4 (16)

Source: Base data from the Neurosurgery Service of Baca Ortiz Pediatric Hospital

TABLE 3: Variables of valvular dysfunction according to infectious and mixed etiology in paediatric patients operated in the Neurosurgery Service of the Baca Ortiz Pediatric Hospital, Quito-ecuador, 2016-2019.

Variables	Frequency (%) N = 60
Isolated organism in infectious dysfunction	No.22
<i>S. aureus</i>	9 (40.09)
<i>E. coli</i>	4 (18.18)
<i>S. epidermis</i>	3 (13.63)
<i>C. albicans</i>	3 (13.63)
Other	3 (13.63)
Isolated organism in mixed dysfunction	No.12
<i>S. aureus</i>	5 (41.66)
Other	7 (58.33)
Antibiogram profile of isolated organisms	
Multisensible	21 (80.77)
Collection development	No.17
Pseudoquiste abdominal	7 (11.67)
Subgaleal	4 (6.67)
Extrabdominal	2 (3.33)
Other	4 (6.68)

Source: Base data from the Neurosurgery Service of Baca Ortiz Pediatric Hospital

DISCUSSION

During the 3 years of the study, 376 valve bypass surgeries were performed, 71 valve dysfunctions were evident, of which 60 met inclusion criteria, 48 being treated in our hospital (BOPH) and 12 in other hospitals.

As in reports from previous multicenter studies, the etiology of hydrocephalus was not a factor related to dysfunction, however, as in most studies, dysfunctions were mostly associated with etiologies that produce communicating hydrocephalus.^{2,10} Regarding age and etiology, our findings are similar to the trend in other studies, which show that the younger the patient is, the more frequent the etiology associated with communicating hydrocephalus, and within this, the hydrocephalus associated with hemorrhage. Thus, Tuli et al. found that age less than 1 year was a risk factor for valve dysfunction.¹⁰ Piatt and Carlson found that age is an independent risk factor for malfunction ($p < 0.001$)¹¹. In our research, age less than one year was related to infectious dysfunction ($p = 0.002$).

Previous studies show that valve dysfunction occurs more frequently within two years after surgery (40-50% failure).² In our experience, valve dysfunction occurs more frequently within one year after placement of a valve, particularly in cases of dysfunction of infectious etiology.

Likewise, we found results that were different from those of several cited series, in these studies it is mentioned that the mechanical causes of dysfunction can occur at any age and that the valve age (time of dysfunction since valve placement) is generally related to late dysfunction^{2,7}. In our study, mechanical dysfunction was significantly associated

with the age of 6 to 10 years; In turn, valve age (dysfunction time) was in the range of 1 to 5 years, with a low incidence of hyperdrainage (1 case), unlike other studies that mention it as one of the most frequent causes of mechanical dysfunction. (3.4%)^{2,9}

In the medical literature it is also reported that factors such as malnutrition associated in turn with hypoalbuminemia, decreased oncotic pressure and the development of ascites and peritoneal insufficiency, hydrocephalus related to myelomeningocele with scoliosis, and mechanical changes such as short catheter, bleeding during surgery and fibrosis, and the causative agent of infectious dysfunction due to *S. epidermidis* were strongly present in the dysfunction^{2,7,9,12,13}. In contrast, our experience with these factors was in a few cases.

Other intraoperative factors such as time or intraoperative bleeding mentioned in some series¹⁴ were also not frequent in our study.

Regarding infectious data, in our study it was found that 35% had a history of external diversion, and *Staphylococcus aureus* was also the etiological agent in cases of dysfunction of infectious etiology in contrast to *S. epidermidis* mentioned in the bibliography¹⁵.

Finally, we did not report an association or a higher frequency of valve dysfunction in factors mentioned in other studies such as: cardiovascular factors, endoscopic procedures, 6 which were not considered in our findings.

CONCLUSION

In our study we found as relevant findings that the patient's age less than 1 year, the elapsed time of valve placement (valve age) less than one year, and proximal catheter dysfunction are statistically significant characteristics more related to infectious dysfunction; while the age of 6-10 years, the valve age of 1-5 years and distal catheter dysfunction are more frequent in mechanical dysfunction. Mixed dysfunction (infectious and mechanical) is predominantly associated with school-age patients, valve age less than 1 year and up to 5 years), dysfunction located in the distal catheter and dysfunction of both catheters.

In the placement of a valve in lactating patients, infectious prophylaxis protocols should be discussed, while at older ages, measures that prevent mechanical dysfunction should be considered and emphasized.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

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