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**REVISTA DE NEUROCIRUGÍA**

**2024**



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# ÍNDICE

- Mensaje del Presidente.
- Assessing pediatric neurosurgery capacity in La Paz, Bolivia: an illustrative institutional experience of a lower-middle-income country in South America.
- Pediatric neurosurgical medulloblastoma outcomes in La Paz, Bolivia: How a Lower Middle-Income Country (LMIC) institution in South America compares to the United States.
- Surgical Management Of Patients with Giant Vestibular Schwannomas (T4 A And B Hannover Classification): Analysis Of 36 Cases.
- Abdominal cerebrospinal fluid pseudocyst: a comparative analysis between children and adults.
- Cirugía de Epilepsia en Bolivia
- Manejo del hematoma subdural agudo con la técnica enrejado de duramadre en la Caja Nacional de Salud del 2005 al 2019

# MENSAJE DEL PRESIDENTE

## XXII CONGRESO BOLIVIANO DE NEUROCIRUGÍA BODAS DE ORO

*“La vida es siempre un millón de probabilidades. Esto hace la garantía y la justificación del arte”. Franz Tamayo*

La Paz – Bolivia 28 – 29 – 30 de agosto 2024

Carta del Presidente.



Distinguidos colegas de Bolivia, Latinoamérica y el mundo:

La Neurocirugía Boliviana cumple 50 años de fundación, son medio siglo de historia y de trabajo esforzado y está ahora en una posición privilegiada, al recibir a grandes referentes de la Neurocirugía; el esfuerzo de cada uno de sus miembros por salvar la vida a sus pacientes, cumple con los más altos niveles técnicos y éticos para lograr ese fin, somos un país y una sociedad que se preocupa de mejorar cada vez nuestros niveles de atención, con la firme convicción de formar futuras generaciones que cumplirán estos objetivos.

Somos los guerreros llamados a combatir con nuestras manos y conocimiento la realidad mundial en el ámbito de las enfermedades quirúrgicas del sistema nervioso y en estos tiempos donde el conflicto entre seres humanos – la guerra – toca nuestras puertas y pone en alerta a los Neurocirujanos del planeta para enfrentar grandes retos.

Definitivamente, la Neurocirugía, siendo el arte de abordar las estructuras más nobles del ser humano y estas por supuesto la creación de Dios, necesita del trabajo médico dedicado a esta obra perfecta con la participación de especialidades como el Neurointensivismo, Traumatología, Neurología, Psiquiatría, Anestesiología, Otorrinolaringología, Pediatría, entre otras, cumpliendo el verdadero trabajo en equipo. Por esa razón nuestro mayor reconocimiento a los médicos involucrados en esta tarea y que hoy nos acompañan de la mano en este evento.

Hay regalos que no se compran, hay regalos que no tienen precio y ellos son la vida y funciones de nuestros pacientes, son regalos supremos que tenemos que conservarlos en los que en determinado momento nos necesitan. Quiero agradecer a la directiva de la Sociedad Boliviana, que con trabajo y esfuerzo hicieron posible recibirlos en el XXII congreso Boliviano de Neurocirugía Bodas de oro, La Paz

Bolivia 2024, para ellos y todos los Neurocirujanos Bolivianos mi mayor reconocimiento, con una mención especial a la Sociedad Paceña de Neurocirugía. Agradecemos a las empresas que nos apoyan, las grandes instituciones como la FLANC, WFNS y a todos los que hacen posible este encuentro científico – social.

Queremos recordar a los Neurocirujanos Bolivianos que a lo largo de esta historia de 50 años ya no están con nosotros, pero su legado perdura y será siempre el fuego que nunca se apaga porque vivirán en nuestras páginas escritas por el destino con el presente y futuro de nuestra Sociedad Boliviana de Neurocirugía (SBN).

Bienvenidos colegas del mundo, La Paz declarada ciudad maravilla acoge 40 Neurocirujanos activos, que cuidan de una población de cerca 5 millones de habitantes, vivimos en comunidad, somos cálidos, afectuosos, empáticos y sobre todo creyentes de que nuestras vidas y la de nuestros pacientes son voluntad divina.

Gracias al Comité organizador del congreso, gracias a la Federación Mundial de Neurocirugía, gracias a la Federación Latinoamericana de Neurocirugía por apoyar y acompañar los logros de la Sociedad Boliviana de Neurocirugía, que Dios bendiga nuestras manos y nuestro cerebro para convertirnos en instrumentos de vida de la especie humana.

Muchas gracias.



# Assessing pediatric neurosurgery capacity in La Paz, Bolivia: an illustrative institutional experience of a lower-middle-income country in South America

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**OBJECTIVE** The current pediatric neurosurgery capacity in lower-middle-income countries (LMICs) in South America is poorly understood. Correspondingly, the authors sought to interrogate the neurosurgical inpatient experience of the sole publicly funded pediatric hospital in one of the largest regional departments of Bolivia to better understand this capacity.

**METHODS** A retrospective review of all neurosurgical procedures performed at the Children’s Hospital of La Paz, Bolivia (Hospital del Niño “Dr. Ovidio Aliaga Uriá”) between 2019 and 2023 was conducted after institutional approval using a recently implemented national electronic medical record system.

**RESULTS** A total of 475 neurosurgical admissions satisfied inclusion for analysis over the 5-year span. The majority of admissions were from within the La Paz Department (87%) via the emergency department (77%), without private insurance (83%). The most common indications for neurosurgical intervention were trauma (35%), followed by hydrocephalus (28%), congenital disease (12%), infection (5%), and craniostomy (3%). Overall, the median age at time of surgery was 2.0 years, and the median operating time was 1.5 hours with a minority of intraoperative complications (2%). The most common inpatient complication was unplanned return to the operating room (19%), most commonly seen in congenital indications. At final discharge, the median postoperative length of stay was 10 days. Twenty-seven (6%) of the 475 patients died during hospitalization, most commonly seen in tumor indications. Of the 448 patients who were discharged, 299 (67%) returned for at least one follow-up appointment.

**CONCLUSIONS** There is restricted breadth in neurosurgical indications and outcomes achievable at the Children’s Hospital of La Paz, Bolivia. As such, the capacity of pediatric neurosurgery at institutions in LMICs in South America such as this one is very limited. Identifying and prioritizing actionable interventions to improve this capacity is institution- and LMIC-dependent, and as such, future efforts will need to be tailored appropriately.

<https://thejns.org/doi/abs/10.3171/2024.3.PEDS24126>

**KEYWORDS** pediatric neurosurgery; hydrocephalus; La Paz; Bolivia; South America; lower-middle-income country; LMIC; global surgery

**B**OLIVIA is a landlocked country in South America with a population of approximately 12 million inhabitants. Per the World Bank, it is officially a lower-middle-income country (LMIC) whose gross domestic product per capita ranks 96th in the world and the Human Development Index ranks 114th in the world.<sup>1-3</sup> Given how epidemiological studies have shown pediatric neurosurgical conditions such as hydrocephalus,<sup>4,5</sup> neural tube defects,<sup>6</sup> and tumors<sup>7</sup> can vary according to parameters related to these macroeconomic rankings, there is a need then to understand how pediatric neurosurgery fares from

resource and capacity perspectives at institutional levels in LMICs such as Bolivia (Fig. 1A).

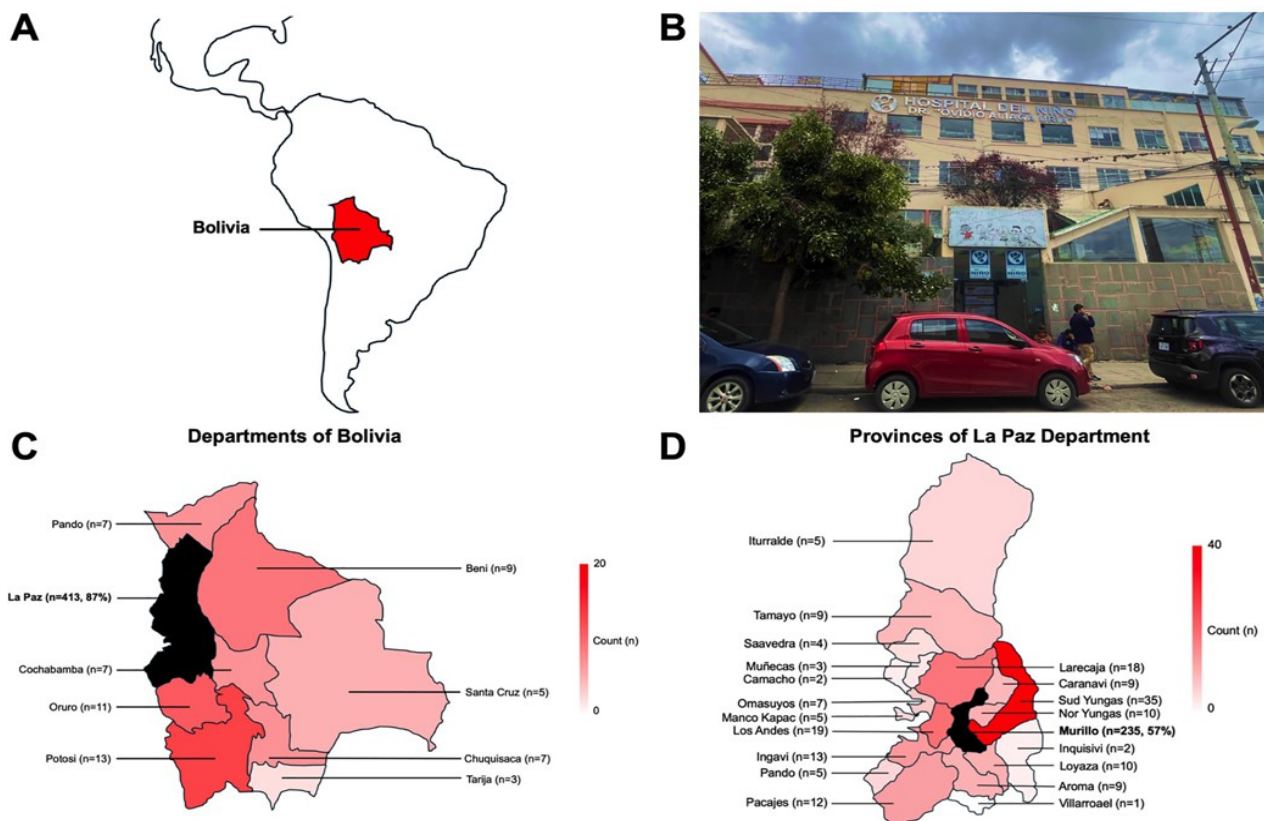
Pediatric neurosurgery in Bolivia is limited to a handful of specialized centers in the country.<sup>8</sup> The Hospital del Niño “Dr. Ovidio Aliaga Uriá” in La Paz, Bolivia, is one such center (Fig. 1B). It was founded in 1972 and offers multiple tertiary pediatric surgical specialties including neurosurgery, cardiothoracic surgery, orthopedic surgery, and general surgery.<sup>9</sup> The hospital serves as the sole publicly funded pediatric hospital in the La Paz Department, a region with approximately 2.9 million inhabitants of

**ABBREVIATIONS** ED = emergency department; EMR = electronic medical record; HIC = high-income country; LMIC = lower-middle-income country; LOS = length of stay; OR = operating room; SICE = Sistema de Información Clínico Estadístico; SUS = Sistema Unico de Salud.

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**FIG. 1.** Bolivia is a landlocked country in South America (A) where the Hospital del Niño “Dr. Ovidio Aliaga Uriá” is located in La Paz (B). The hospital services all patients, including patients from all 9 departments of the country (C), and all 20 provinces within the La Paz Department in which the hospital is located in the Murillo Province (D). All maps were created using software Paint (Microsoft). The photograph is courtesy of the first author V.M.L. Figure is available in color online only.

whom 21% are estimated to be aged 18 years or younger.<sup>10,11</sup> The hospital itself has 4 operating rooms (ORs) and 212 inpatient beds in total, of which there are 8 functional pediatric ICU beds shared across all specialties, 12 neonatal ICU beds, and 11 dedicated neurosurgical (neuroscience) floor beds.

Since the last decade, the Department of Neurosurgery at the Hospital del Niño has been serviced by two board-certified attending neurosurgeons. There is no mandatory neurosurgery residency rotation at the hospital; however, both local and national neurosurgery residents have the option to participate as a mid- to senior-level resident on away rotation if desired.

Many resources in the operative workup and surgery for neurosurgical patients are extremely limited at the Hospital del Niño. With respect to imaging, there is no MRI available, and the only CT scanner is very commonly not functional or available. Within the OR, there is no operating microscope or endoscope available for use. Finally, the limited size of the ICU that is shared between all surgical services limits the inflow of operative cases requiring ICU-level care after neurosurgery.

To date, it is unclear as to the neurosurgical capacity at

the Hospital del Niño. Efforts to audit and review surgical outcomes have been limited within this institution, like many other poorly resourced institutions in the LMICs of South America, due to inconsistencies in how data can be reported and stored.<sup>8,12</sup> We sought to perform the first-ever retrospective review of all neurosurgical patients at the Hospital del Niño “Dr. Ovidio Aliaga Uriá,” La Paz, Bolivia, using a recently implemented national electronic medical record (EMR) system to shed light on the pediatric neurosurgical capacity of institutions like this in LMICs in South America.

## Methods

### Data Collection

Primary data were retrospectively obtained from the national EMR system Sistema de Información Clínico Estadístico (SICE) at the Hospital del Niño “Dr. Ovidio Aliaga Uriá,” La Paz, Bolivia, after institutional approval. In the SICE, inpatient and operative outcomes are manually entered by physicians on the care team for each patient. All laboratory values and hospitalization reports are accessible, including operative reports, pathology reports,

and radiology reports. Patient data in the SICE can be queried based on department, diagnosis, and procedure bookings. For this study, patients were identified by searching all procedure bookings under the Department of Neurosurgery between 2019 and 2023.

## Selection Criteria

Selection focused on inpatient outcomes following neurosurgical intervention during a hospital admission. Inclusion criteria were 1) patients aged 18 years and younger, 2) who underwent at least one neurosurgical procedure during admission, and 3) with a documented operative report. Exclusion criteria were 1) patients with scheduled neurosurgery that was ultimately not performed, and 2) admissions with incomplete documentation.

## Outcomes

In addition to sociodemographic parameters of all patients, the primary outcomes of interest were types of neurosurgical procedures performed during admission, intraoperative complications, duration of surgery, postoperative length of stay (LOS), complications during hospitalization, status at time of discharge, and follow-up. Secondary outcomes included nature of hospital admission, insurance status, and the need for further neurosurgical procedures after index admission. For summative purposes, all neurosurgical procedures were classified as congenital (including neural tube defects and congenital intracranial cysts), craniosynostosis, hydrocephalus, infection, trauma, tumor, or miscellaneous.

## Results

### Overall Admissions

A total of 475 admitted patients with neurosurgical procedures at the Hospital del Niño, La Paz, Bolivia, satisfied all requirements for selection into the study cohort (Table 1). Twenty-one patients were excluded because of incomplete documentation. The majority of included patients were from the La Paz Department (413/475, 87%), within which the majority resided in the Murillo Province (235/413, 57%), where both La Paz and El Alto cities are located (Fig. 1C and D). Nevertheless, all Bolivian departments and La Paz provinces were represented in the overall cohort, indicating the wide networks from which the hospital draws its patient population.

The most common nature of admission was through the emergency department (ED; 368/475, 77%) compared with patients presenting electively to the hospital (107/475, 23%), with more ED patients having been transferred from another hospital (206/368, 56%) than those presenting primarily (162/368, 44%) (Fig. 2A). Across the 5-year span, the most admissions occurred in 2019 (124/475, 26%), and the least admissions were in 2021 (50/475, 11%) (Fig. 2B). The low admission rate in 2021 was associated with the worldwide COVID-19 pandemic and local government shutdowns of nonemergent hospital services,<sup>13</sup> and this was evident by the greatest proportion of admissions through the ED in the year 2021 (39/50, 78%).

Finally, the majority of patients presented with Sistema Unico de Salud (SUS) insurance (394/475, 83%), an

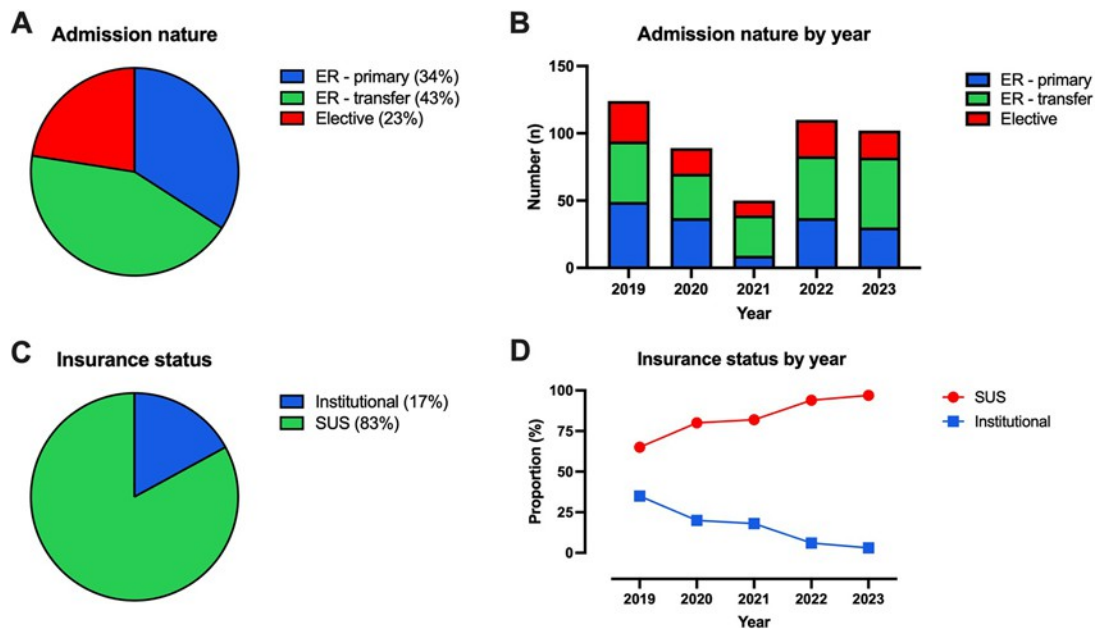
**TABLE 1. Summary of inpatient characteristics for 475 patients seen at Hospital del Niño, La Paz, Bolivia, between 2019 and 2023**

Characteristic	Value
<b>Residence</b>	
w/in La Paz Department	413 (87)
w/in Murillo Province*	235/413 (57)
Outside Murillo Province	178/413 (43)
Outside La Paz Department	62 (13) Admission route
ED	368 (77)
Transfer	206/368 (56)
Primary	162/368 (44)
Elective	107 (23)
<b>Insurance status†</b>	
SUS	394 (83)
Institutional	81 (17)
Time from admission to op, days	2 (1–6) Surgical indication
Trauma	165 (35)
Hydrocephalus	131 (28)
Congenital	59 (12)
Tumor	52 (11)
Infection	23 (5)
Craniosynostosis	16 (3)
Miscellaneous	29 (6)
Age at op, yrs	2.0 (0.3–6.9)
<b>Sex</b>	
Male	257 (54)
Female	218 (46)
Op time, hrs	1.5 (1.0–2.0)
Intraop complications	9 (2)
<b>Postop hospitalization</b>	
Postop imaging	130 (27)
Unplanned return to OR	90 (19)
Shunt failure	17/90 (19)
Postop infection	54 (11)
Inpatient mortality	27 (6)
LOS, days	10 (6–21)
<b>Follow-up</b>	
Return to ED in 30 days	37/448 (8)
Readmitted for neurosurgery	11/37 (30)
≥1 follow-up	299/448 (67)
Mean follow-up time, mos	4.1 (0.7–12.9) Final status
Alive	283/299 (95)
Dead	16/299 (5)

Continuous variables are presented as median (IQR) and categorical variables are presented as count (percentage of total cohort) unless otherwise stated.

\* Hospital del Niño is located within Murillo Province within La Paz Department.

† SUS is the equivalent to universal healthcare insurance, and institutional insurance is the equivalent to private insurance.



**FIG. 2.** Proportion of admission route overall (A) and by year during 2019–2023 (B). Proportion of insurance status overall (C) and by year (D). SUS insurance is the equivalent to universal healthcare insurance, and institutional insurance is the equivalent to private insurance. Figure is available in color online only.

equivalent to universal healthcare insurance in high-income countries (HICs), and a minority presented with institutional insurance (81/475, 17%), which is the equivalent to private insurance in HICs (Fig. 2C). Within the recent 5-year span, the proportion presenting with SUS insurance continued to increase (Fig. 2D). Similarly, the median time from admission to surgery was 2 (IQR 1–6) days, increasing from 1.9 days in 2019 to 2.5 days in 2023.

