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Management of Advanced Child Hydrocephalus From 2008 to 2018 in Parakou, Benin

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ABSTRACT

Introduction: Hydrocephalus is a common neurosurgical condition in children. The objective of this study was to report our experience and the outcome of the management of advanced child hydrocephalus in Benin.

Patients & Methods: This was a retrospective and transversal study performed at the teaching hospital of Borgou in Benin. From 2018 to 2018, children admitted for hydrocephalus were registered. The cranial circumference, the condition of the cerebral parenchyma and ventricular dilation, and the psychomotor state were the major criteria used to indicate an advanced hydrocephalus. Clinical condition, postoperative evolution and child outcomes were the variables studied. The follow-up was 1 to 10 years.

Results: During the study period, 153 (52.6%) among 291 cases of hydrocephalus were classified as advanced hydrocephalus. The mean age was 9.37 ± 16.79 months. The sex ratio of 0.9. A ventriculoperitoneal shunt was performed for 116 (75.8%) children. Post-operative complications were divided into infections, valve migration and dysfunction in 13.4%, 4.3% and 12% of cases respectively. Post-operative mortality was 16.4%. During follow-up, 28 (28.9%) patients were lost to follow-up and 32 (33%) died. No motor acquisition was observed in 26 children. Four children have developed complete autonomy. One of these children has entered school.

Conclusion: The long-term survival and psychomotor development of children with advanced hydrocephalus is almost non-existent. To establish criteria for operability and determine the short and long-term prognosis, it is important that so-called "advanced" hydrocephalus be defined on the basis of precise clinical