## Neurosurgical Indications

Primary neurosurgical indications were related most commonly to trauma (165/475, 35%), followed by hydrocephalus (131/475, 28%), congenital disease (59/475, 12%), tumor (52/475, 11%), infection (23/475, 5%), craniosynostosis (16/475, 3%), and miscellaneous (29/475, 6%) (Fig. 3A). Collectively, trauma and hydrocephalus indications accounted for the majority of all indications for neurosurgery, as well as for each individual year (Fig. 3B).

For congenital indications, the most common diagnosis was myelomeningocele (28/59, 47%), followed by lipomeningocele (10/59, 17%), arachnoid cyst (10/59, 17%), and encephalocele (7/59, 12%). For craniosynostosis indications, the most common diagnoses were plagiocephaly (7/16, 44%), brachycephaly (3/16, 19%), and scaphocephaly (2/16, 12%). For hydrocephalus indication, the most common diagnoses were congenital hydrocephalus (76/131, 58%), shunt failure (31/131, 24%), and hydrocephalus not otherwise stated (16/131, 12%). For infection indications, the most common diagnoses were subdural empyema (10/23, 43%), cerebral abscess (6/23, 26%), and epidural abscess (3/23, 14%). For miscellaneous indications, the most common diagnoses were pilonidal cyst (7/29, 24%), dermoid cyst (6/29, 21%), and hemangioma (4/29, 14%). For trauma indications, the most common

diagnoses were epidural hematoma (102/165, 62%), subdural hygroma (19/165, 12%), and depressed skull fracture (19/165, 12%). Finally for tumor indications, the most common diagnoses were medulloblastoma (11/52, 21%), pilocytic astrocytoma (8/52, 15%), and ependymoma (7/52, 13%).

## Surgery Details

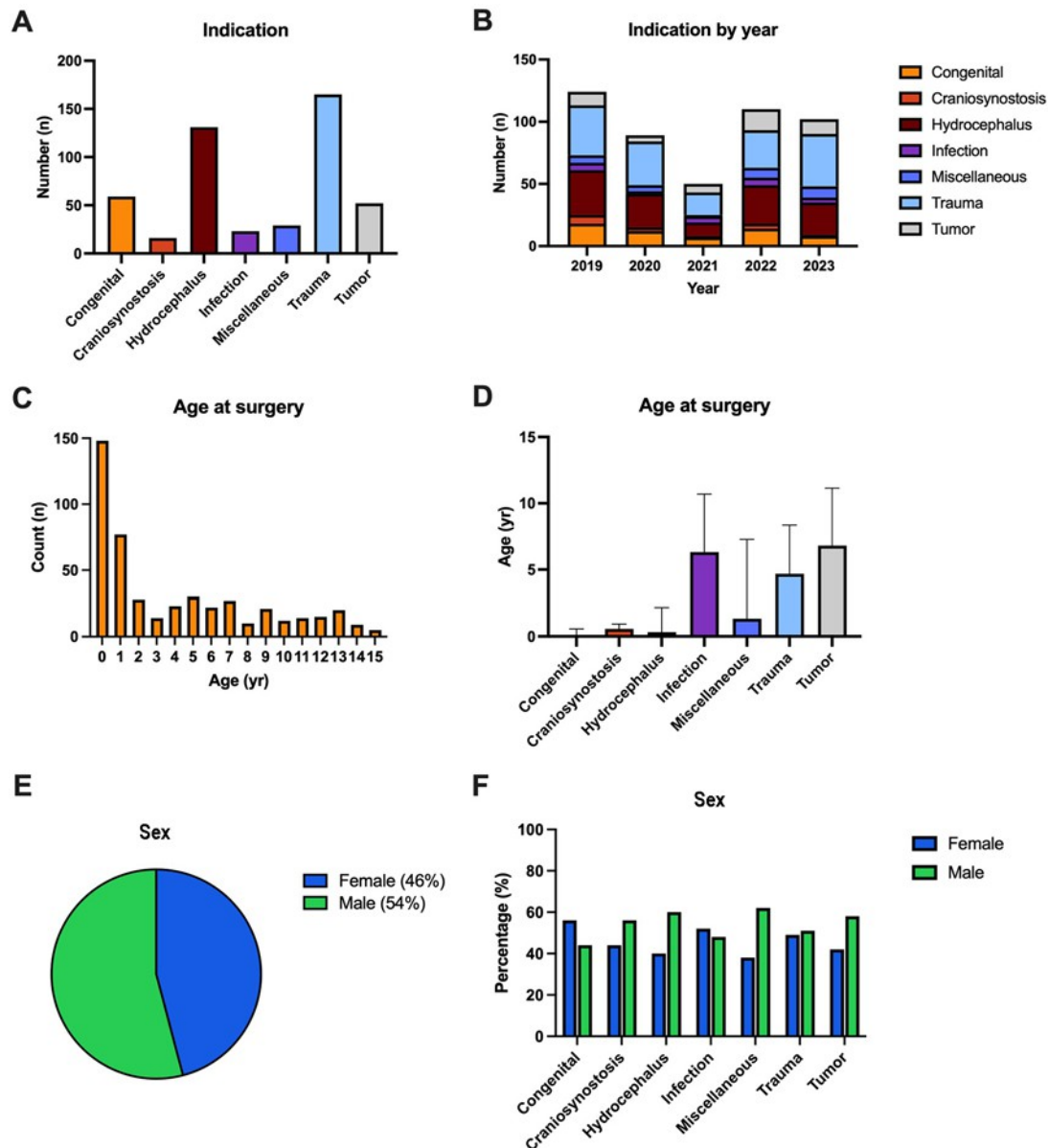
Within the cohort, the median age at the time of surgery was 2.0 (IQR 0.3–6.9) years (Fig. 3C). The indication with the youngest median age was congenital (newborn), and the indication with the oldest median age was tumor (6.8 years) (Fig. 3D). In terms of sex, there were 218 (46%) female and 257 (54%) male patients (Fig. 3E). The only indications for which there were more female than male patients by proportion were congenital (56% vs 44%) and infection (52% vs 48%) (Fig. 3F).

The most common neurosurgical procedures performed were craniotomy (184/475, 39%), followed by shunting (142/475, 30%), neural tube defect closure (45/475, 9%), craniectomy (38/475, 8%), and peripheral lesion excisions (29/475, 6%).

In terms of surgery, the median operative time was 1.5 (IQR 1.0–2.0) hours; there were no intraoperative mortality events and only a minority of intraoperative complications (9/475, 2%), none of which required additional intervention. The indication with the longest median OR time was tumor (2.5 hours), and that with the shortest median OR time was miscellaneous (1.0 hours) (Fig. 4A).

## Hospitalization Outcomes

Postoperatively, 130 (27%) of the 475 patients underwent postoperative imaging typically either in-house if the



**FIG. 3.** Composition of neurosurgical indications overall (A) and by year (B). Histogram of age at the time of surgery overall (C) and by indication (D). Overall sex proportions (E) and by indication (F). Continuous data are reported as median (IQR). Figure is available in color online only.

CT scanner was functional or at an adjacent institution. The indication with the highest rate of postoperative imaging was for infection (17/23, 74%) and the lowest rate was for craniosynostosis (0%).

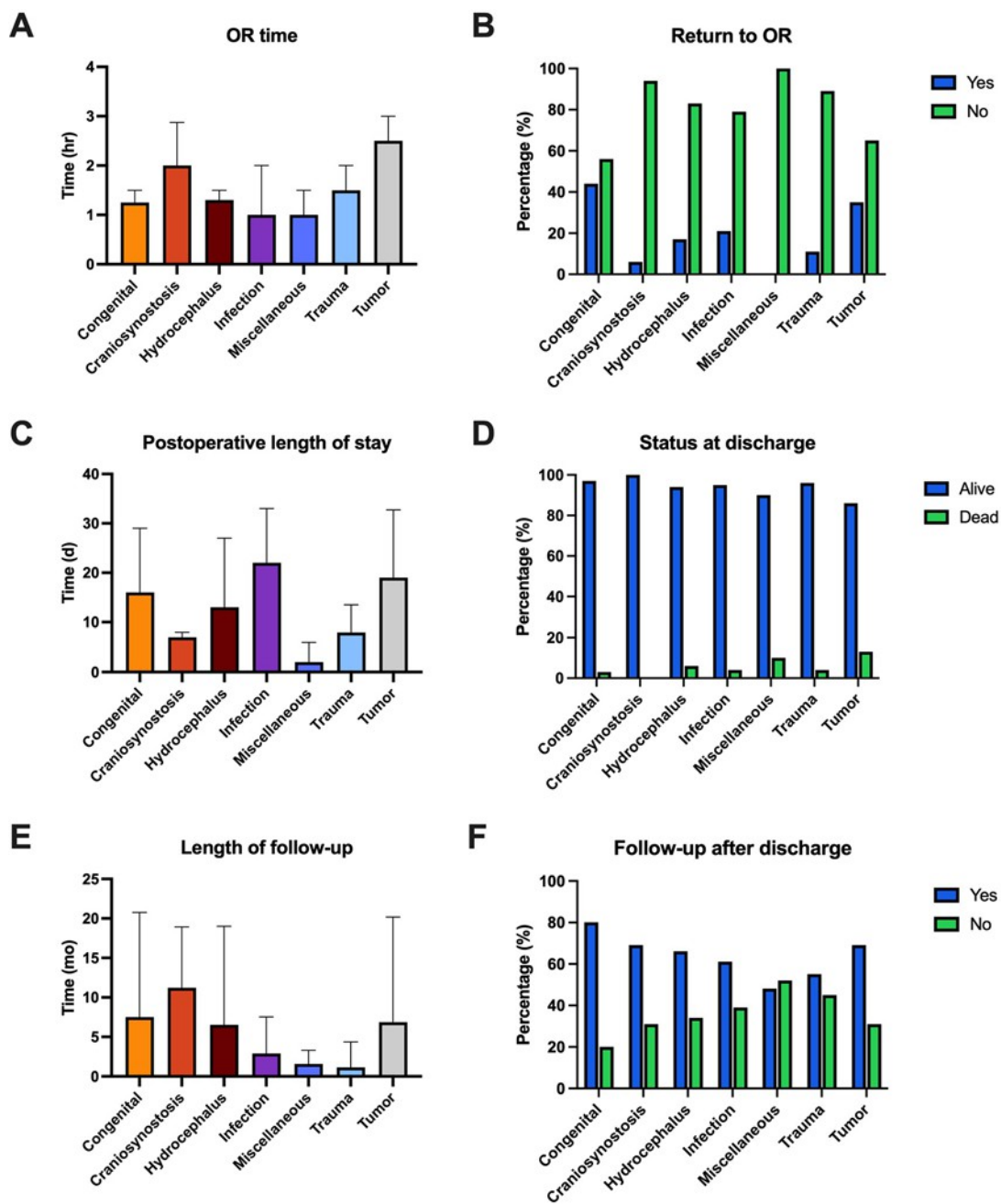
There were 90 (19%) instances in which patients were returned to the OR during the same admission, with 25 (28%) of them returning to the OR more than once during the same admission. Separately, 17 (19%) of those returning to the OR were because of shunt failure. The indication with the highest rate of return to OR was congenital (26/59, 44%) and that with the lowest rate was miscellaneous (0%) (Fig. 4B).

Infections were detected in 54 (11%) of the 475 patients after their index neurosurgery procedure during admis-

sion. The most common infections were systemic bacteremia (18/54, 33%), meningitis (13/54, 24%), urinary tract infection (10/54, 19%), and pneumonia (8/54, 15%). The indication with the greatest rate of infection was congenital (10/59, 17%) and that with the lowest rate was miscellaneous (2/29, 7%). Among the 131 shunt procedures for hydrocephalus, there were 9 (7%) cases of shunt infection requiring surgical revision.

By final discharge, the median LOS after index surgery was 10 (IQR 6–21) days. The indication with the longest median postoperative LOS was infection (22 days) and the shortest was miscellaneous (2 days) (Fig. 4C).

Twenty-seven (6%) of the 475 patients died during hospitalization (Fig. 4D). The remaining 448 (94%) patients



**FIG. 4.** Operative outcomes of all patients by indication. OR time (A), return to OR (B), postoperative LOS (C), status at discharge (D), length of follow-up (E), and follow-up after discharge (F). Follow-up data only relevant to the 448/475 (94%) patients who were alive at time of discharge. Continuous data are reported as median (IQR). Figure is available in color online only.

were ultimately successfully discharged to outpatient status. The indication with the highest rate of inpatient mortality was tumor (7/52, 13%), and the lowest was craniosynostosis (0%).

## Follow-Up Outcomes

Thirty-seven (8%) of the 448 discharged patients returned to the ED within 30 days of discharge, with 11 (30%) requiring subsequent readmission for further neurosurgical intervention. The indication with the highest rate

of return to the ED within 30 days was infection (3/22, 14%), and the lowest was craniosynostosis and miscellaneous (both 0%).

Of the 448 patients discharged, 299 (67%) returned for at least one follow-up appointment with a median follow-up time of 4.1 (IQR 0.7–12.9) months (Fig. 4E and F). The indication with the highest rate of follow-up was congenital (47/59, 80%) and that with the lowest was miscellaneous (14/29, 48%). The indication with the longest median follow-up time was craniosynostosis (11.2 months)

and the shortest was trauma (1.1 months). By last follow-up, 283/299 (95%) of patients were alive, and 16/299 (5%) had died.

## Discussion

The inpatient neurosurgical experience at the Hospital del Niño “Dr. Ovidio Aliaga Uria,” La Paz, Bolivia, has to date never been audited. In our study, we reveal for the first time that many of the outcomes in this South American LMIC institution are quantitatively very different from those of more contemporary experiences from HICs. These differences are in part explainable by the differences in resources available, and to improve capacity and outcomes, many institution- and country-specific initiatives need to be considered.

## A Large Serviceable Population

The surgical indications seen in our contemporary series mirrors those of previous efforts to describe the most common pediatric indications seen in Bolivia as a country.<sup>8</sup> Geographically, our study showed that the majority of patients derive from the most populous province, the Murillo Province (inclusive of the largest metropolitan cities of La Paz and El Alto), where the Hospital del Niño is located. Nevertheless, over the 5-year span, the neurosurgery department serviced patients from all provinces of the La Paz Department, and indeed, from all departments within the country of Bolivia. As such, there is a clear demand throughout the region Hospital del Niño serves for neurosurgery services. This is evident also by the appreciable proportion of ED transfers for neurosurgical admission. In addition, despite the COVID-19 pandemic, our study shows that the patient volume is returning effectively to prepandemic levels, which is not dissimilar to other neurosurgery experiences in HICs.<sup>14,15</sup>

## Surgical Indications

There were some noticeable disparities in some of the surgical indications at Hospital del Niño compared with more typical HIC experiences, likely driven by the limited resources available. Arachnoid cysts have proven amenable to surgical treatment if symptomatic after conservative management, including by endoscopic techniques, in many HIC settings.<sup>16</sup> However, the absence of endoscopic technology and intraoperative imaging modalities at the Hospital del Niño restricts neurosurgeons to open surgery only when a patient is symptomatic. Similarly, plagiocephaly is not typically the most common operative craniosynostosis in HICs, as it often responsive to conservative treatment by orthoses and helmeting;<sup>17</sup> however, in the absence of these orthotic services due to both cost and availability, neurosurgeons are often limited to only open surgery for definitive treatment. Finally of note, pilonidal cysts are not commonly treated by neurosurgeons in HICs. Yet, in the absence of complete imaging services at the Hospital del Niño and surrounding centers, many of these cases are referred to neurosurgery to treat in the possibility the perceived cyst is rather a meningocele or some other extension of the caudal central nervous system.

## Reliance on Insurance

The Bolivian government formally introduced the SUS in 2018,<sup>18</sup> a universal healthcare program similar to that of the Medicaid system in the United States. Being a publicly funded hospital, the Hospital del Niño was an early utilizer of this system. In Bolivia, it is estimated that 50% of the population are users of SUS,<sup>19</sup> which is lower than the 80% seen in the neurosurgical cohort at the Hospital del Niño. We found over the subsequent 5 years that there has been an increasing reliance on this insurance by neurosurgical patients. Correspondingly, there was also possibly a correlative increase in time from initial admission to index surgery, which could be explained by the time approval required for crucial elements of neurosurgical procedures, from preoperative imaging (particularly if needed to be done at outside facilities) to implants such as shunt valves.

## Limited OR Equipment

Without an operative microscope or intraoperative navigation, the capacity to pursue microsurgery, particularly in the setting of brain tumor surgery, is extremely limited. With loupe magnification only, neurosurgeons are limited as to how aggressive they can be intraoperatively for particular indications. The comparable operating times among most indications highlight the limited, macroscopic approach available at hand. Longer-term documented follow-up of indications that can recur after a subtotal resection, such as tumor, will be telling as to the consequential severity of not having microsurgery available in this setting.

Next, the absence of a functional endoscope with effective imaging also likely impacts the ability to perform effective hydrocephalus surgery in many of the infants presenting with congenital hydrocephalus amenable to endoscopic third ventriculostomy; hydrocephalus alone was present in 84 (43%) of the 193 treated patients who were younger than 1 year. Trends in HICs suggest that endoscopic third ventriculostomy can be considered upfront surgery and effective in a large proportion of infants in obviating the need for permanent shunting in the future.<sup>20,21</sup> Unfortunately, the Hospital del Niño is not able to perform such a procedure, and as such, all patients who could otherwise potentially avoid a shunt are treated by shunting.

## Postoperative Outcomes

After surgery, the return to OR rate was 19% with a postoperative infection rate of 11%. This rate is notably higher than that of many large experiences in pediatric neurosurgery in HICs.<sup>22,23</sup> It is worth noting that despite the discrepancy in rates, congenital indications appear to be associated with the highest rate of postoperative infections in both our series and series from HICs.<sup>22</sup> Although within the hydrocephalus shunting subgroup, the overall shunt infection rate during admission of 7% was somewhat comparable to national HIC experiences reported elsewhere within the range of 5%–15%.<sup>24,25</sup> Similarly, the overall surgical site infection rate of 2% was comparable to that of a large HIC pediatric neurosurgery experience.<sup>26</sup> Ultimately, it would appear that greater disparity in postoperative infection rates is more systematic than surgical, although a single source as to this difference is unlikely.

Rather, there are likely many contributors specific to the LMIC setting including variability in sterile technique, surveillance timing, and perioperative antibiotics available.<sup>27</sup>

The median LOS was over a matter of weeks at the hospital, which, among various indications in HICs, is overall higher; for example, the median LOS for tumor cases was 19 days at the Hospital del Niño, which is greater than the mean LOS for tumor patients at various HIC pediatric centers ranging from 2 to 8 days.<sup>23,28,29</sup> Similar to infection rates, this difference in hospitalization duration is likely multifactorial. One factor more prominent in LMICs is that many patients may come from families and households not immediately able or equipped to deal with non-simple postoperative care.<sup>30</sup>

Finally, we note that 6% of patients died during hospitalization, which was as high as 13% among patients with tumor. This is higher than what is reported in HICs, with a large United States pediatric neurosurgery series reporting a mortality rate of less than 1% across all indications.<sup>23</sup> In part, these poorer outcomes mirror outcomes of other LMICs limited by late diagnosis, delayed treatment, and heterogeneous socioeconomic conditions that impact postoperative care.<sup>31</sup> Of note within tumor indications, not all chemotherapy services are coverable for outpatients, leading many tumor patients to experience extended LOS to receive adjuvant therapy, naturally then increasing their risk of developing inpatient infection and other complications that increase comorbidity burden.

## Follow-Up Rates

It is important to note that follow-up rates after surgery were not perfect, and indeed this pales in comparison to some of the published pediatric neurosurgery literature from HICs with perfect follow-up.<sup>27</sup> Indeed, our study showed that only two-thirds of patients returned for at least one follow-up visit. Geography, parental education and understanding, and the financial ability to attend follow-up are all likely factors that contribute to this finding. Given the large catchment area of the La Paz Department and its 20 provinces, as well as the absence of telemedicine technology which has proven effective in many HIC experiences,<sup>14</sup> it is difficult to envisage this rate drastically improving in the near future. A proactive step intrinsic to acknowledging this is that part of the discharge protocol at Hospital del Niño is comprehensive postoperative wound instructions if follow-up does not happen.

## Absence of Functional and Vascular Subspecialties

In many pediatric hospitals in HICs, all neurosurgical subspecialty services are available. This is not the case at Hospital del Niño. Functional surgeries (including epilepsy surgery) and vascular diagnostics such as angiography and open vascular surgery are not available. This is in part due to the lack of resources in the OR, as well as imaging technology, surgical instruments, and medical devices needed to perform such procedures. As it is currently, pediatric patients who require surgical treatment for these pathologies can only pursue treatment in private neurosurgery settings that treat both adults and children.<sup>8</sup> As such, there remains a large deficiency in treating pediatric patients in

the La Paz Department with functional and vascular disorders who do not have private insurance.

## Limitations

There are limitations to our study. First, this was a retrospective review of data and is vulnerable to reporting and selection biases. The EMR system is not specific to neurosurgery or neurosurgical outcomes, and as such, outcomes could not be more specific at this time. Second, the imaging system at the Hospital del Niño remains film-based, not digital. Therefore, it was not possible to review all images of all indications given that film images are not always stored after surgery (often patients return home with the films in hand). Lastly, the generalizability of these findings to all pediatric institutions in Bolivia is not advised. Currently, the exact landscape at a national level is unclear; however, we are aware of other pediatric neurosurgery units in the country with access to an operative microscope and more functioning in-house imaging systems, which would likely lend themselves to broader neurosurgical indications and more robust operations. Greater collation of national-level data is required to ascertain how exactly these outcomes in La Paz compare in the context of Bolivian pediatric neurosurgery.

## Moving Forward

There are many resource deficiencies at the Hospital del Niño that need to be addressed to stabilize the current capacity of the neurosurgery department. Immediate priorities include reliable imaging modalities, such as a CT scanner, followed by an operative microscope and endoscope. These aspects alone will ensure that the current performable surgeries are optimized. Then, patient and family education will remain critical to improve follow-up rates. The challenge that La Paz faces is that it is in an extensive geographical province that requires transport and may not be affordable or available to all. Hospital-based transport services may in the future address in part this concern. It remains most practical that these considerations should precede any discussion as to the expansion of the subspecialties (i.e., functional and vascular) by the neurosurgery department. As is the general trend among many upper-middle-income countries and LMICs of South America, strategic centralization of different resource-heavy subspecialties in pediatric neurosurgery is likely the optimal solution to expansion of capacity in the future of both La Paz and Bolivia long-term.<sup>32</sup>

## Conclusions

The Hospital del Niño “Dr. Ovidio Aliaga Uria,” La Paz, Bolivia, is the only publicly funded pediatric hospital that offers neurosurgery to a geographic region of 2.9 million inhabitants. We conducted the first audit of the inpatient outcomes within the neurosurgery department and report a limited capacity for a growing population with increasing reliance on universal healthcare. Inpatient outcomes for this illustrative institution of a South American LMIC remain very different from those of HICs, and as such, to improve capacity and outcomes, institution- and country-specific factors need to be considered.



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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

Conception and design: Lu, Niazi, Jorge David Brun. Acquisition of data: Lu, Jorge David Brun. Analysis and interpretation of data: Lu. Drafting the article: Lu, Niazi. Critically revising the article: Lu, Niazi, Jorge David Brun. Reviewed submitted version of manuscript: Lu, Niazi, Jorge David Brun. Approved the final version of the manuscript on behalf of all authors: Lu. Study supervision: Jorge Daniel Brun, Jorge David Brun.

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# Pediatric neurosurgical medulloblastoma outcomes in La Paz, Bolivia: How a Lower Middle-Income Country (LMIC) institution in South America compares to the United States

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## Abstract

**Background** How pediatric medulloblastoma patients fare in Lower Middle-Income Country (LMICs) in South America is not well understood. Correspondingly, the aim of this study was to summarize the pediatric neurosurgical experience of an institution in La Paz, and compare outcomes to that of a generalized High Income Country (HIC) United States (US) experience.

**Methods** A retrospective review of all pediatric neurosurgical medulloblastoma patients at the Children's Hospital of La Paz, Bolivia (Hospital del Niño "Dr. Ovidio Aliaga Uria") between 2014 and 2023 was conducted and compared to a generalized US experience abstracted from the US National Cancer Database (NCDB) and National Inpatient Sample (NIS) databases. Categorical, continuous and survival data were statistically summarized and compared.

**Results** A total of 24 pediatric medulloblastoma patients underwent neurosurgical treatment at the Hospital del Niño. In this La Paz cohort, there were 15 (63%) males and 9 (38%) females, with a mean age of 5.6 years old at diagnosis. The majority of patients underwent subtotal resection (STR, 79%), while the remaining patients underwent biopsy only. Ten (42%) patients expired during their hospitalization, and mean length of stay overall was 39 days. Only 8 (33%) patients received adjuvant treatment after surgery. Median overall survival from diagnosis in the La Paz cohort was 1.9 months. Compared to the US databases, the La Paz cohort experienced significantly more emergency room admissions for surgery, less gross total resection, more STR, more return to operating room for ventriculoperitoneal shunting, more bacteremia, more tracheostomy procedures, more percutaneous gastrostomy placements, longer lengths of stay, less adjuvant chemotherapy, less radiation therapy, shorter follow-up, and ultimately, significantly shorter overall survival (all  $P < 0.050$ ).

**Conclusions** Pediatric neurosurgical medulloblastoma outcomes at the Children's Hospital of La Paz, Bolivia are significantly inferior to that of a generalized US experience. Future research is required to identify institution- and country-specific initiatives to improve discrepancies between institutions in LMICs in South America compared to HICs.

**Keywords** Medulloblastoma · Pediatric · Neurosurgery · La Paz · Bolivia · South America · LMIC

## Introduction

Medulloblastoma is a malignant brain tumor that is the most common pediatric central nervous system (CNS) tumor, constituting up to 20% of them [1]. Although prognosis is variable depending on molecular subtype, surgery and adjuvant therapy remain the key components for treatment [2, 3]. Epidemiologic data reveals that the 5-year survival in a contemporary High Income Country (HIC) United States cohort is 72% with surgical treatment alone compared to 35% without surgery [4]. Given how resource dependent surgical intervention is, our understanding of how pediatric

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medulloblastoma patients fare in more resource-limited setting in Low and Lower-Middle Income Countries (LMICs) in South America is not well understood.

Bolivia is one such example, a landlocked country in the South America with a population of approximately 12 million inhabitants. Per the World Bank, it is LMIC whose gross domestic product (GDP) per capita ranks 96th in the world, and Human Development Index (HDI) ranks 114th in the world [5–7]. In the La Paz Department, the Hospital del Niño “Dr. Ovidio Aliaga Uriá” serves as the sole publicly funded pediatric hospital to treat all neurosurgical patients in the region with approximately 2.9 million inhabitants [8]. This hospital remains severely resource limited in terms of neurosurgery and medulloblastoma care. There is no magnetic resonance imaging (MRI), operative microscope, neuronavigation, or intraoperative imaging available, and while there are limited chemotherapy services, there is no radiation therapy available on site.

The objective of this study was to summarize the clinical outcomes of pediatric medulloblastoma patients treated at the Hospital del Niño, La Paz and compare them to a generalized HIC US experience to understand if and what anecdotal disparities exist in La Paz, Bolivia.

## Methods

### Design

This study sought to compare the Hospital del Niño, La Paz experience of pediatric medulloblastomas to that of an anecdotal experience within the US using US national databases

- the National Cancer Database (NCDB) was used to define treatment outcomes and the National Inpatient Sample (NIS) was used to define inpatient outcomes.

### Hospital Del Niño, La Paz

The primary La Paz data was retrospectively obtained from the national electronic medical record system ‘Sistema de Información Clínico Estadístico’ (SICE) at the Hospital del Niño “Dr. Ovidio Aliaga Uriá”, La Paz, Bolivia between 2014 and 2023 after institutional approval. This hospital is located in the Murillo Province within the La Paz Department. The Department of Neurosurgery is currently staffed by two pediatric neurosurgeon attendings accredited by the national Bolivian Society of Neurosurgery, and perform an average of 95 neurosurgical cases a year between them. Inclusion criteria for the La Paz cohort were (1) patients aged 18 years and younger, (2) who underwent at least one neurosurgical procedure during admission for tumor resection, (3) with histological tissue diagnosis of

medulloblastoma, and (4) a documented operative report. Exclusion criteria were (1) patients with scheduled neurosurgery which was ultimately not performed, and (2) admissions with incomplete documentation.

### NCDB data

Data representative of the medulloblastoma treatment outcomes in the US were extracted from the 2016 iteration of the National Cancer Database (NCDB), a database maintained by the Commission on Cancer (CoC) and the American Cancer Society since 2004 which describes over 70% of new cancer diagnoses from 1,500 hospitals in the United States [9]. The database was queried for all patients that satisfied the following; inclusion criteria of (1) pediatric patients aged  $\leq 18$  years, with (2) surgically proven (biopsy or resection) primary medulloblastoma diagnosis (ICD-0-3 947X/3), (3) with known treatment (surgical resection, chemotherapy and radiation therapy) statuses and overall survival outcome. Exclusion criteria were (1) recurrent pathology, (2) diagnosis by autopsy after death, (3) adult patients, and (4) incomplete clinical data.

### NIS data

Data representative of the medulloblastoma inpatient outcomes in the US were extracted from the 2020 iteration of the National Inpatient Sample (NIS) database, a publicly available database developed by the Agency of Healthcare Research and Quality for the Healthcare Cost and Utilization Project (HCUP). The NIS is the largest all-payer healthcare database in the United States and is derived from 4,550 hospitals in 48 states, representing 20% of all inpatient admissions in the country. Inclusion criteria used were (1) pediatric patients aged  $\leq 18$  years, (2) with medulloblastoma by International Classification of Diseases (ICD) 10 diagnosis (C71.6), and (3) were treated with suboccipital decompression and tumor resection during admission (ICD-10 procedure codes 00BC0ZX/Z). Exclusion criteria were (1) incomplete outcome data and (2) unspecified surgical procedures.

### Outcomes

The primary outcome of this study was the overall survival from index surgery. In addition to sociodemographic parameters of all patients, other outcomes of interest were nature of hospital admission, insurance status, neurosurgical procedures performed during admission, extent of resection (EOR), intraoperative complications, duration of surgery, postoperative length of stay (LOS), complications during hospitalization, status at time of discharge, follow-up,

and the use of adjuvant chemotherapy and radiation therapy. The EOR in the La Paz cohort was based on the operating neurosurgeons judgement at the end of the surgery given the lack of postoperative imaging available to all patients.

## Statistics

Outcome comparisons between groups were conducted using chi-square exact test and Wilcoxon rank-sum test for categorical and continuous data, respectively. Kaplan-Meier estimations using log rank testing were used to compare overall survival (OS). A comparative NCDB cohort was created matching for age at surgery and gender to the La Paz cohort. All analyses were conducted using STATA 14.1 (StataCorp, College Station, Texas); tests were two-sided, and statistical significance was defined using the alpha threshold of 0.05.

**Table 1** Presentation characteristics of the La Paz, Bolivia and the NCDB, US medulloblastoma patients with comparative *P*-values when applicable. Categorical data reported as count (% total) and continuous data reported as mean  $\pm$  SD, unless otherwise stated

Characteristic	La Paz, Bolivia ( <i>n</i> = 24)	NCDB, US ( <i>n</i> = 163)	<i>P</i> -value	NIS, US ( <i>n</i> = 80)	<i>P</i> -value
Age, yr	5.6 $\pm$ 3.8	6.4 $\pm$ 5.4	0.947	7.1 $\pm$ 4.4	0.098
Gender			0.614		0.587
Female	9 (38%)	70 (43%)		35 (44%)	
Male	15 (63%)	93 (57%)		45 (56%)	
Residence*					
La Paz Department	19 (74%)	.	.	.	.
Murillo Province	12/19 (63%)	.	.	.	.
Admission route					<b>0.035</b>
Emergency room (ER)	21 (88%)	.	.	52 (65%)	
Primary	15/21 (71%)	.	.	.	
Transfer	6/21 (29%)	.	.	.	
Elective	3 (12%)	.	.	28 (35%)	
Insurance			0.338		
Private	10 (42%)	85 (52%)		.	.
Other**	14 (58%)	78 (48%)		.	.

\*The Hospital del Niño is located in the Murillo Province, one of 20 provinces in the La Paz Department, which is one of the 9 departments of Bolivia

\*\*Other includes universal healthcare insurance and no insurance statuses

## Results

### Demographics

A total of 24 patients from the Hospital del Niño, La Paz, satisfied all criteria for selection into the study (Table 1). There were 15 (63%) males and 9 (38%) females, with a mean age of 5.6  $\pm$  3.8 years old (range 0.8–14.7 years old). The majority of patients were from within the La Paz Department (*n* = 19/24, 79%) and within the Murillo Province (*n* = 12/19, 63%). With respect to route of admission, the majority of patients presented via the emergency room (ER, *n* = 21/24, 88%) versus electively (*n* = 3/24, 12%). Of those who presented through the ER, more patients presented as a primary patient (*n* = 15/21, 71%) than as a transfer from an outside hospital (*n* = 6/21, 29%). Finally, more patients were reliant on public universal healthcare insurance (*n* = 14/24, 58%) than private insurance (*n* = 10/24, 42%) during their admission.

### Surgical outcomes

All 24 patients underwent suboccipital craniectomy for attempted tumor resection, with three (*n* = 3/24, 13%) patients requiring ventriculoperitoneal shunting (VPS) prior to tumor resection surgery due to obstructive hydrocephalus (Table 2). There were no instances of patients presenting with posterior fossa mass that did not proceed to surgery. Mean operating time was 2.8  $\pm$  0.9 h, and there were no significant intraoperative complications reported. No case was deemed to have achieved macroscopic gross total resection (GTR). The majority of patients underwent subtotal resection (STR, *n* = 19/24, 79%) with a mean estimated EOR of 69  $\pm$  18%. The remaining patients underwent biopsy only (*n* = 5/24, 21%).

### Hospital course

After index surgery, most patients (*n* = 19/24, 79%) were able to obtain postoperative imaging by computed tomography (CT) (Table 2). Nearly one half of patients (*n* = 11/24, 46%) required a return to the operating room (OR) during their admission, with all returns ultimately involving the placement of VPS for postoperative hydrocephalus. There were 6 (*n* = 6/11, 55%) patients who returned to the OR multiple times during admission due to VPS failure, repeat resection attempt and cerebrospinal fluid (CSF) leak (each *n* = 2). Postoperatively, the most common complications seen in hospital were septicemia following bacteremia (*n* = 4/24, 17%), tracheotomy (*n* = 3/24, 12%) and percutaneous feeding tube placement (*n* = 3/24, 12%). Meningitis preceded septicemia in two cases. Ultimately of the 24 patients mean

**Table 2** Treatment characteristics of the La Paz, Bolivia and the NCDB, US medulloblastoma patients with comparative *P*-values when applicable. Categorical data reported as count (% total) and continuous data reported as mean  $\pm$  SD, unless otherwise stated. GTR, gross total resection; STR, subtotal resection; OR, operating room; LOS, length of stay

Characteristic	La Paz, Bolivia (n=24)	NCDB, US (n=163)	<i>P</i> -value	NIS, US (n=80)	<i>P</i> -value
Operating time, hr	2.8 $\pm$ 0.9				
Extent of resection			< 0.001		
GTR	0	73 (45%)			
STR	19 (79%)	46 (28%)			
Biopsy only	5 (21%)	44 (27%)			
Postoperative imaging	19 (79%)				
Return to OR					
Repeat surgery	5 (21%)			8 (10%)	0.159
VP shunt	11 (46%)			7 (9%)	< 0.001
Complications					
Bacteremia	5 (21%)			0	< 0.001
Tracheotomy	5 (21%)			0	< 0.001
PEG	4 (17%)			2 (3%)	0.009
LOS, d	39 $\pm$ 51			10 $\pm$ 9	< 0.001
Status at discharge					
Alive	14 (58%)			80 (100%)	< 0.001
Dead	10 (42%)			0	
Adjuvant treatment					
Chemotherapy	8 (33%)	127 (78%)	< 0.001		
Radiation therapy	5 (21%)	95 (58%)	0.001		

LOS was 39  $\pm$  51 days, although only 14 (58%) patients were discharged from hospital alive, with 10 (42%) patients expiring during their hospitalization. No cases of cerebellar mutism were noted in the survivors. Causes of death for the 10 patients that expired included septicemia ( $n = 4/10$ , 40%), respiratory failure ( $n = 3/10$ , 30%), shunt failure ( $n = 3/10$ , 30%), and one case was not reported.

## Treatment and follow-up

After surgery, 8 (33%) patients were able to receive chemotherapy and of those, 5 (21%) were also able to receive radiation therapy (Table 2). Mean time to therapy initiation for chemotherapy was 14.5  $\pm$  5 days and 2.1  $\pm$  1 months

**Table 3** Follow-up outcomes of the La Paz, Bolivia and the NCDB, US medulloblastoma patients with comparative *P*-values when applicable. Categorical data reported as count (% total) and continuous data reported as mean  $\pm$  SD, unless otherwise stated. CI, confidence interval

Characteristic	La Paz, Bolivia (n=24)	NCDB, US (n=163)	<i>P</i> -value	NIS, US (n=80)	<i>P</i> -value
At least one follow-up	11/14 (79%)				
Follow-up time, mo	20 $\pm$ 20	30 $\pm$ 25	< 0.001		
Final status			0.220		
Alive	9 (38%)	83 (51%)			
Dead	15 (63%)	80 (49%)			
Median survival, mo (95% CI)	1.9 (0.7–36)	32.9 (12–51)	< 0.001		

respectively. For chemotherapy, the most common agents used were vincristine and vinblastine over 4–12 cycles. For radiation therapy, all patients were aged above 3 years and received a fractionated 30 Gy dose to the craniospinal axis, followed by a 25 Gy boost dose to the posterior fossa over 6 weeks. There were no associated complications observed with any of these adjuvant therapies. Of the patients discharged to outpatient, most of them ( $n = 11/14$ , 79%) completed at least one follow-up visit (Table 3). Mean follow-up time for these patients was 20  $\pm$  20 months.

## Survival in La Paz cohort

At last contact, only 9 of the initial 24 (38%) of medulloblastoma patients were still alive with a median follow-up time of 7.3 months (range 1–64) (Table 3). In sum, of the 15 patients that had expired, the majority of these events occurred during the postoperative inpatient stay period after index surgery ( $n = 10/15$ , 67%), with the remaining cases occurring after discharge following index surgery hospitalization. Median overall survival from diagnosis of this cohort was 1.9 months (95% CI 0.7–36).

## Comparisons with NCDB and NIS

Outcomes of the La Paz cohort ( $n = 24$ ) in Bolivia were then compared to NCDB ( $n = 163$ ) and NIS ( $n = 80$ ) cohorts from the United States (Tables 1, 2 and 3). All cohorts were statistically comparable in terms of mean age at diagnosis and gender.

Comparison with the NCDB cohort revealed that the La Paz cohort underwent significantly less GTR (0% vs. 45%), more STR (79% vs. 28%), and less biopsy (21% vs. 27%) ( $P < 0.001$ ), and received significantly less chemotherapy (33% vs. 78%,  $P < 0.001$ ) and significantly less radiation therapy (21% vs. 58%,  $P = 0.001$ ) compared to the NCDB cohort (Table 1). In terms of

survival, the overall proportion of patients alive at last follow-up did not differ between the La Paz cohort and the NCDB cohort. However, follow-up time after diagnosis was significantly shorter in the La Paz cohort (20 vs. 30 months,  $P < 0.001$ ), and median overall survival from diagnosis was also significantly shorter (1.9 vs. 32.9 months,  $P < 0.001$ ) (Fig. 1).

Comparison with the NIS cohort revealed that the La Paz cohort presented significantly more via the ER (88% vs. 65%,  $P = 0.035$ ) than electively, and that they experienced significantly more return to the OR for VPS (46% vs. 9%,  $P < 0.001$ ), and more postoperative complications of bacteremia (17% vs. 0%,  $P < 0.001$ ), requiring tracheostomy (21% vs. 0%,  $P < 0.001$ ) and PEG placement (17% vs. 3%,  $P < 0.001$ ) (Table 1). The La Paz cohort experienced significantly longer mean lengths of stay after surgery (39 vs. 10 days,  $P < 0.001$ ) and ultimately, significantly higher proportion of in-hospital death compared to the NIS cohort (42% vs. 0%,  $P < 0.001$ ).

## Discussion

How pediatric medulloblastoma patients perform in LMIC Bolivia in South America has not been reported before. We report for the first time the outcomes of the sole publicly funded pediatric hospital in one of the largest geographical departments of Bolivia. We demonstrate that surgical outcomes, postoperative outcomes, and treatment outcomes at the Hospital del Niño, La Paz are largely inferior to that of a HIC experience from the US.

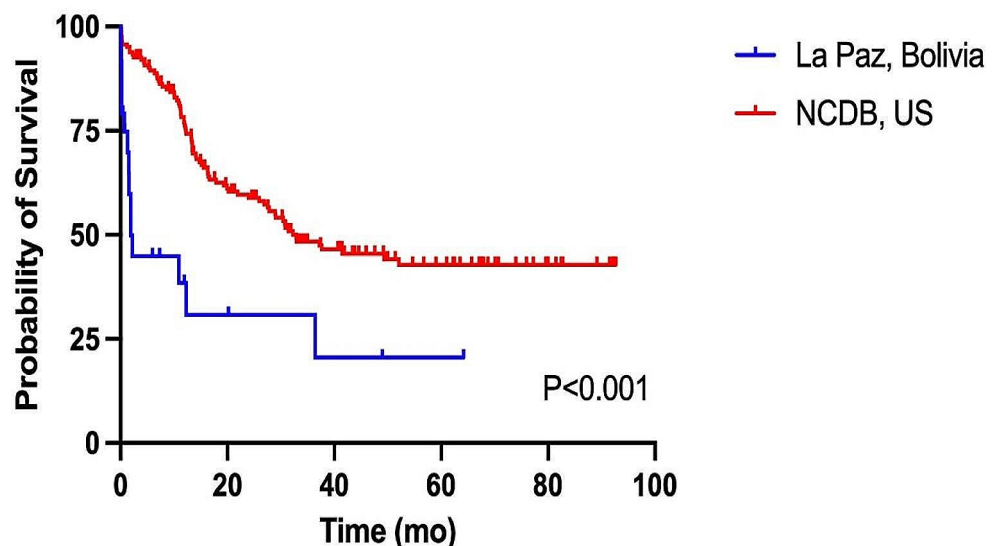
At time of surgery, the patients that presented in La Paz were comparable in both age and gender to that of a US experience. Specific to the hospital, although the majority of patients were from the local geographic

province and department, a notable number of patients were from areas outside the local catchment area. This broad catchment area may contribute to the difficulty many patients face in accessing adjuvant therapy and maintaining effective post-operative follow-up [10]. This would align with the trend that geographical distance has been reported elsewhere to contribute to inferior neuro-oncological care in other LMIC in other regions of the world [11, 12].

Most of the patients seen in La Paz were admitted via the ER for eventual surgical treatment. This rate was significantly higher than in the US, where more elective admissions were seen. Reasons as to this trend can be reflected in the barriers to timely diagnosis and workup. Access to primary care, access to imaging modalities, and the appropriate insurance for elective workup are just some of the barriers that have been reported in the broader literature as socio-economic predictors of delayed care in LMICs [13]. This is important to recognize as a barrier to care specifically for medulloblastoma because in HICs, workup for posterior fossa masses include full spine magnetic resonance imaging (MRI) to investigate drop metastases as well as CSF cytology from lumbar puncture sampling to assist in staging of disease. The reality in La Paz is that these adjuncts to staging are not available due to cost and severity of presentation. As a result, patients are not as effectively staged prior to surgery, and it is likely this contributes to the inferior outcomes we observe in La Paz, Bolivia when compared to the US experience.

More delayed care and treatment can result in more emergent presentations to the ER rather than elective concerns seen in clinic [14]. These emergent presentations can be related to obstructive hydrocephalus that medulloblastoma can cause within the posterior fossa if given enough time to develop, and include signs such as vomiting, gait

## Medulloblastoma survival



**Fig. 1** Kaplan-Meier survival curve for medulloblastoma patients. Overall survival from diagnosis was significantly shorter in the La Paz cohort compared to the NCDB cohort (median survival 1.9 vs. 32.9 months,  $P < 0.001$ )

disturbances and altered mental status. What is concerning and specific to this setting is the unavailability of external ventricular drains (EVDs) and other emergent materials for CSF diversion to treat such presentations. This is because these materials need to be purchased emergently by the patient and family, which is typically not possible in this hospital. As a result, there were no EVDs seen in the La Paz cohort.

The Bolivian government formally introduced the 'Sistema Unico de Salud' (SUS) in 2018 [15], a public universal healthcare program similar to that of the 'Medicaid' system in the US. Being a publicly funded hospital, the Hospital del Niño was an early utilizer of this system to enable care for patients. However, this also has placed financial constraints on the care available to medulloblastoma patients. This is because although the plan will cover the basic hospital care, it does not readily cover indirect costs and disposable materials such as transportation to other facilities to obtain further imaging and surgery materials such as shunt valves, dural grafts. Adjuvant treatment is typically coverable however time to approval can take a matter of weeks. As such, the cost of care is a significant barrier to achieving comparable care for these patients out-side of surgery.

Operatively, no patient in the La Paz cohort was deemed to have undergone a GTR compared to 45% in the US experience. It is suspected that the absence of an operating microscope, preoperative MRI for all patients, and intraoperative neuronavigation and ultrasound in the Hospital del Niño all contribute to the difficulty in achieving GTR. This trend in less GTR for pediatric brain tumors due to limited resources is not uncommon in LMICs [16]. There were a number of postoperative infections (meningitis and bacteremia) that occurred in this cohort which is difficult to attribute to a single source. Nonetheless, their contribution to burden of disease in LMICs has been noted previously [17]. Specific to surgery, meticulous closure and appropriate concurrent hydrocephalus management will ideally assist in minimizing these complications in the future which are already an active part of the surgery in La Paz [18].

Pathologically all tumors in this cohort were diagnosed as medulloblastoma, however currently there is no molecular service available at the hospital to further provide information on molecular subtype. As such, it is difficult to determine how detrimental the absences of these surgical adjuncts are as extent of resection has proven significantly prognostic in the progression free survival of particular medulloblastoma molecular subtypes [19].

However, it is known that postoperative adjuvant therapy for pediatric medulloblastoma is a significant predictor of overall survival [2, 3, 20]. As such, our results

indicate there remains a great disparity in the access to adjuvant care in the La Paz cohort compared to the US experience, with only one third of patients in La Paz able to access chemotherapy alone. It is likely this disparity itself contributes to the significantly shorter overall survival of pediatric medulloblastoma patients in La Paz compared to the US experience. Reasons for those patients to not undergo adjuvant therapy are difficult to understand in retrospect, although when documented decisions to proceed with palliative care and the cost for further care were listed in two separate cases. Specific to radiation therapy, as this service is not available at the Hospital del Niño, patients must search for these services at outside facilities and be able to meet the financial costs. The authors speculate that patient and family education can play a significant role in improving the decision and ability to proceed with adjuvant therapy. There are local charitable services that can provide information to families, as well as host families during cancer care should accommodation be an issue. Improved understanding of these services will greatly enhance the quality of future care for these patients after surgery.

Additionally, there are likely local- and hospital-based challenges specific to La Paz and Bolivia as well contributing to these outcomes, for the survival in neighboring countries in South America such as Peru [21] and Brazil [22] are longer (> 10 months) for the same pediatric diagnosis than what we report here. One example specific to Hospital del Niño is the availability of postoperative intensive care to these complex neurosurgical patients. There are currently only 8 functional intensive care unit (ICU) beds available for all services in the hospital, which very much limits the capacity in which patients can be managed after surgery given the high risk of respiratory issues, swallowing issues and hydrocephalus issues in the immediate postoperative period. Improved ICU infrastructure in the future will likely correlate with improved postoperative care and inpatient outcomes for these high-risk pediatric medulloblastoma patients.

There are limitations to this study. Firstly, the retrospective nature of the La Paz cohort limits our ability to expand the data reported, and further, limits our ability to generalize trends to the whole country. There are currently an unknown number of private clinics and adult neurosurgeons in La Paz that can also operate on pediatric brain tumors. Understanding how those patients fare, as well as patients in other pediatric hospitals in other major Bolivian cities will greatly enhance our understanding as to how typical the La Paz cohort and its outcomes are to the country. This can be overcome in the future with a more prospective national database collection approach. Secondly, the absence of molecular technologies in the hospital prevents us from making any further

understanding as to the natural history of pediatric medulloblastoma in the La Paz Department. Currently the costs of doing this is not something the majority of patients and their families can meet. Thirdly, the limited resource setting of the La Paz hospital can be argued to limit the comparability between patients treated there versus patients treated in other countries with more resources. Statistically it can confound outcomes and any regression analyses should take this into account if trying to create a predictive model in the future. Finally, the US experience is based on two national databases which themselves are limited. Although they are sampled across many hospitals in the US, how exactly one singular institution may fare in the US compared to the Hospital del Niño in La Paz, Bolivia may vary. As such the differences reported in the study remain anecdotal and not necessarily generalizable to every US or high-income country setting.

## Improving care in the future

A future challenge for quality improvement in the Hospital del Niño is the inpatient survival outcome. Two thirds of the overall deaths in the La Paz cohort occurred during index admission for surgery, an occurrence that was not singularly seen in the US experience. In part these poorer outcomes mirror outcomes of other LMICs limited by late diagnosis, delayed treatment, limited medical surveillance protocols, and heterogeneous socioeconomic conditions that impact postoperative care [23]. Resources that are low-cost need to be considered when initially attempting to improve timeliness of diagnosis and staging, including low-field, portable MRI machines and limited-stain cytology of CSF [24].

In terms of surgery outside the obvious need for an operating microscope and MRI services, we propose multiple lower-cost considerations for the future. The first is to continue to enhance the surgical training of attendings in the hospital by having them participate in surgeries with formal feedback [25]—this can include via video link, via mission trip visits, and even via visiting the operating room of neurosurgeons in HICs. Further to that, ‘twinning’ the Hospital del Niño with a pediatric hospital in the US could also prove effective in improving surgical decision-making. This concept of twinning involves continued communication (by email, by video link) to regularly discuss cases which has been shown to be effective in other domains of surgery in LMICs [26].

From a neuro-oncology perspective, one noteworthy endeavor will be the establishment of a formal multi-disciplinary team (MDT) involving neurosurgeons, oncologists

and radiologists. Currently this does not exist at the Hospital del Niño in the same form as it does in HICs. It has been shown that MDTs improve the decision-making in the care of brain tumors [27]. The challenges faced at the Hospital del Niño is the limited infrastructure to formalize this team, but efforts are in place to continue to build a more collaborative approach in parallel the continued improvements in communicating technology available at the hospital in the future.

## Conclusions

The current prognosis for pediatric medulloblastoma in the Hospital del Niño, La Paz, Bolivia remains inferior to that of a generalized United States experience. Poorer outcomes may be attributed to, in part at the very least, to the limited resources available at the hospital, and prospective efforts are required to further identify actionable items for translational improvement.

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**Data availability** No datasets were generated or analysed during the current study.

## Declarations

**Competing interests** The authors declare no competing interests.

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**Surgical Management Of Patients with Giant Vestibular Schwannomas (T4 A And B Hannover Classification): Analysis Of 36 Cases**

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**Keywords:** neuro oncology, vestibular schwannomas

## Abstract

**Background:** Vestibular Schwannoma (VS) is a tumor with benign histopathological characteristics that may present aggressive behavior. Literature on giant VS is limited, surgical techniques and manners to minimize complications and improve postoperative outcome are not clearly defined. **Objective:** Analyze the data of patients with giant VS, aiming to statistically evaluate the pre- and post-surgical parameters, techniques used for tumor resection and the results of facial nerve monitoring.

**Methods:** This is a cross-sectional retrospective study in which the data from 36 patients with giant VS (T4 Hannover classification) who underwent surgery between 1999 and 2021 were analyzed. These patients were selected from a major group of 173 patients with vestibular schwannomas. Statistical analysis was performed adopting significance less than 5% ( $p \leq 0.05$ ).

**Results:** A significant correlation was found between facial nerve function in the immediate postoperative period and the intraoperative monitoring of this nerve with evoked potentials ( $p=0.024$ ). Patients which were not submitted to facial nerve monitoring showed worse facial nerve function rates (IV/V House-Brackmann classification) in the immediate postoperative period when compared to those who were monitored. We obtained facial nerve preservation in 66,7 % of patients after 6 months.

**Conclusion:** Preserving the facial nerve and avoiding post-surgical complications are factors that should always be aimed. Our study proved that intraoperative monitoring of the facial nerve with evoked potentials improved facial nerve functionality. Unfortunately, the use of evoked potentials is still not a reality for many centers, especially the public ones.

**Keywords:** facial nerve, functional outcome, giant vestibular schwannoma, Hannover classification, vestibular schwannoma.

## Introduction

Vestibular Schwannoma (VS) is a benign tumor located in the cerebellopontine angle, which originates from the sheath of the VIII cranial pair, the vestibulocochlear, from an exacerbated growth of Schwann cells. It is commonly located on the upper and/or lower vestibular nerve, being more common in the upper one, but can also occur in the cochlear, a fact that explains the lower use of the term acoustic neurinoma. [3, 6] Among brain tumors, it represents 8% of primary intracranial neoplasms, with an incidence of 1.09:100,000 in the United States.[3, 4]

It is noteworthy that the VS has a very close relationship with important cranial nerves, such as the oculomotor and trigeminal nerves at the upper limit, facial immediately adjacent to the superior and inferior vestibular nerves, and glossopharyngeal and vagus nerves at the lower limit.[15] Symptoms in patients with VS usually start with those related to the VIII pair, and hearing loss, imbalance and the presence of tinnitus are frequent. Nausea and short-term dizziness may also be reported.[19] After these, changes in motricity and sensitivity of the face and ocular motricity demonstrate the progression of the disease. At advanced levels there may be compression of the brainstem, making the tumor possibly fatal.[4]

The classification of VS is usually done by its extension. The most used classification in the literature is the Hannover classification, differentiating them from T1 to T4b, being T1 purely intracanalicular, T2 intrameatal or extrameatal, T3a fills the cerebellopontine cistern, T3b reaches the brainstem, T4a makes compression of the brainstem and T4b makes brainstem compression and fourth ventricle deviation. [7, 19] Consistency concerns whether the tumor is solid or cystic.[5]

Comprehension of the eloquent areas around the VS is vital. Cranial nerves, when altered, interfere in daily human practices and, in the most serious cases, directly in the lives of individuals. Correctly employed and performed neurosurgical techniques and treatments, knowledge of the anatomy surrounding this tumor, method of approach and resection (total, subtotal or partial) are decisive issues in the patient's prognosis.[16] A major concern during resection surgery for these giant tumors is the preservation of facial nerve function. Some factors can influence this result, such as size and adhesion of the tumor, whether it is cystic or solid, the position of the nerve in relation to the tumor and the extension of the resection.[8] The House-Brackmann classification system assesses the function of the VII nerve by moving the face and can be used in the pre- and post-surgical evaluation of patients with VS, being divided into grades from I (normal function) to VI (total paralysis).[10]

Surgical access and patient positioning are still much debated, and access through the middle fossa, translabyrinthine and retrosigmoid, and different positions for each one of them, can be used. Each combination of access and position has advantages and disadvantages. The semi-sitting position presents a risk of air embolism, but it allows the use of the third-hand technique for irrigation and cleaning of the surgical field, enabling continuous bimanual resection. The dorsal decubitus position (DDP), on the other hand, facilitates the accumulation of fluid and blood, difficulting the visualization of the cranial nerves and increasing the risk of injury to them.[3]

Consequently, considering the few cases of giant VS in the literature, the current study aims to assess the pre- and post-surgical facial nerve functionality, comparing it with the techniques used during giant VS surgery (patient positioning, use of evoked potential, surgical access, extension of the resection, if radiosurgery was used) and with the complications, and with it, to verify the post-surgical prognosis of 36 patients.

### **Materials and methods**

This is a cross-sectional retrospective study, in which pre- and post-surgical data from 36 patients with giant vestibular schwannomas (Hanover T4 classification) who underwent surgery between 1999 to 2021 by the senior surgeon were analyzed. Those patients were selected from a major group of 173 patients with Hannover T1, T2, T3 and T4 classification (20, 42, 75 and 36, respectively). These data were obtained by consulting his personal file and the following information were considered for analysis: gender, age, size, extension, laterality, consistency, concomitant fibromatosis, preoperative hearing, facial nerve functionality (preoperative, post-immediate and after 6 months), facial nerve anastomosis after tumor resection, previous surgical procedure, preoperative radiosurgery, shunt placement in the postoperative period, surgical positioning of the patient, craniotomy technique, intraoperative monitoring of the facial nerve with evoked potentials and if there was a drop in this potential during surgery (50% decay alarm criterion), size of tumor resection, fistula or other immediate postoperative complications and tumor progression within 1 year.

Facial nerve functionality was assessed according to the House-Brackmann (HB) classification.[14] It is important to mention that the analyzes performed on facial nerve functionality 6 months after surgery and tumor progression within 1 year had a smaller patient sample, as 2 patients died during the period (glottis edema and pneumonia) and 4 patients had no clinical follow-up. Thus, this specific parameter after 6 months and 1 year had 30 patients and the others had 36 patients.

The size of the bone opening was not considered. Tumor resections were defined as Gross Total Resection (GTR) when more than 95% of the tumor was removed and Subtotal Resection (STR) or Debulking when those values were between 90 and 95% and 40-90%, respectively. This was verified by the surgeon during surgery. The parameter complications considered fistula, other complications and/or death after the tumor removal surgery.

Tumor progression in 1 year considered whether there was tumor regrowth or not during this period.

Statistical analysis was performed in the Laboratory of Research Design and Scientific Writing of [BLINDED FOR REVIEW]. Initially, a descriptive analysis was performed in which qualitative variables were expressed as absolute and relative frequencies and quantitative variables as means, standard deviations, minimum and maximum values. The analysis of the obtained results was performed adopting a significance lower than 5% ( $p \leq 0.05$ ). The variable age was presented as mean and 95% confidence interval, using the Shapiro-Wilk's test for data normality. To compare qualitative variables, Fisher's exact test was used. Data tabulation was performed using Microsoft Excel and statistical analysis using Stata version 14.0.

The study was approved by the [BLINDED FOR REVIEW] Hospital Clinical Research Ethics Committee with the registration number 4.987.301 and CAAE 50580521.7.0000.5670. Patient consent was given for figure 1 publication.

## **Results**

The 36 patients included in this study were operated between 1999 and 2021 in a general neurosurgery center. We had an incidence of 1.6 case per year of T4 A and B VS. Figure 1 exemplifies a case among our patients. The data obtained can be seen in table 1.

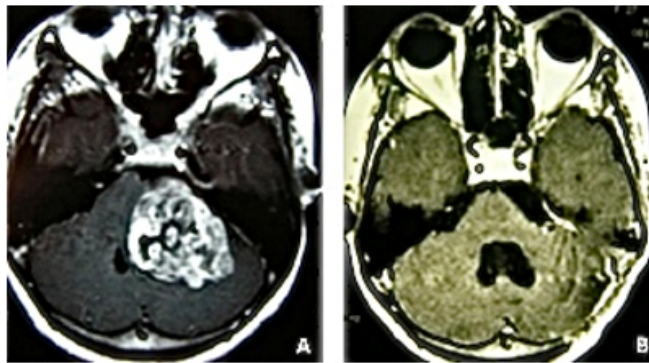


Figure 1: 22-year-old female patient, with a T4B (Hannover Classification) Vestibular Schwannoma. A: Axial MRI T1 with gadolinium showing the tumor at the left cerebellopontine angle. B: Axial MRI T1 with gadolinium showing total resection of giant vestibular schwannoma.

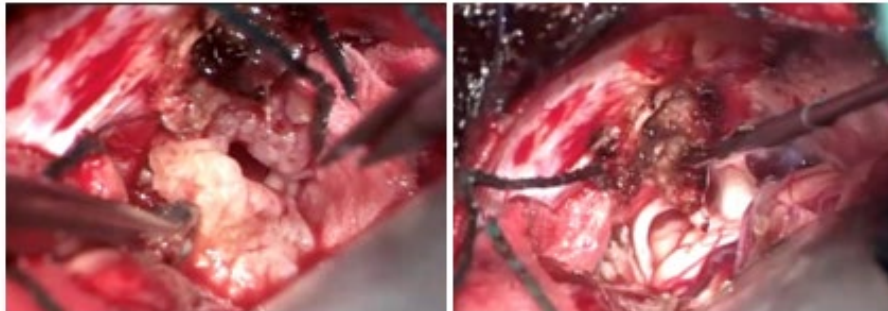


Figure 2: Debulking of the giant vestibular schwannoma using the two hands technique to dissect its pseudocapsula.

Table 1A. Data obtained from medical records of the 36 patients included in the study

N°	Sex	Age	Tumor classification before surgery (Hannover)	Tumor extension	Laterality	Consistency	Etiology	Pre-operative hearing	HB before surgery	HB immediately after surgery	HB 6 months after surgery	Facial nerve procedure	Previous treatment
1	M	45	T4a	Lateral	Left	Solid	Sporadic	Absent	I	V	-	-	-
2	M	40	T4a	Tentorial	Left	Solid	Sporadic	Absent	I	IV	II	Hypoglossal facial anastomosis	-
3	F	24	T4a	Laterotentorial	Left	Solid	Sporadic	Absent	III	III	III	-	Regrowth but was operated before in another place
4	F	20	T4b	Lateral	Right	Solid	Sporadic	Absent	I	I	Death due to glottic oedema	-	-
5	M	36	T4a	Tentorial	Left	Cystic	Sporadic	Absent	I	III	III	-	-
6	M	35	T4a	Tentorial	Left	Solid	Sporadic	Absent	I	I	I	-	Regrowth but was operated before in another place
7	F	22	T4b	Lateral	Right	Solid	Sporadic	Absent	I	I	I	-	-
8	F	17	T4a	Tentorial	Right	Solid	Sporadic	Present	I	I	I	-	-
9	M	19	T4a	Tentorial	Right	Solid	Sporadic	Absent	I	I	I	-	Regrowth
10	M	29	T4a	Lateral	Right	Solid	Sporadic	Absent	I	II	II	-	-
11	M	38	T4a	Lateral	Left	Solid	Sporadic	Absent	I	III	IV	-	-
12	M	36	T4a	Tentorial	Left	Solid	Sporadic	Absent	I	IV	IV	-	-
13	F	48	T4a	Tentorial	Left	Solid	Sporadic	Absent	I	II	II	-	-



14	F	64	T4a	Tentorial	Right	Solid	Sporadic	Absent	I	IV	IV	-	-
15	F	56	T4a	Tentorial	Right	Solid	Sporadic	Absent	I	III	III	-	-
16	M	40	T4a	Lateral	Right	Solid	Sporadic	Absent	I	V	V	Masseteric facial anastomosis	-
17	F	17	T4a	Lateral	Left	Cystic	NF2	Absent	I	II	Death due to pneumonia	-	-
18	F	48	T4a	Tentorial	Right	Solid	Sporadic	Absent	I	III	III	-	-
19	F	27	T4b	Laterotentorial	Right	Solid	Sporadic	Absent	III	IV	II	Facial and eyelid reconstruction	Regrowth but was operated in another place
20	F	25	T4a	Tentorial	Right	Solid	Sporadic	Absent	I	II	I	-	-
21	M	28	T4b	Laterotentorial	Right	Solid	Sporadic	Absent	II	III	IV	-	-
22	F	23	T4a	Tentorial	Left	Solid	Sporadic	Present	I	I	I	-	-
23	F	54	T4a	Tentorial	Right	Solid	Sporadic	Absent	I	III	III	-	-
24	M	36	T4a	Tentorial	Left	Solid	Sporadic	Absent	I	II	III	-	-
25	F	57	T4a	Tentorial	Left	Solid	Sporadic	Absent	I	II	II	-	-
26	F	46	T4a	Lateral	Left	Solid	Sporadic	Absent	I	III	II	Masseteric facial anastomosis	-
27	M	72	T4b	Laterotentorial	Right	Cystic	Sporadic	Present	I	I	I	-	-
28	F	29	T4a	Laterotentorial	Right	Cystic	Sporadic	Absent	I	III	II	Masseteric facial anastomosis	Regrowth
29	F	53	T4a	Lateral	Right	Cystic	Sporadic	Present	I	II	II	-	-
30	F	40	T4a	Lateral	Left	Solid	Sporadic	Absent	I	III	-	-	-

31	M	30	T4a	Tentorial	Right	Solid	Sporadic	Absent	I	I	I	-	-
32	F	50	T4b	Laterotentorial	Right	Solid	Sporadic	Absent	I	III	II	-	-
33	F	51	T4a	Tentorial	Right	Solid	Sporadic	Absent	I	III	II	-	-
34	F	34	T4a	Tentorial	Left	Solid	Sporadic	Present	I	II	II	-	-
35	F	35	T4a	Lateral	Left	Solid	Sporadic	Absent	I	II	-	-	-
36	F	42	T4a	Lateral	Right	Solid	Sporadic	Absent	I	III	-	-	-

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Table 1B. Data obtained from medical records of the 36 patients included in the study

n	Shunt	Positioning 1	Positioning 2	Bone opening	Intraoperative evoked potential monitoring	Drop in intraoperative evoked potential	Extent of surgical resection	Radiosurgery	Fistula after surgery	Post-operative complications	Tumor progression in 1 year
1	-	DDP w/ head rotation	-	Craniectomy	No	No	GTR	No	No	No	No
2	-	DDP w/ head rotation	-	Craniectomy	No	No	GTR	No	No	No	No
3	EVD	Semi seated	-	Craniectomy	Yes	Yes	GTR	No	No	No	-
4	-	Semi seated	-	Craniectomy	Yes	No	GTR	No	No	No	-
5	-	Semi seated	-	Craniotomy	Yes	No	GTR	No	Liquoric fistula	No	No
6	-	Semi seated	-	Craniectomy	Yes	No	GTR	No	No	No	No
7	EVD	Semi seated	-	Craniectomy	Yes	No	STR	No	No	Hematoma	No
8	-	Semi seated	-	Craniectomy	Yes	No	GTR	No	No	No	No
9	-	Semi seated	DDP w/ head rotation	Craniectomy	Yes	No	STR	No	No	No	No
10	-	DDP w/ head rotation	-	Craniectomy	Yes	No	GTR	No	No	No	No
11	-	Semi seated	-	Craniectomy	Yes	No	GTR	No	Liquoric fistula	No	No
12	-	Semi seated	-	Craniectomy	Yes	No	GTR	No	No	No	No
13	-	Semi seated	-	Craniectomy	Yes	No	GTR	No	No	No	No
14	-	DDP w/ head rotation	-	Craniectomy	Yes	No	GTR	No	No	No	No
15	VP	DDP w/ head rotation	-	Craniectomy	Yes	No	GTR	No	No	No	No
16	-	DDP w/ head rotation	-	Craniectomy	Yes	No	GTR	No	No	No	No
17	-	DDP w/ head rotation	-	Craniectomy	Yes	No	GTR	No	Liquoric fistula	Hematoma	-
18	-	DDP w/ head rotation	-	Craniectomy	Yes	No	GTR	No	No	No	No
19	EVD	DDP w/ head rotation	-	Craniotomy	Yes	No	GTR	No	Endolymphatic fistula	No	No

20	-	Semi seated	-	Craniectomy	Yes	Yes	GTR	No	No	Pons Stroke	No
21	VP	Semi seated	-	Craniotomy	Yes	No	GTR	No	No	No	No
22	-	Semi seated	-	Craniotomy	Yes	No	GTR	No	No	No	Yes
23	-	DDP w/ head rotation	-	Craniectomy	Yes	No	GTR	No	No	Mesencephalon bleeding	No
24	-	Semi seated	-	Craniectomy	Yes	No	GTR	No	No	No	No
25	-	DDP w/ head rotation	-	Craniectomy	Yes	No	GTR	No	No	No	Yes
26	-	Semi seated	-	Craniotomy	Yes	No	GTR	No	No	No	No
27	-	DDP w/ head rotation	-	Craniectomy	Yes	Yes	Debulking	No	No	No	No
28	-	Semi seated	DDP w/ head rotation	Craniotomy	Yes	No	Debulking	Yes	Liquoric fistula	No	No
29	-	Semi seated	-	Craniotomy	Yes	No	GTR	No	No	No	No
30	-	DDP w/ head rotation	-	Craniotomy	Yes	Yes	GTR	No	Liquoric fistula	No	-
31	-	Semi seated	-	Craniectomy	Yes	No	GTR	No	No	No	No
32	EVD	DDP w/ head rotation	-	Craniotomy	Yes	Yes	Debulking	No	No	No	No
33	-	Semi seated	-	Craniotomy	Yes	No	GTR	No	No	No	No
34	-	Semi seated	-	Craniotomy	Yes	No	GTR	No	No	No	No
35	-	DDP w/ head rotation	-	Craniectomy	Yes	No	GTR	No	No	No	-
36	-	DDP w/ head rotation	-	Craniectomy	Yes	Yes	GTR	No	No	No	-

n: patient; M: male; F: female; VP: ventriculoperitoneal; EVD: external ventricular drain; HB: House-Brackmann scale; DDP: Dorsal Decubitus Position; GTR: Gross Total Resection; STR: subtotal resection

Figure 1: 22-year-old female patient, with a T4B (Hannover Classification) Vestibular Schwannoma. A: Axial MRI T1 with gadolinium showing the tumor at the left cerebellopontine angle. B: Axial MRI T1 with gadolinium showing total resection of giant vestibular schwannoma. C: Debulking of the giant vestibular schwannoma using the two hands technique to dissect its pseudocapsula. D: After total removal of the tumor, it is possible to see the preserved facial nerve (blue arrow) and brain stem (white arrow). E and F: Post-operative picture of the patient showing the preserved facial nerve.

The assessment of the patients' age presented normal distribution, with a mean of 37.9 years, standard deviation of 13.7 years and values minimum and maximum of 17 and 72 years, respectively. Females were more frequent (23/36; 63.9%) than males (12/36; 36.1%). Only 6 patients (16.7%) had T4b tumors according to the Hannover classification, and the others T4a (30/36; 83.3%). Most tumors were in the right hemisphere (20/36; 55.6%) and had a solid consistency (31/36; 86.1%). Cystic tumors were found in only 5 patients (5/36; 13.9%), and 1 of these patients had type 2 neurofibromatosis. The etiology of the other 35 patients' tumors was sporadic. As this is a study of giant VS, with tumors classified as T4a and T4b only, only 5 patients (8.3%) had hearing before surgery. Five patients (13.9%) had already presented tumor regrowth, having been previously submitted to resection. Facial nerve functionality before surgery was considered good, with 33 patients classified as I on the HB scale (91.7%), 1 as II (2.8%) and 2 as III (5.5%). Immediately after surgery, these values changed to 8 with I (22.2%), 9 with II (25.0%), 13 with III (36.1%), 4 with IV (11.1%) and 2 with V (5.6%).

Anastomosis of the facial nerve was performed in 4 patients after surgery, the techniques were: anastomosis of the hypoglossal and facial nerves or anastomosis of the masseteric and facial nerves.

Only 1 patient underwent postoperative tarsorrhaphy. In the postoperative period, 2 patients died (glottis edema and pneumonia). In addition, 4 patients were not evaluated after 6 months and 1 year due to lack of clinical follow-up.

In 6 patients (16.7%) some type of shunt was placed, 3 of them ventriculoperitoneal (VP) and the other 3 external ventricular drain (EVD).

Regarding the positioning and bone opening of the patient during surgery, 55.6% (20/36) underwent surgery in the semi-seated position and the others (16/36; 44.4%) in dorsal decubitus position with head rotation, and in 69.4% (25/36) craniectomy was performed and in 30.6% (11/36) craniotomy.

Only 2 patients were not submitted to intraoperative monitoring with evoked potentials in the facial nerve and those surgeries were performed in 1995 and 1999. Of those monitored, 6 had a drop in evoked potential.

Most tumors were resected as GTR (31/36; 86.1%), and the others as STR (2/36; 5.6%) or debulking (3/36; 8.3%). Radiosurgery was performed in only 1 patient before resection.

Regarding post-surgical complications, 9 patients (25.0%) had them, which were cerebrospinal fluid (CSF) fistula, endolymphatic fistula, pons stroke, midbrain bleeding and/or hematoma. As mentioned above, 2 patients died after surgery, one of which had no other post-surgical complications and the other had CSF fistula and hematoma). It is important to note that no patient had air embolism.

After 6 months of surgery, the facial nerve functionality found was 8 patients with HB I (26.7%), 11 with HB II (36.7%), 6 with HB III (20.0%), 4 with HB IV (13.3%) and 1 with HB V (3.3%). After 1 year of follow-up, tumor regrowth was found in only 2 patients (6.7%). Table 2 summarizes the facial nerve (FN) functionality values before surgery, in the immediate postoperative period and after 6 months of follow-up, considering good HB I or II function, regular HB III or IV, and poor HB V and VI. In terms of facial nerve functionality, considering good HB values (I or II) and/or maintenance of the pre-surgical condition, we obtained 66.7% (20/30) of preservation 6 months after tumor resection.

Regarding the associations made in this work, the results can be seen in Tables 2-9. A significant correlation was found between facial nerve function in the immediate postoperative period and intraoperative monitoring of this nerve with evoked potential ( $p=0.024$ ). Thus, it can be said that 100% of patients who were not submitted to this monitoring had worse facial nerve function indexes (HB IV and V) in the immediate postoperative period when compared to those who were submitted. The extent of surgical resection showed a significant difference with the classification of tumor size ( $p=0.024$ ), with GTR being possible in 93.4% of T4a tumors, while this value was 50% in T4b.

Considering p values between 0.05 and 0.09, it can be said that there was a trend towards a significant difference when comparing facial nerve function before surgery with tumor classification ( $p=0.066$ ) and post-operative fistula with the type of bone opening ( $p=0.057$ ). Consequently, 16.7% of patients with T4b tumors had worse pre-surgical functionality (HB > II) compared to T4a tumors, whose value was only 3.3%. In addition, 36.4% of the patients undergoing craniotomy had a postoperative fistula, while craniectomy had only 8% of this complication.

## **Discussion**

One of the main goals of giant VS resection is the preservation of facial nerve functionality. The data found in this study regarding the use of facial nerve evoked potential monitoring are aligned with the current trend in the literature. The non-use of intraoperative evoked potential was related to worse facial nerve functionality within the immediate postoperative period ( $p=0.024$ ). The use of evoked potential since its implementation resulted in significantly lower decay of facial nerve functionality.[1] In the study by Matthies et al. it is possible to analyze the drop in percentage of evoked potential on the facial nerve and its correspondence to the worst outcomes regarding its functionality, demonstrating how fundamental this technique is in the contemporary scenario.[13]

In addition to the use of evoked potential monitoring, it is necessary to adjust the threshold decay of the potential, in order to improve its sensitivity.[1] The criterion of decay alarm in our study was 50%. Earlier alarms (with a smaller decay), while they may decrease the chances of facial nerve involvement, may also restrict the extent of tumor removal. That way it is recommended to apply a maximum of 50% decay.[1]

A trend towards significance was found between preoperative facial nerve functionality and tumor size classification, with 16.7% of patients with T4b tumors having the worst functionality (HB > II) compared to T4a tumors whose value was 3.3% only ( $p=0.066$ ). In giant VS, the location of this nerve changes and it tends to become thinner by the tumor mass effect and relationship with the vestibular nerve.[11] Thus, the larger the size of the tumor, the greater the previous involvement of the facial nerve, in agreement with what is described in the literature.[8]

Our study did not find a relationship between tumor consistency and facial nerve functionality ( $p=1.000$ ) or post-surgical complications ( $p=0.119$ ), although cystic tumors presented higher rates of complications than solid ones (60.0% vs. 22.6%, respectively). The study by Samii and Matthies found that cysticity decreases facial nerve preservation and increases the risk of complications.[19] In addition, Han et al. identified that cystic VS tend to be larger than solid ones at the time of diagnosis.[9] Cystic VS are rarer and more aggressive than solids, accounting for 5.7-20% of all VS. [16] These tumors generally grow faster and adhere more to the facial nerve and other structures, making their resection more difficult. As a result, surgical and facial nerve function outcomes are less favorable in those cases.[12, 18] However, other articles found a significant association between VS with cystic components and better results of facial nerve functionality due to faster and earlier decompression of the lesion.[2, 14] On the other hand, tumors with more than 90% of cystic component had greater difficulty in preserving this nerve due to the lack of a well-defined plane between the arachnoid membrane and the thin tumor capsule. [14]

Hearing function is affected in most patients with VS, especially in those with giants tumors. [8, 14, 18, 21] In the current study, only 5/36 (13.9%) had some degree of hearing preservation before surgery, and in the postoperative period these patients had their hearing abolished. According to a work by Schneider et al. with 37 patients with giant VS, 75.68% (28/37) of them had hearing loss as the most common symptom at presentation.[21] In the study by Samii et al., among the 50 patients in the group with tumors larger than 4 cm, all of them had some degree of hearing impairment, with 9 (18%) of these having a minor dysfunction degree (classes 1 and 2 of the New Hannover Classification). There was hearing preservation in only 1 patient ( $p<0.05$ ) in this group who had the minor dysfunction.[18]

The meta-analysis by Zou et al. [23], as well as our work, considered HB I and II as good facial nerve function, HB III and IV as regular and HB V and VI as poor. The proportions found of good, regular and poor facial nerve function in the postoperative period were, respectively, 62.9% (95% CI, 50.0-74.9%;  $I^2=91.1\%$ ), 25.8% (95% CI, 18.2-34.2%;  $I^2=81.9\%$ ) and 8.9% (95% CI, 3.3-16.7%;  $I^2=89.1\%$ ). It was commented that facial nerve function generally improved during follow-up, but the presence of paralysis in the immediate postoperative period indicated a worse prognosis.

In addition, with 1 year of follow-up, patients with HB equal to or less than III generally showed gradual improvement, while patients with HB greater than or equal to IV were at higher risk of maintaining this impaired function.[23] Our study showed similar results, with better facial nerve function after 6 months than in the immediate postoperative period. These values were 63.4% (19/30) for good function, 33.3% (10/30) for regular and 3.3% (1/30) for poor, and we had no patients with HB VI (Table 2).

Schneider et al. [21] also evaluated the facial nerve function 6 weeks and 1 year after surgery and compared the groups with GTR and NTR (Near Total Resection). After 6 weeks, in the GTR group, 44.4% (12/27) presented HB I or II and, in the NTR group, 90% (9/10), with a significant difference between the groups ( $p < 0.02$ ). At 1 year of follow-up, the facial nerve function values with HB I or II were 78% (21/27) for GTR and 100% (10/10) for NTR, with significance ( $p < 0.0001$ ). This suggests that there is a significant difference in the outcome of facial nerve function depending on the extent of tumor resection. [21] Our study found values of 45.2% (14/31) of HB I or II with GTR, 100% with STR (2/2) and 33.3% (1/3) with debulking in the immediate postoperative period and 56.0% (14/25) with GTR, 100% (2/2) with STR and 100% in debulking (3/3) after 6 months of follow-up. Although an improvement in the number of patients with HB I or II was observed regardless of the extent of resection, these values were not statistically significant.

The post-surgical complications rate was compared with the patient positioning type (semi-seated vs dorsal decubitus with head rotation). However, our sample did not show statistically significant data. Recent data by Schackert et al. [20], in a study with 544 patients, demonstrated that in patients with VS classified as T3 and T4, the use of the semi-seated position significantly reduced surgical time and blood loss. The same study evaluates complications related to this position, and one of the most cited in the literature is its potential association with pulmonary air embolism. When comparing the lateral decubitus vs semi-seated, they found a value for this complication of 0/68 (0%) vs 7/163 (4.3%), respectively, however without statistical significance ( $p = 0.108$ ). Analyzing these risks, Saladino et al. [17], in a study with 425 patients who underwent surgical resection in the semi-seated position, found a rate of 21% of pulmonary air embolism.[17] They found that the highest risk was at the time of the skin incision until the opening of the dura mater. The same authors propose the use of pulmonary air embolism protocols of diagnosis and treatment in order to mitigate the risks.

We found a trend of significance regarding bone opening and postoperative fistula ( $p = 0.057$ ). Craniectomy had better results than craniotomy, with 92.0% of patients undergoing craniectomy having no postoperative fistulas, while this value was 63.6% for craniotomy. However, a recent meta-analysis with pediatric patients found that the rate of cerebrospinal fluid leakage was 4.4% in patients undergoing resection of intradural tumors (8.0% in the posterior fossa tumors subgroup), and this risk was higher in patients undergoing craniectomy compared to those undergoing craniotomy (OR 4.7, 95% CI 1.7-13.4).[28] This comparison is scarce in the literature, requiring further studies.



## Conclusion

The main factor to be considered in the treatment of giant VS is the maximum safe resection. Among the major challenges that involve its resection, the preservation of the facial nerve and avoidance of post-surgical complications should also be considered.

Our study proved that intraoperative monitoring of the facial nerve with evoked potentials improved facial nerve functionality. Unfortunately, the use of evoked potentials is still not a reality for many centers, especially the public ones. We hope that in the future, with more studies showing the same results, this might change.

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# Abdominal cerebrospinal fluid pseudocyst: a comparative analysis between children and adults

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## Abstract

**Purpose** Abdominal cerebrospinal fluid (CSF) pseudocyst is a rare but important complication in patients with ventriculoperitoneal shunt (VPS). In addition to presenting our experience, we performed a comparative analysis between children and adults with this entity. To the author's knowledge, there are no studies in which this condition has been compared.

**Methods** The PubMed database was searched for all relevant reports published from 1954 to 2012. The differences were statistically compared, especially regarding clinical investigations, etiology of the hydrocephalus, shunt revision, CSF infection, treatment, and recurrence. Chi-square test or Fisher's exact test was used to find associations among the variables.

**Results** Compiled from literature, we found 393 cases of abdominal pseudocyst: 295 children, including our cases, and 55 adults, with age not informed in 43 cases. In children, 33 % of the patients have a positive culture on presentation, with higher incidence in children younger than 10 years. In contrast, only 15 % among adults were positive CSF culture. In total, 287 abdominal pseudocyst cases who underwent shunt revision have been reported; 78.4 % of children and 62.2 % of adults. The main occurrence of this complication according to the etiology of hydrocephalus in children was different from adults. The recurrence of pseudocyst occurred in 19.8 and 24.2 % of children and adults, respectively.

**Conclusions** The differences between children and adults might represent distinct trends on the etiology and treatment of this entity. Hence, additional well-designed cohort studies will be necessary to strengthen our findings.

**Keywords** Cerebrospinal fluid pseudocyst · Ventriculoperitoneal shunt · Shunt complication · Abdominal pseudocyst · Hydrocephalus

## Introduction

The modern ventriculoperitoneal shunt (VPS) era started in 1948 by Cone, Lewis, and Jackson [26, 52] and later by Ames, who between 1950 and 1957 performed several polyethylene tubing VPS with almost unsatisfactory results [6, 7]. Now, VPS is the standard therapy for the management of hydrocephalus. However, as more patients with hydrocephalus survive and live longer, more complications developed [59]. Approximately 5 to 47 % of abdominal complications by VPS are reported [44, 71]. One of the less frequent but important complications is the abdominal CSF pseudocyst, with a reported rate of less than 1 % on all patients with shunt [14], although higher rates were reported as well [94]. Even more uncommon are hepatic [37, 62, 65, 66, 85, 95, 110] or splenic pseudocyst as a complication of VPS insertion [72].

The cyst is called pseudocyst because its walls consist only of peritoneal serous membrane, thickened by a chronic inflammatory process [18, 70]. The invasion of lymphocytes in the specimen may have suggested that the inflammatory reaction to the shunt tube was associated with cyst formation [80]. We agree with others who prefer the term "abdominal pseudocyst" [31, 84, 94, 98, 103, 105] rather than peritoneal

[14, 18, 45, 52], intraperitoneal [75], or omental cyst [41].

The aim of this paper was to report a comparative analysis between children and adults with abdominal CSF pseudocyst

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on the clinical presentation, etiology of the hydrocephalus, rate of CSF infection and shunt revision, treatment, and results on the literature of the last 58 years. In this review, we have included five new cases treated at our institution (Figs. 1 and 2).

## Methods

An analysis of the 502 shunt placement procedures performed for an 8-year period (January 2005 to December 2012) at our hospital, revealed that five patients developed abdominal CSF pseudocyst. Details of history, clinical features, radiological finding, cerebrospinal and abdominal CSF pseudocyst culture, and clinical outcomes were summarized (Table 1). In addition, the information for this review was retrieved from the author's files and the National Library of Medicine database (PubMed) for the period 1954–2012 using the following keywords: “intra-abdominal pseudocyst”, “cerebrospinal fluid pseudocyst”, “abdominal cerebrospinal fluid pseudocyst”, “peritoneal pseudocyst”, “intraperitoneal pseudocyst”, “omental cyst”, “shunt infection”, and “shunt complications.” In total, 100 articles of case-control studies, case reports, and case series were included in our revision (Table 2).

Our reviewed included 393 cases of abdominal pseudocyst related to the use of VPS. Because of the possible difference between children and adults, the analysis was performed for each subgroup, considering individuals younger than 19 years as children and individuals older than 19 years as adults. Chi-square test or Fisher's exact test was used to find associations among the variables and verified the odds ratio for recurrence related to the etiology of hydrocephalus, CSF infection and surgical treatment. The Stata software version 13.0 (Copyright

1985–2013; Stata Corp. LP, College Station, TX) was used for statistical analysis. A  $p$  value  $< 0.05$  was considered significant.

## Discussion

### Epidemiology

Since Harsh [54] described in 1954 a periumbilical cyst associated among 12 ventriculofallopian shunts and Jackson and Snodgrass [60] also reported another case 1 year later, large series were published in the world literature [14, 36, 42, 52, 53, 63, 74, 94, 98], some of them in Latin America [31, 38, 102]. Our literature review included a total of 100 articles, including five cases of the current study, totaling 393 cases of abdominal CSF pseudocyst since the first case reported by Harsh [54] (Table 2). According to several authors, this condition accounted for approximately 0.25 to 10 % of all VPS [11, 14, 18, 36, 52, 71, 74, 94, 103]. In our series, abdominal pseudocyst formation represents only 1 % of all patients with VPS (5/502). However, the prevalence rate found in 21 case-control studies, including our study, recompiled from the literature was 2.3 % of the cases based on the total of patients evaluated, that is, 10,803 controls with 183 cases [4, 10, 11, 14, 18, 29, 33, 36, 49, 52, 53, 60, 71, 74, 90, 91, 94, 101, 103, 108].

Many authors have reported this complication in pediatric patients because hydrocephalus is more common in children. However, in a series, the child-adult ratio was 1.8:1 [94]. According to this survey, 295 children, including our cases, and 55 adults have been reported until 2012; age was not provided in 43 cases (Fig. 3). There are reports of abdominal CSF pseudocyst in a 75-year-old man and an 84-year-old woman [100, 109]; on the other hand, one of the youngest patients was 2 months old [94, 102]. In children, our study identified 48.2 % males and 51.8 % females, and among adults, 43 % males and 57 % females. We found a mean age of  $7.2 \pm 4.7$  years in children; 80 % of the cases were younger than 10 years, and only 20 % were children between 10 and 19 years old. In adults, the mean age was  $36.7 \pm 15.1$  years. We observed a proportion of 47.3 % of the cases for adults 30 years and younger and 52.7 % for adults older than 30 years (Table 3).

### Predisposing factors

Although the etiology of abdominal CSF pseudocyst is not clear, different authors attribute cyst formation to several predisposing factors, with no clinical or scientific support. The most frequent factor is the inflammatory process, either sterile or infectious. In some patients, it could be an inflammatory response to some components of the shunting system or to a high concentration of protein in the peritoneal cavity

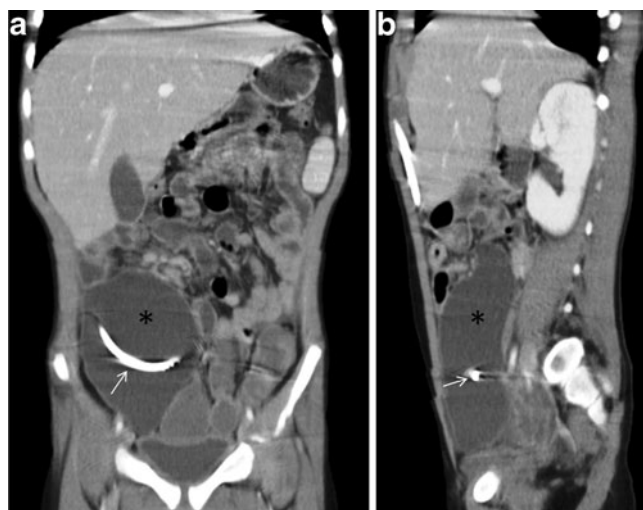
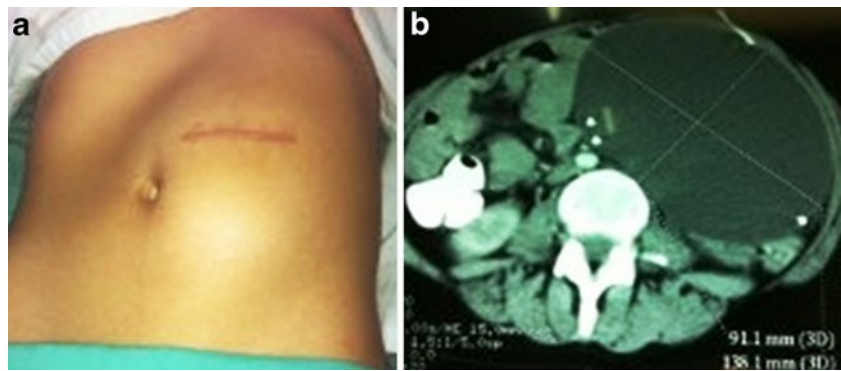


Fig. 1 a Abdominal CT scan (coronal view) showing a large abdominal pseudocyst (asterisk) with the distal segment of the VPS (arrow) located within the cyst. b Abdominal CT scan (sagittal view) demonstrating the pseudocyst (asterisk) pushing the bowels superiorly

Fig. 2 a Photograph showing an abdominal mass that regularly contoured at the left flank in the abdomen. b Abdominal CT scan showing a large cystic mass measuring 13×9 cm



[2, 28, 30], associated sometimes with an antigen–antibody reaction by an unidentified protein moiety in the CSF [8]. This hypothesis would explain the cases of abdominal pseudocyst in which CSF culture proved negative. Other predisposing causes are as follows: (a) prior abdominal surgeries or multiple revisions of the shunt in the abdomen; (b) allergic reactions to silicone or ethylene oxide [55] and a reaction due to the starch granules from the surgical gloves; for this reason, some neurosurgeons recommend the use of latex-free gloves while inserting a VPS or excising pseudocyst [103]; and (c) in a lesser proportion, liver dysfunction [67].

Most authors agree that infection is the main cause of the formation of abdominal CSF pseudocyst [18, 33, 73]. One paper reviewed eight studies with 128 cases showing that infection rates vary between 17 and 80 %, with an average of 42 % [74]. Some neurosurgeons recommend a routine culture of the abdominal catheter tip. In the series of Salomão et al. [102], cultures of the CSF and the tip were positive in 44.4 and 61 % of the samples, respectively. In their opinion, the latter seems to be more reliable. The most common organisms isolated from the culture are

*epidermidis* and *Staphylococcus aureus*. According to Erşahinet al. [36], when the abdominal CSF pseudocyst associated with a shunt infection occurs within 1 year after a shunting procedure, the probable organism is *S. epidermidis*. Mobley et al. [74] stated that it is possible to find slow-growing bacteria such as *P. acnes* in culture more than 7 days. Some few isolated cases of infected abdominal CSF pseudocyst with other bacteria have been reported [36, 56, 63, 74, 102]. In contrast, similar to our five cases, Bartolek et al. [11] did not find infection signs in their five patients, nor did Sena et al. [105] and Agha et al. [4].

In our analysis of 295 pediatric cases, 92 patients (33 %) have a positive culture on presentation, 188 cases (67 %) have sterile CSF, and 15 children have infection as missing data. The positive culture rate was 41 % for those 5 years and younger, 39 % for children between 5 and 10 years old, 16 % for patients between 10 and 15 years old, and only 4 % positive culture for those 15 years and older. In contrast, only 6 cases (15 %) among 41 adults were infected. The positive culture rate was 66 % for those 30 years and younger. The other 14 adults were not informed of the CSF culture. This information confirms

Table 1 Clinical characteristics of five patients with abdominal CSF pseudocyst

Patient no.	1	2	3	4	5
Sex/age	F/1 years	F/14 years	F/2.5 years	F/6 years	F/6 years
Hydrocephalus etiology	Congenital	PMTB	MM	Dandy Walker	Congenital
Age at last VPS	3 months	13.5 years	1 month	2.2 years	16 days
Previous shunt revision	Yes	No	No	No	No
Shunt	Malfunction	Malfunction	Malfunction	Function	Function
Symptoms	Abdominal mass and pain, headache, vomiting	Abdominal mass and pain, sonolence	Abdominal pain and distention, headache, vomiting	Abdominal pain and distention, tenderness	Abdominal pain, distention, abdominal mass
Abdominal CT scan	4.5×6.5 cm	9.1×13.8 cm	7×12 cm	4×3 cm	8.1×10.7 cm
CSF culture	Negative	Negative	Negative	Negative	Negative
Fluid pseudocyst protein	1,350 mg/dl	1,250 mg/dl	1,098 mg/dl	501 mg/dl	827 mg/dl
Follow-up	2 years	1 year	1 year	1 year	1 year
Outcome	Resolution	Resolution	Resolution	Resolution	Resolution

Abbreviations: CSF cerebrospinal fluid, MM myelomeningocele, PMTB postmeningitis tuberculosis, VPS ventriculoperitoneal shunt

Table 2 Review of the literature on abdominal CSF pseudocysts(1954–2012)

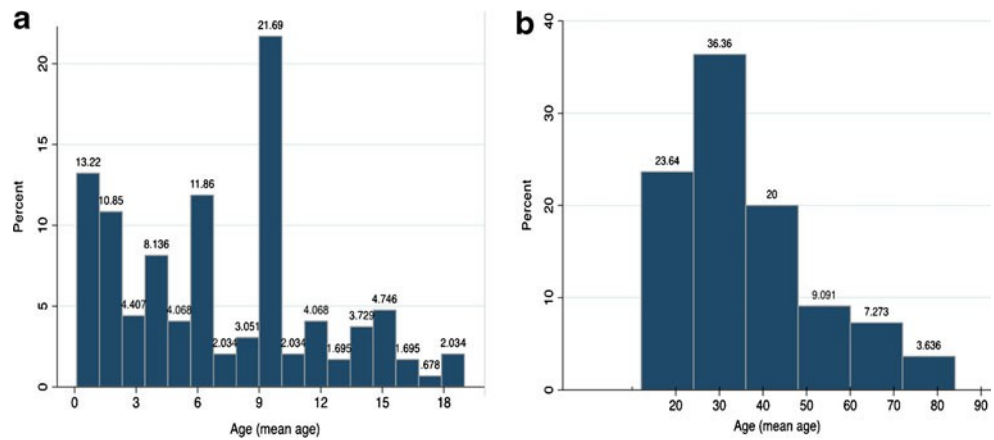
First author/reference	Year	N
Achayra [1]	2001	1
Agarwal [3]	2009	1
Agha [4]	1983	6
Aguirre-Rivero [5]	1998	6
Anderson [8]	2003	1
Aparici-Robles [9]	2008	6
Bartolek [11]	2010	5
Baumgartner [12]	1990	1
Bauni [13]	2007	2
Besson [14]	1995	22
Birbilis [15]	2008	1
Briggs [16]	1984	5
Bryant [17]	1988	5
Burchianti [18]	1988	7
Buyukyavuz [19]	2012	1
Castellucio [20]	2006	1
Chandra [21]	1992	2
Chitkara [22]	2004	1
Chuang [23]	1978	2
Chung [24]	2009	6
Coley [25]	2004	7
Coşkun [27]	1999	1
Davidson [30]	1975	1
De Oliveira [31]	2007	12
Deindl [32]	1986	1
Egelhoff [33]	1986	8
Ekong [34]	1979	3
Engelhard [35]	1992	1
Erşahin [36]	1996	10
Faraj [37]	2011	1
Figueiredo [38]	1981	3
Fischer [39]	1969	3
Fortea-Sanchis [40]	2011	1
Gamal [41]	1988	1
Gaskill [42]	1989	12
Gebarski [43]	1984	1
Ghidirim [44]	2010	1
Ghritlaharey [45]	2006	3
Goldfine [47]	1978	1
Gomutbutra [48]	2004	1
Grosfeld [49]	1974	3
Grunebaum [50]	1988	5
Guice [51]	1978	1
Gutierrez [52]	1976	11
Hahn [53]	1986	26
Harsh [54]	1954	1
Hashimoto [55]	2004	1
Hernández [56]	2004	1
Horikawa [57]	1999	1
Hsieh [58]	2006	1
Jackson [60]	1955	1

Table 2 (continued)

First author/reference	Year	N
Jain [61]	2003	1
Kaplan [62]	2007	1
Kariyattil [63]	2007	15
Kim [64]	1995	1
Koçak [65]	2004	1
Kolić [66]	2010	1
Latchaw [67]	1981	1
Lee [68]	1978	1
Leung [69]	2010	1
Mobley III [74]	2005	36
Nakagaki [75]	1979	1
Nfonsam [76]	2008	4
Norfray [77]	1979	1
Nugent [78]	1986	1
Oh [79]	2001	1
Ohba [80]	2012	1
Palomar [81]	1977	2
Parrish[82]	1973	1
Parry [83]	1975	4
Pathi [84]	2004	1
Peltier [85]	2011	1
Pérez Moreno [86]	2012	1
Pernas [87]	2004	1
Popa [90]	2009	2
Price [91]	1981	2
Raghavendra [93]	1981	3
Rainov [94]	1994	14
Rana [95]	1985	1
Redman [96]	1977	1
Roitberg [98]	1998	27
Rovlias [99]	2001	1
Ruiz-Tovar [100]	2010	2
Rush [101]	1985	4
Salomão [102]	1998	18
Sanal [103]	2007	8
Seçer [104]	2011	1
Sena [105]	2010	1
Sharma [106]	2004	1
Sivalingam [107]	1976	1
Suematsu [108]	1984	2
Takeuchi [109]	2012	1
Verma [110]	2012	1
Wang [111]	2012	1
Wang [112]	1989	1
White [113]	1991	6
Wolbers [114]	1987	1
Yamamoto [115]	1979	1
Yamashita [116]	1990	1
Yuh [117]	2012	1
Dabdoub	2013	5
Total		39

Fig. 3 Histogram of age between

a children and b adults. Distribution of age among children, showing that the vast majority of the sample had children younger than 10 years, 13 % had younger than 1 year, and only 4.4 % had 15–19 years old. Among adults, we observed a right-skewed distribution, showing that most individuals were 20 to 25 years (25.5 %) and 20 % were older than 45 years



that the prevalence of infection is lower in adults than that in children and shows a higher prevalence in children younger than 10 years ( $p=0.042$ ) and in adults younger than 30 years ( $p=0.355$ ). Furthermore, some authors suggest that a smaller pseudocyst tends to be infected and a larger abdominal CSF pseudocyst tends to be sterile [46, 84].

Table 3 Clinical characteristics and etiology of hydrocephalus reported in the literature in children ( $n=295$ ) and adults ( $n=55$ )

	Children		Adults	
	<i>n</i>	%	<i>n</i>	%
Age <sup>a</sup>	7.2±4.7		36.7±15.1	
Gender <sup>b</sup>				
Male	136	48.2	22	43.1
Female	146	51.8	29	56.9
Etiology				
Congenital HCF	64	21.7	03	5.5
Myelomeningocele	57	19.3	03	5.5
Intraventricular hemorrhage	43	14.6	06	11
Previous infection	23	7.8	09	16.3
Tumor	23	7.8	12	21.8
Aqueductal stenosis	16	5.4	02	3.6
Dandy-Walker cyst	15	5	0	0
Trauma	05	1.7	04	7.2
Chiari malformation	04	1.3	01	1.8
TB meningitis	12	4.1	01	1.8
Others	20	6.8	06	11
Unknown	13	4.4	08	14.5
Symptoms				
Abdominal <sup>c</sup>	289	89.9	46	83.6
Neurological <sup>d</sup>	102	38.6	19	34.6

Abbreviations: HCF hydrocephalus, TB tuberculosis

<sup>a</sup> Mean±standard deviation

<sup>b</sup> 17 missing

<sup>c</sup> 29 missing

<sup>d</sup> 31 missing

One of the most suggested predisposing factors of CSF pseudocyst formation is multiple shunt revisions. Hahn et al. [53] found that the average number of previous shunt revisions in patients with abdominal CSF pseudocyst was 11.2, and Rainov et al. [94] described 50 % of the patients with 5 to 10 shunt revisions. This revision rate is significantly higher compared with our cases and other reports [18, 31, 36, 42, 102].

In total, 287 abdominal CSF pseudocyst cases who underwent shunt revision have been reported; 196 (78.4 %) of 250 children and 23 (62.2 %) of 37 adults evaluated were submitted to VPS revision. In children, of the 196 patients who had at least one shunt revision, 68 (35 %) had positive cultures on presentation. On the other hand, of the 54 patients who did not have a revision, only 12 (22 %) had an infection ( $p=0.09$ ). In contrast, of the six adult patients reported with positive CSF culture, two had previous shunt revision (2/19), 2 had not undergone shunt revision (2/12), and the other two were not informed of VPS revision (2/6) ( $p=0.507$ ). Comparing adults and children, we found an association between shunt revision and positive cultures ( $p=0.017$ ), suggesting that children who had a previous shunt revision had the most positive cultures compared with adults, with an odds ratio of 1.84.

Additionally, the etiology of the hydrocephalus has been thought to predispose the development of abdominal pseudocyst. Gutiérrez and Raimondi [52] observed the high incidence of abdominal CSF pseudocyst in patients with Dandy-Walker cyst (4 of 11). Likewise, myelomeningocele seemed to be an important etiology factor for pseudocyst formation in de Oliveira's [31] series (6 of 12). Central nervous system (CNS) tumors have been associated with this pathology, such as astrocytomas, meningioma, papilloma of plexus choroideus, ependymoma, and primitive neuroectodermal tumors [9, 11, 20, 23, 30, 62, 95, 106, 115, 116].

In international literature, the most common etiology-related hydrocephalus in children was congenital hydrocephalus (21.7 %), followed by myelomeningocele (19.3 %), intraventricular hemorrhage in premature infants (14.6 %), brain

tumor (7.8 %), and meningitis (7.8 %). On adult patients, the main causes of hydrocephalus included tumor (21.8 %), meningitis (16.3 %), intraventricular hemorrhage (11.3 %), trauma (7.2 %), and congenital hydrocephalus (5.5 %) (Table 3). Although 21 and 19 % of children had congenital hydrocephalus and myelomeningocele, respectively, these diagnoses do not represent predisposition to positive CSF cultures ( $p=0.421$  for congenital hydrocephalus and  $p = 0.325$  for myelomeningocele). On the other hand, almost 22 and 16 % of the adults had brain tumor and meningitis, respectively, as a main cause of hydrocephalus. Of the adult cases with negative culture, 34 and 20 % had brain tumor and meningitis, respectively, as opposed to none of the adults with these conditions had infection ( $p = 0.106$  for brain tumor and  $p = 0.299$  for meningitis). Hence, the etiology of hydrocephalus has not seemed to be a risk factor in the development of abdominal CSF pseudocyst in either children or adults.

#### Clinical aspects

Although there are no pathognomonic signs in patients with abdominal CSF pseudocyst, the common presentations in children are headache, vomiting, and decreased level of consciousness [94]. Moreover, this pathology may be presented as an acute abdomen [117]; therefore, this presentation in children with VPS requires a skillful diagnostic workup [92, 97]. In adults, the abdominal signs are more important than the infectious signs or neurological complaints, which usually appear days or weeks after abdominal symptoms [42, 77]. Ohba et al. [80] found abdominal symptoms in most of the adult cases, where approximately 30 % presented with symptoms because of shunt malfunction. In abdominal CSF pseudocyst has been also described hyponatremic seizure, hydronephrosis, inferior vena caval obstruction with bilateral hydronephrosis, ureteropelvic junction obstruction, caliectasis with both ureters deviated laterally, cholelithiasis, suggestive of a full-term pregnancy, hemorrhage as a complication of anticoagulant therapy, markedly elevated liver function test, and a right pleural effusion [19, 35, 47, 48, 57, 67, 69, 88, 89, 111].

In our series, three patients (60 %) had shunt dysfunction. However, in literature revision, the neurological symptoms occurred only in 38.6 % of the children and 34.6 % of the adults. Hence, there was no tendency toward presentation with neurological symptoms in the children compared with the adults ( $p=0.569$ , chi-square test). On the other hand, abdominal symptoms were more frequent, representing 89 % of the sample, with 89.9 % in children and 83.6 % in adults. Hence, there was no tendency toward presentation, with abdominal symptoms compared in both groups ( $p=0.184$ , chi-square test). However, to determine the association between the types of symptoms assessed, we identified 35 % of children with both abdominal and neurological symptomatology ( $p=0.000$ ) and 24 % of adults ( $p=0.000$ ), demonstrating that the

association of abdominal and neurological symptoms was obtained for children and adults.

The time interval from the shunt application to the onset of symptoms had a mean of approximately 2 years [18], but frequently within the first 6 months [108]. The shortest interval was 5 days in one patient [24] and 6 days in other patients [52]. The longest interval was 10 years [99, 105] and 15 years [31, 87]. The length of time from the last shunting procedure to the development of abdominal CSF pseudocyst in our series ranged from 6 months to 5.9 years (mean 2.6 years). The fact that abdominal CSF pseudocyst occurs within a short amount of time after a shunt revision implies that an infectious state is probably present, despite the low infection rates published in the literature.

#### Diagnosis

For radiological diagnosis, ultrasound is the method of choice because it is noninvasive, easy to perform, not expensive, and sufficient for a satisfactory diagnosis for larger, localized, or loculated abdominal CSF collection, and generally, it can be displaced in the tip of the peritoneal catheter [7, 42]. Grunebaum et al. [50] drew attention to the sonographic signs of the noninfected pseudocyst, infected pseudocyst, or abscess. For these authors, the noninfected abdominal CSF pseudocyst is characterized by a well-defined sonolucent mass without septa and increased echogenicity beneath it. The shunt tip will produce an echo of higher amplitude than a septum, and the two parallel sides of the shunt will produce the characteristic "railroad sign". In an infected abdominal pseudocyst, a mixture of echoes is demonstrated. The presence of septa or even a fluid level may be detected [93].

The CT scan of the abdomen provides a more accurate diagnosis, especially when these abdominal CSF pseudocysts are large and deform the normal architecture of the abdomen [43]. CT scan may be a better first choice in the initial evaluation to exclude other causes such as peritonitis, appendicitis, volvulus, or diverticulitis [8]. The CT scan shows a cyst containing homogeneous water density fluid with a fine and well-defined margin. The distal catheter of the shunt appears as a higher-density structure with the tip within the pseudocyst [91]. Our patients were diagnosed based on CT scan and reflect the trend for using it as the basis for evaluation, especially when there is a suspected mass.

#### Treatment

On the basis of treatment, there has been no consensus for both handling the VPS as pseudocyst itself because of the treatment changes according to the characteristics of patients, the experience of the surgeon, and the findings during the operation [104]. All patients with abdominal pseudocyst must have CSF from the shunt, the pseudocyst, or both



preoperatively. When the CSF is positive for bacteria on Gramstain or had a suspicious glucose, protein, or white blood cellcount, the treatment tends to be an externalization of the shuntand proper antibiotic therapy [74].

In the absence of infection, the most widely accepted opinions are replacement of the distal catheter into the nonperitoneal space, especially if there are signs of peritonitis and peritoneal adhesions or laparotomy for catheter replace- ment in the opposite quadrant of abdomen pseudocyst. Therefore, removing the peritoneal shunt entirely became preferable [31, 103]. Although an acute infection may not be evident, prophylactic antibiotic therapy is warranted until infection is ruled out [8].

About the treatment of pseudocyst, some authors propose full or partial exeresis by laparotomy or laparoscopy, especial-ly if the large cavity is attached to the abdominal wall [76]. Some surgeons had demonstrated that the simple remotion ofthe catheter diverted the CSF, making the pseudocyst disap-pear [42]. Sonographically guided abdominal CSF pseudocyst aspiration is also an effective technique, allowing the exclu- sion or confirmation of CSF infection and providing the relief of abdominal symptoms [25, 93]. If there is no sign of infec- tion, a single operative shunt revision may be a reasonable andless invasive method of treatment, sparing the patient a surgi- cal procedure and shunt externalization [25].

In summary, the most common definitive procedures in children and adults include seven main categories: (1) directexploratory laparotomy or laparoscopic procedure with ab- dominal pseudocyst excision or aspiration and (a) reposition of the distal shunt into the opposite site of peritoneum, (b) replacement of the distal shunt, and/or (c) reposition of the distal catheter into the nonperitoneal space; (2) direct explor- atory laparotomy or laparoscopic procedure with abdominal pseudocyst excision or aspiration and VPS externalization orexternal ventricular drainage (EVD) and (a) new VPS and/or (b) reposition of the distal catheter into the nonperitoneal space; (3) aspiration only; (4) VPS externalization or EVD and new VPS; (5) reposition of the shunt; (6) replacement ofthe shunt; and (7) removal or reposition of the distal shunt catheter into the nonperitoneal space. Lately, for selected cases, after shunt externalization, de Oliveira et al. [31] had advocated the endoscopic third ventriculostomy in these casesbecause CSF flow is reestablished and the peritoneal cavitywould be preserved for future use.

Regarding the risk of the recurrence of abdominal CSF pseudocyst in the literature, we performed an analysis for each subgroup of children and adults according to the etiology of hydrocephalus, shunt revision, and infection (Tables 4 and 5). The recurrence of abdominal pseudocyst occurred in 19.8 and 24.2 % of children and adults, respectively, with no informa- tion in 48 children and 22 adults. Age was not a relevant factorin the recurrence even for children ( $p=0.198$ ) or adults ( $p= 0.801$ ), with no statistical difference between those who had

Table 4 Proportion of abdominal pseudocyst recurrence risk related tothe etiology of hydrocephalus, shunt revision and infection reported in theliterature in children ( $n=49$ )

	n (%)	OR	IC95 %	p
<b>Etiology</b>				
Congenital HCF	13 (26.5)	1.22	0.7–2.5	0.574
Myelomeningocele	12 (24.5)	1.24	0.6–2.7	0.458
Intraventricular hemorrhage	11 (22.5)	1.75	0.8–3.8	0.153
Previous infection	03 (6.1)	0.65	0.2–2.2	0.505
Tumor	03 (6.1)	0.74	0.2–2.5	0.645
Aqueductal stenosis	02 (4.1)	1.64	0–7.6	0.557
Dandy-Walker cyst	0 (0)	0	0–1.36	0.091
Trauma	0 (0)	0	0–3.89	0.316
Chiari malformation	0 (0)	0	0–7.85	0.479
TB meningitis	0 (0)	0	0–1.36	0.128
Others	03 (6.1)	0.93	0.3–3.2	0.910
Unknown	02 (4.1)	–	–	–
Previous shunt revision	37 (86.1)	2.01	0.8–4.9	0.135
Infection	15 (31.3)	0.95	0.5–1.85	0.871

Abbreviations: OR odds ratio, IC95% 95 % confidence interval, HCF hydrocephalus, TB tuberculosis

the recurrence and those who had not. However, among malechildren, we identified a higher proportion statistically differ- ent ( $p=0.011$ ), representing 65.3 % of the children who hadrecurrence.

The recurrence of abdominal pseudocyst in children sub- mitted previous shunt revision (86 %) found an odds ratio oftwo, although without significant association. The most fre- quent hydrocephalus etiologies found in these children wereco ngenital hydrocephalus (27 %, OR = 1 .2), myelomeningocele (25 %, OR= 1.2), and intraventricular hemorrhage (23 %, OR=1.8); however, they also have not found significant statistical associations. In adults, congenital

Table 5 Proportion of abdominal pseudocyst recurrence risk related tothe etiology of hydrocephalus, shunt revision and infection reported in theliterature in adults ( $n=08$ )

	n (%)	OR	IC95 %	p
<b>Etiology</b>				
Congenital HCF	02 (25)	8.0	0.8–	0.072
Intraventricular hemorrhage	01 (12.5)	1.04	0–9.0	0.969
Previous infection	01 (12.5)	0.45	0–3.6	0.489
Tumor	02 (25)	1.33	0.2–7.8	0.763
Others	02 (25)	–	–	–
Previous shunt revision	02 (40)	0.6	0.09–3.8	0.615
Infection	01 (20)	1.19	0–10.9	0.890

Abbreviations: OR odds ratio, IC95% 95 % confidence interval, HCF hydrocephalus

hydrocephalus (25 %, OR=8,  $p=0.07$ ) was suggested to be a relevant risk factor for CSF abdominal pseudocyst recurrence. With respect to the surgical treatment developed, we described all surgeries reported in the literature for adults and children with the odds ratio of recurrence for each intervention evaluated (Table 6). For children, the odds of recurrence was statistically associated with VPS reposition (odds 9.4 and  $p=0.000$ ). In contrast, the lowest odds of recurrence was identified for repositioning the distal catheter into the nonperitoneal space without exploratory laparotomy procedure after removing the VPS (odds=0.08 and  $p=0.001$ ), and for adults, we have not found any statistical association. Therefore, the re-position of the peritoneal catheter on the abdominal cavity seems to carry more risk of recurrence in children compared with adults, and the reposition of the distal shunt into the nonperitoneal space is not a risk factor for recurrence in children.

By checking the different surgeries performed after or without exploratory laparotomy for CSF pseudocyst excision or aspiration, we found that the number of recurrence in children was higher for treatments without exploratory laparotomy (55 % and  $p=0.853$ ) than for the group with laparotomy technique (39 % and  $p=0.255$ ), but there was no statistical relationship with recurrence. In adults, the situation is different because 75 and 13 % of the number of recurrences were identified for the group of treatment with and without

exploratory laparotomy, respectively. However, we do not identify statistical significance ( $p=0.556$  and  $p=0.544$ ).

In children, the abdominal pseudocyst recurrence with the reposition of the distal shunt after exploratory laparotomy treatment was not statistically significant among patients with negative CSF cultures; however, for treatment with the reposition of the distal shunt without exploratory laparotomy, we identified a suggestive probability for recurrence ( $p=0.050$ ).

## Conclusions

In this literature survey, we have analyzed a relatively large number of abdominal CSF pseudocysts in children compared with adults. The level of evidence in most of the studies in the literature is low. However, from our comprehensive literature review, we conclude the following:

1. As opposed to the literature reported, this review showed no statistical differences in the clinical picture between children and adults with this entity.
2. The prevalence of infection is lower in adults than that in children and higher in children younger than 10 years.
3. We identified a statistical significance association between shunt revision and positive cultures in children when compared with adults.

Table 6 Surgical treatment of abdominal pseudocyst in children ( $n=247$ ), adults ( $n=33$ ) and risk of recurrence

	Children			Adults		
	<i>n</i> (%)	OR	<i>p</i>	<i>n</i> (%)	OR	<i>p</i>
<b>Surgical treatment</b>						
( <sup>a</sup> ) and repositioning of the distal shunt	56 (18.9)	1.56	0.239	23 (41.8)	0.92	0.922
( <sup>a</sup> ) and replacing the distal shunt	23 (7.8)	1.1	0.890	11 (20)	0	0.566
( <sup>a</sup> ) and repositioning the distal catheter into non-peritoneal space	15 (5.1)	0	0.316	04 (7.3)	0	0.409
( <sup>a</sup> ) and ( <sup>b</sup> ) and new VP shunt	13 (4.4)	1.77	0.411	07 (12.7)	1.75	0.566
Aspiration only	03 (1.02)	8.4	0.101	01 (1.8)	0.13	0.072
( <sup>b</sup> ) and new VP shunt	82 (27.8)	0.96	0.928	04 (7.3)	0	0.700
VP shunt repositioned	14 (4.8)	9.46	0.000	03 (5.5)	0	0.566
VP shunt replacement	05 (1.7)	2.77	0.258	02 (3.6)	0	0.409
VP shunt removing and repositioning the distal catheter into non-peritoneal space	43 (14.6)	0.08	0.001	–	–	–
( <sup>a</sup> ) and ( <sup>b</sup> ) and repositioning the distal catheter into non-peritoneal space	11 (3.7)	0	–	–	–	–
( <sup>b</sup> ) and repositioning the distal catheter into non-peritoneal space	18 (6.1)	0	–	–	–	–
Others	08 (2.7)	0	–	–	–	–
NI	04 (1.4)	–	–	–	–	–
<b>Pseudocyst recurrence</b>						
Yes	49 (19.8)	–	–	08 (24.2)	–	–
No	198 (80.2)	–	–	25 (75.8)	–	–

Abbreviations: VP ventriculoperitoneal, EVD external ventricular derivation, VAS ventriculo atrial shunt, NI not informed, OR odds ratio

<sup>a</sup> Direct exploratory laparotomy or laparoscopic procedure with cyst excision

<sup>b</sup> VP shunt externalized and/or external ventricular derivatio

4. We did not identify the predisposing causes of hydrocephalus related to the development of abdominal CSF pseudocyst between children and adults. However, congenital hydrocephalus suggests being a risk factor for recurrence in adults.
5. Age was not a relevant factor in the abdominal CSF pseudocyst recurrence, although there is a higher proportion of recurrence in male children.
6. In children, pseudocyst recurrence was statistically associated in treatment with repositioned distal catheter. In adults, we did not find any statistical association.
7. In children, the number of recurrence was higher for treatments without exploratory laparotomy (55 %) than for the group with laparotomy technique (39 %). In adults, it was the opposite situation, that is, 75 % after exploratory laparotomy and 13 % without laparotomy.
8. Future research in these patients not only will allow a better understanding of the pathogenesis of abdominal CSF pseudocyst but also will determine which procedure should be the best neurosurgical treatment in children and adults with these conditions.

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## **Título: Cirugía de Epilepsia en Bolivia**

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## **Resumen.**

Procedimientos microneuroquirúrgicos funcionales tienen como prioridad la modificación de la función de agrupamientos neurales o circuitos neurales específicos, y no la remoción de las lesiones que colocan en riesgo la viabilidad del sistema nervioso o la vida del paciente. Así de esta manera, está íntimamente asociado al abordaje de la neurocirugía funcional encontrándose el concepto de intratabilidad clínica o farmacológica de la Epilepsia. Este es el aspecto más crucial en la selección de pacientes epilépticos para tratamiento quirúrgico, sin embargo, no es estrictamente excluyente. Nuestro trabajo muestra los criterios, técnicas y resultados en cirugía de epilepsia en 77 pacientes seleccionados por equipo multidisciplinario beneficiados de este procedimiento microneuroquirúrgico.

## **Summary.**

Functional microneurosurgical procedures have as a priority the modification of the function of neural groupings or specific neural circuits, and not the removal of lesions that put the viability of the nervous system or the life of the patient at risk. Thus, it is closely associated with the approach of functional neurosurgery, finding the concept of clinical or pharmacological intractability of Epilepsy. This is the most crucial aspect in the selection of epileptic patients for surgical treatment, however, it is not strictly exclusive. Our work shows the criteria, techniques and results in epilepsy surgery in 77 patients selected by a multidisciplinary team who benefited from this microneurosurgical procedure.

## **Introducción.**

Es cierto que la entidad “epilepsia”, tomada genéricamente, trae consigo una perspectiva pronóstica muy favorable, apenas un quinto y un sexto de los pacientes con epilepsia tienen dificultad en controlar sus crisis con las Drogas Anti Epilépticas (DAE). Estos pacientes con crisis así denominadas “refractarias” las DAE pueden encajarse en uno de los 3 escenarios clínicos que merecen la consideración quirúrgica. El primer escenario es representado por aquellos pacientes que están recibiendo dosis y combinaciones adecuadas de DAE, no refieren efectos colaterales significativos, mas persisten con crisis que interfieren de forma significativa en el funcionamiento social. El segundo se compone de pacientes que también persisten en crisis que, además, sufren de forma importante con la carga medicamentosa prescrita. Por tanto, el tercer escenario incluye aquellos pacientes que consiguen un control satisfactorio de las crisis, en términos de frecuencia e intensidad, pero solamente con dosis y combinaciones de las DAE que virtualmente los incapacitan para una vida normal. Todos estos pacientes pueden ser considerados como incapacitados por el “problema de la epilepsia” –crisis o carga de medicamentosa. A esta discusión se debe aumentar también la incapacitante situación representada por aquellos pacientes, principalmente en los países en desarrollo, que obtienen un control adecuado de las crisis solamente con el empleo de alguna (o algunas) de las nuevas DAE, las cuales representan un costo muy elevado. Varios de estos pacientes y sus familias sufren con este tipo peculiar de incapacitación social, representado por el problema que es de ingerir medicamentos de alto costo, para uso crónico, por tiempo indeterminado, en una base de salario familiar ya bastante comprometido.

En nuestra institución hace siete años y gracias al apoyo de autoridades, jefes de servicio, médicos de planta, residentes, enfermeras, trabajadoras sociales y personal en general la cirugía de Epilepsia es una realidad, constituye un paso importante que en nuestro medio seamos pioneros de este tipo de procedimientos de alta complejidad, que se vienen realizando de forma rutinaria en el Hospital Materno Infantil. Contamos actualmente con un equipo multidisciplinario de Neurocirugía Funcional que en interacción con las diferentes especialidades se siguen conductas quirúrgicas en pacientes Epilépticos, todos ellos evaluados y estudiados ampliamente.



## **Objetivos.**

Demostrar los beneficios de la cirugía para la epilepsia, mediante las diferentes técnicas microquirúrgicas de acuerdo al tipo de Epilepsia sintomática, revisar los criterios de selección de pacientes candidatos a tratamiento quirúrgico de epilepsia, conforme realización de estudios pre operatorios y analizar los resultados clínicos obtenidos mediante la escala de Eagle de pacientes operados seguidos en consulta externa.

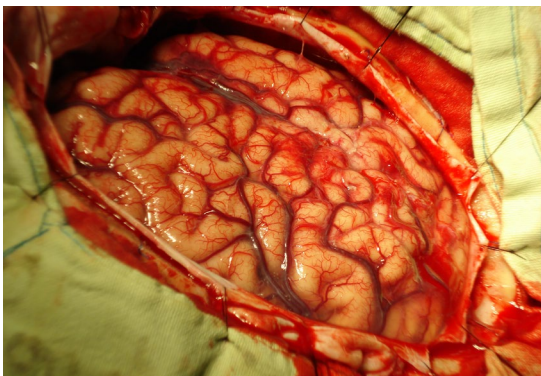
## **Casuística y método.**

77 pacientes operados 48 de sexo masculino, 29 de sexo femenino edades entre 8 y 45 años, todos ellos seleccionados por criterios clínicos de refractariedad, estudios neurofisiológicos como Electroencefalograma (EEG), Video electroencefalograma (Video EEG), Estudio imagenológico como Resonancia Nuclear magnética (RNM) de encéfalo con secuencias T2 y FLAIR coronal. Todos los pacientes contaban valoración Neuropsicologica pre y pos operatorias. Para la valoración de los resultados se evaluó el estado clínico de pos operatorio mediante la escala de Eagle con seguimiento no menor a 12 meses.

Fueron realizadas las siguientes cirugías en el periodo de Abril 2007 a Diciembre 2016:

Hemisferotomia Funcional Derecha

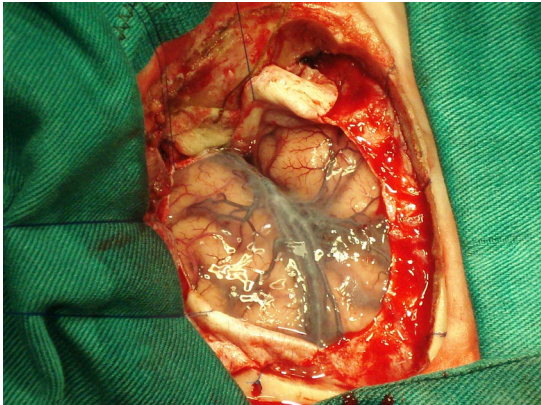
(fig 1)



*Fig 1 Exposición de hemisferio para Hemisferotomia funcional derecha*

Neocorticectomia Amigdalohipocampectomia Derecha 31

Neocorticectomia Amigdalohipocampectomia Izquierda 12 (fig.2)



(fig. 2) Exposicion de lobulo temporal para Amigdalohipocampectomia

Lesionectomias Derechas 15

Lesionectomias Izquierdas 6 (fig3)



fig. 3 Posición para lesionectomia guiada por estereotaxia.

Reseccion de foco Epileptogenico Derecho 7

Reseccion de foco epileptogenico Izquierdo 4

Los resultados fueron analizados en pacientes operados del hemisferio derecho y del hemisferio izquierdo siendo que todos ellos tenian dominancia cerebral Izquierda

## **Resultados.**

De acuerdo a la escala funcional modificada de Engel cuyos parámetros son los siguientes:

Grupo I = libre de crisis epilépticas

Grupo II = rara ocurrencia de crisis

Grupo III = meritoria mejoría

Grupo IV = Sin mejoría

Del universo de pacientes todos mejoraron, no hubo óbitos ni secuelas limitantes en el pos operatorio, 49 pacientes (63.3%) se encuentran en el Grupo I, 25 pacientes (32,4%) en el grupo II y 3 pacientes en el grupo III.

## **Discusión y conclusión.**

Según nuestro estudio, los resultados fueron buenos con control de las crisis parciales y generalizadas con control total con uso de un solo medicamento anticonvulsivo y otro porcentaje libre de crisis y sin uso de medicación.

Actualmente son menos las contraindicaciones para la cirugía de epilepsia. El paciente epiléptico en Bolivia a lo largo de muchos años no a tenido posibilidad de optar por otros tratamientos que no sean los medicamentos anticonvulsivos, pero en la actualidad existen procedimientos neuroquirúrgicos capaces de brindar una opción muy válida en el tratamiento de la Epilepsia.

Los trabajos de centros donde se practican cirugías de Epilepsia tienen además de resultados similares en relación proporcional al volumen de pacientes atendidos, diagnosticados e incluidos en el protocolo de cirugía de Epilepsia, son aun varios los elementos que se deben tomar en cuenta para estructurar un centro para realizar este tipo de cirugía con patrones y cánones internacionales, sin embargo en nuestros primeros casos, la expectativa de los pacientes ha sido llenada positivamente, pues todos ellos al tener menos crisis o ninguna llegaron a realizarse no solamente del punto

de vista familiar, profesional, sino personal ya que 7 de ellos contrajeron matrimonio y tener hijos que es la mayor satisfacción para el equipo que nos acompañó en este tiempo.

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**Título:** Manejo del hematoma subdural agudo con la técnica enrejado de duramadre en la Caja Nacional de Salud del 2005 al 2019

**Title:** Management of acute subdural hematoma with the dura grid technique in the Caja Nacional de Salud from 2005 to 2019

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## **Resumen.**

**Introducción.** Los traumatismos craneoencefálicos (TCE) son lesiones físicas producidas sobre el tejido cerebral cuando una fuerza cinética externa intercambia energía con este. Los hematomas subdurales agudos a menudo requieren intervención quirúrgica (craniectomía descompresiva) con apertura amplia de la duramadre, por lo que es importante desarrollar nuevas técnicas que puedan reducir complicaciones inmediatas o tardías de este procedimiento.

**Objetivos.** Identificar los beneficios de la técnica en la evolución de los pacientes con HSDa.

**Materiales y métodos.** Es un estudio descriptivo en el periodo comprendido junio 2005 a septiembre del 2019 en el Hospital de tercer nivel perteneciente a la Caja Nacional de Salud, Hospital Materno Infantil regional La Paz en el Servicio de Neurocirugía, que incluye a un total de 77 pacientes.

**Resultados.** Mortalidad de 9% (n=7), con un tiempo de permanencia en la Unidad de Terapia Intensiva de  $X= 23$  (2 a 44 días), con un promedio de internación de  $X= 38$  (7 a 69 días). La escala de repercusión de Glasgow (GOS) Grado 4: 51 % (n= 39), grado 3: 26% (n=20), grado 2: 14% (n=11) y grado 1: 9% (n=7).

**Discusión.** Esta técnica de abertura dural puede permitir la remoción de un hematoma subdural sin lesiones de la corteza del cerebro edematizado contra los bordes de la duramadre abierta o de la misma craneotomía.

**Conclusión.** Esta técnica ha demostrado un mejor pronóstico funcional con grados de secuelas, diferencia en los días de estancia hospitalaria, y en la Unidad de Terapia Intensiva en nuestra población.

## **Summary.**

**Introduction.** Head injuries (TBI) are physical injuries produced on brain tissue when an external kinetic force exchanges energy with it. Acute subdural hematomas often require surgical intervention (decompressive craniectomy) with wide opening of the dura, so it is important to develop new techniques that can reduce immediate or late complications of this procedure.

**Objectives.** To identify the benefits of the technique in the evolution of patients with HSDa.

**Materials and methods.** It is a descriptive study in the period from June 2005 to September 2019 in the Third Level Hospital belonging to the Caja Nacional de Salud, Hospital Materno Infantil La Paz Regional, in the Neurosurgery Service, which includes a total of 77 patients.

**Results.** Mortality of 9% (n=7), with a length of stay in the Intensive Care Unit of  $X= 23$  (2 to 44 days), with an average hospital stay of  $X= 38$  (7 to 69 days). Glasgow Impact Scale (GOS) Grade 4: 51% (n=39), Grade 3: 26% (n=20), Grade 2: 14% (n=11), and Grade 1: 9% (n= 7).

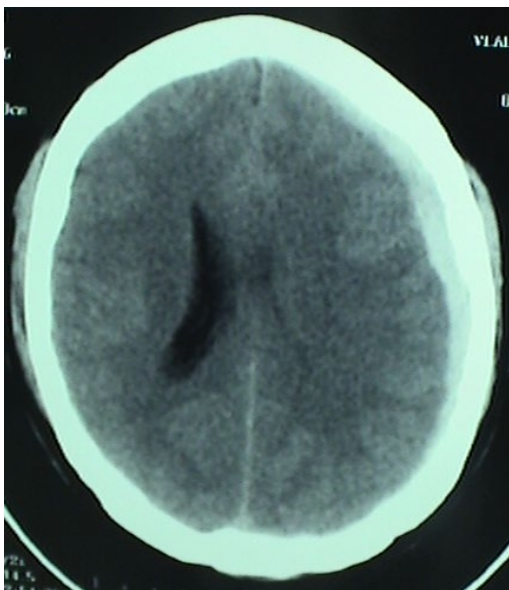
**Discussion.** This dural opening technique may allow removal of a subdural hematoma without injury to the swollen cerebral cortex against the edges of the open dura mater or the craniotomy itself.

**Conclusion.** This technique has shown a better functional prognosis with degrees of sequelae, difference in days of hospital stay, and in the Intensive Care Unit in our population.



## Introducción.

Los traumatismos craneoencefálicos (TCE) son lesiones físicas producidas sobre el tejido cerebral cuando una fuerza cinética externa intercambia energía con este, alterando de forma temporal o permanente la función cerebral, es un problema de salud pública en todo el mundo con un enorme impacto personal y social. Son la causa más frecuente de muerte y discapacidad entre niños y adultos jóvenes. De todos los pacientes con traumatismo aproximadamente el 10% experimenta una lesión cerebral mortal, el 5 al 10 % graves déficits neurológicos permanentes (1). Los traumatismos craneales cerrados causan más daño encefálico que los penetrantes, la aceleración y desaceleración que puede desplazar el cerebro de una forma repentina dentro del cráneo y ocasionar un impacto brusco contra la calota inflexible y los pliegues sólidos y afilados que puede causar contusiones, hemorragias, etc. Una de las emergencias neuroquirúrgicas más serias son los hematomas subdurales agudos, que a menudo requieren intervención quirúrgica, después del tratamiento de un traumatismo craneoencefálico severo, la tasa de mortalidad de un hematoma subdural agudo va de 60 a 90% que se atribuye a las características propias del hematoma, presencia de lesión adicional al parénquima cerebral y lesiones



*HSD agudo izquierdo, paciente masculino de Emergencias del HO1*

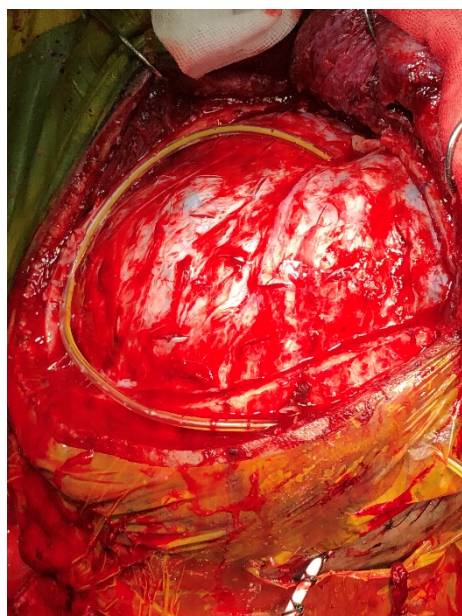
secundarias (2).

Los hematomas subdurales agudos (HSDa) son colecciones hemáticas localizadas entre la superficie interna de la duramadre y la aracnoides. Es una de las principales causas de muerte e incapacitantes, más frecuentes que los epidurales. Por desgarro de las venas emisarias corticales cuando cruzan el espacio subdural para drenar en un seno venoso dural. Más del 95% de los HSDa son supratentoriales, la localización más frecuente es en la convexidad de la zona

frontotemporoparietal, puede ser bilateral, en su mayoría de origen traumático,

un 72% relacionado con caídas y agresiones, y un 24% debido a accidentes de tráfico. Los accidentes de tráfico, accidentes laborales, violencia doméstica, heridas por arma blanca y/o de fuego son las causas más frecuentes de esta noxa(3). En Bolivia, las estimaciones realizadas por el Ministerio de Salud y la Policía Nacional (2004), permiten inferir que ocurren 55 accidentes por día y 2,5 por hora; con las implicaciones consiguientes de gastos, muertes, heridos y seguramente luego de algún tiempo personas discapacitadas (4). Lastimosamente no contamos con estadísticas en nuestro medio sobre la patología, se accede a un estudio realizado en la ciudad de La Paz en población pediátrica gestión 2011 a 2012 donde se estudiaron 479 casos entre las edades de 0 días a 16 años. El grupo con mayor incidencia de TCE fue el sexo masculino con 59,7% , los preescolares de 2 a 5 años fueron los más afectados con 34,2%, predominando el TCE leve (55,7%), el mecanismo de lesión fue producto de caídas por debajo del nivel del plano de sustentación con un 51,6%, seguida por hechos de tránsito en calidad de peatón con un 15,7%, el tipo de lesión más frecuente fue “Lesión no especificada”, es decir con diagnostico impreciso (37%), seguida por edema cerebral (25,9%) (5). Un resultado que permite extrapolar a la población adulta de manera parcial.

La craniectomía descompresiva es el método de remover una porción del cráneo con apertura amplia de la duramadre, es la técnica más utilizada para el drenaje de coágulos en estos pacientes para reducir la PIC y reducir el subsecuente deterioro, sin embargo pese a una técnica bien ejecutada el altamente probable presencia de herniación a través de la craniectomía, la que es seguida por una descompresión rápida debido a lesión de venas cerebrales corticales con el borde del hueso y la abertura dural (6), por lo que es importante desarrollar y emplear nuevas técnicas que puedan reducir complicaciones inmediatas o tardías de este procedimiento. En este sentido se ha descrito en la



*Craniectomía descompresiva con enrejado de dura*

literatura una craniectomía unilateral que se borda como una convencional craniectomía con una variante muy importante que no compromete la abertura completa de la duramadre para el drenaje de coágulos en el HSDa, sino abertura parcial de la misma a través de pequeñas incisión recta en un centro o sentido específico, realizándose una serie de incisiones de 2cm de largo, esto permite expansión de la duramadre en una dirección controlada, manteniendo su integridad y reduciendo la incidencia de herniación (7).

### **Objetivos.**

- Identificar los beneficios de la técnica en la evolución de los pacientes con HSDA.
- Disminuir el tiempo quirúrgico.
- Disminuir la morbimortalidad.
- Evitar grados profundos de secuelas.

### **Material y métodos**

#### **Descripción del estudio de población.**

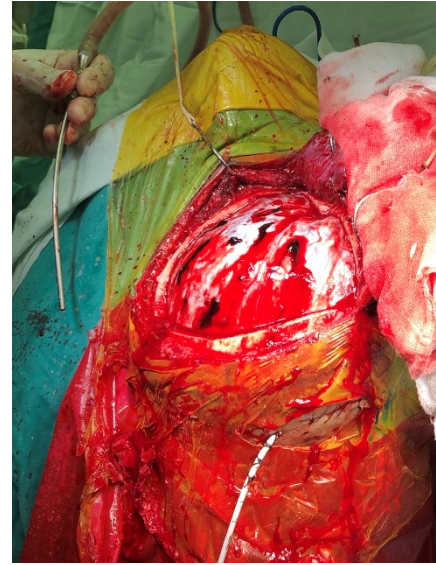
Este es un estudio descriptivo en el periodo comprendido junio 2005 a septiembre del 2019 en el Hospital de tercer nivel perteneciente a la Caja Nacional de Salud, Hospital Materno Infantil regional La Paz en el Servicio de Neurocirugía, que incluye a un total de 77 pacientes. Las características clínicas de los pacientes fueron registradas según género, edad, escala de coma de Glasgow al ingreso, clasificación de Marshal y que fueron operados dentro de las primeras 24 horas posterior al traumatismo. Con seguimiento a 6 meses posteriores al evento quirúrgico, se registra mortalidad, tiempo de permanencia en UTI, tiempo de internación hospitalaria y grado de secuela basaja en "Glasgow outcome scale" (GOS) Todos los pacientes ingresaron a craniectomía descompresiva con abertura de duramadre tipo enrejado, secundario a HSDa.

De los 77 pacientes en las edades comprendidas entre 19 a 57 años, ingresando a quirófano todos aquellos clasificados en la escala de Glasgow como TCS severo (>8 GSC) y rango 3-4 en la escala de Marshal tomográfica.

### Técnica operatoria.

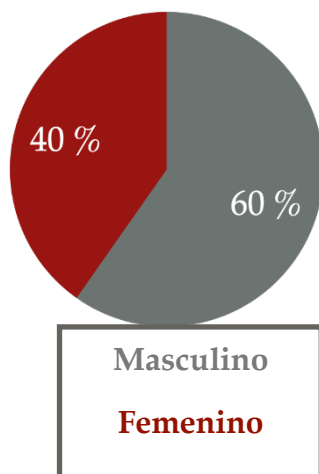
La craniectomía descompresiva consiste en la remoción de una porción del cráneo a través de un flaps de 12x15cm mínimo que estaría asociado con baja mortalidad (26% vs 35%) comparado con flaps más pequeños, siendo la norma lo más amplia posible y descomprimiendo siempre base de cráneo medio (temporal).

Se realizo durotomía en enrejado sobre la localización del HSDa, en líneas verticales y horizontales para distribuir la fuerza de tensión de la dura, a una distancia de 2cm entre incisión e incisión, con posterior lavado y drenaje de coágulos a través de estos.



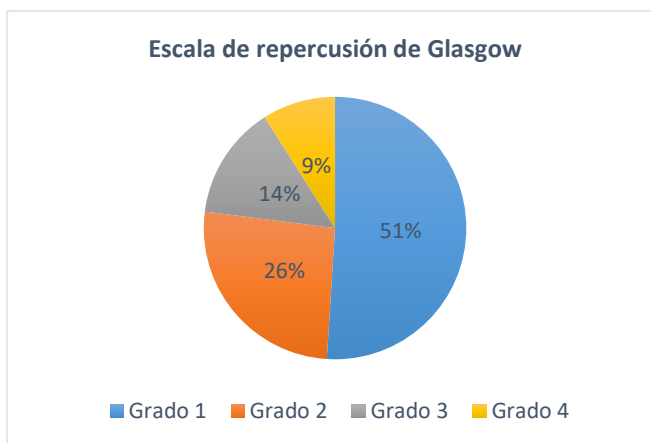
*Drenaje de coágulos a través de enrejado dural*

### Resultados.



Nuestra población compuesta de 77 paciente que ingresaron a quirófano durante las primeras 24 horas posterior a la lesión, 31 (40,2%) fueron femeninas y 46 (59,8%) son masculinos, todos con criterios de TCE grave según escala de coma de Glasgow, en edades comprendidas de 19 a 57 años, con la causa más común del traumatismo craneoencefálico severo secundario a accidentes de tránsito.

Obteniéndose una mortalidad dentro de los primeros tres meses de 9% (n=7), con un tiempo de permanencia en la Unidad de Terapia Intensiva de X= 23 (2 a 44 días), con un promedio de internación de X= 38 (7 a 69 días). La escala de repercusión de Glasgow (GOS) se subdivide en: Grado 1: Muerte; Grado 2: estado vegetativo que se entiende como la incapacidad de interactuar con el medio que lo rodea; Grado 3: discapacidad severa, que es la incapacidad de vivir independientemente, obedece ordenes; Grado 4: Discapacidad moderada, es aquel paciente capaz de vivir independientemente, incapaz de volver al trabajo o escuela, y finalmente Grado 5: Buena recuperación que es capaz de reincorporarse a su vida normal, por lo que nuestros resultados a los 6 meses reporta: Grado 4: 51 % (n= 39), grado 3: 26% (n=20), grado 2: 14% (n=11) y grado 1: 9% (n=7).



## **Discusión.**

El empleo de una nueva técnica para el drenaje de Hematomas subdurales agudos, que da sus primeros pasos desde 1995 de la mano de Guilburd et al, que presenta sus resultados en el Décimo congreso Europeo de Neurocirugía en Berlín (8), donde describe la técnica de fenestración dural , seguido de diferentes autores que con técnicas similares como la “lattice duroplasty” de Mitchell et al. asociada a craniectomía descompresiva para la remoción de hematomas subdurales (9). Hasta nuestro presente, donde reciclando una técnica ya practicada en otros países, podemos empezar a aplicar en nuestro medio, con la finalidad de mejorar los resultados postquirúrgicos de pacientes con traumatismo craneoencefálico severo, en donde aun es controversial el beneficio quirúrgico de pacientes al límite de la vida o la función recuperable (10). Por lo que vale la pena estudiar los resultados obtenidos por los predecesores con este tipo de casos y comparar con nuestros resultados obtenidos con la variación de

la abertura de la duramadre, que se traduce en menor tiempo quirúrgico, menor lesión a la duramadre, mayores probabilidades de evitar una herniación transcalvaria y menor riesgo de fistula de liquido cefalorraquídeo, dichos beneficios que se traducirían en disminución de las secuelas secundarias al drenaje del hematoma subdural agudo.

Esta técnica de abertura dural parece original, elegante y técnicamente fácil de hacer, puede permitir la remoción de un hematoma subdural sin lesiones de la corteza del cerebro edematizado contra los bordes de la duramadre abierta o de la misma craneotomía (11), por lo que crear algoritmos para pacientes susceptibles a esta técnica, ayudaría a escoger pacientes idóneos, pues una de las limitaciones más importantes de esta técnica es que pese al adecuado control de PCI postquirúrgica, puede la abertura no llegar a ser suficiente para controlar una hipertensión intracraneal severa, pese a la extensión de duramadre que podamos lograr con el enrejado, por lo que se requiere más estudios que reporten sus resultados con una población más amplia.

Nuestros resultados son equiparables a otros estudios reportados como serie de casos, en donde se ve una clara diferencia en la disminución de tiempo operatorio, estancia en UTI y estancia hospitalaria, a la que se reporta en la apertura clásica completa de duramadre en sus diferentes variaciones (en C, estrellada, etc).

## **Conclusión.**

La técnica del enrejado de duramadre ha demostrado en el presenta trabajo disminución en la mortalidad, un mejor pronóstico funcional con grados de secuelas, diferencia en los días de estancia hospitalaria, así como reducción en la utilización de camas en la Unidad de Terapia Intensiva, por lo que consideramos que es una herramienta en el arsenal técnico quirúrgico a considerar, una vez establecido los criterios de selección de paciente susceptible a este procedimiento.

## **Palabras claves.**

Hematoma subdural agudo, fenestración dural, técnica Lattice, craniectomía descompresiva.

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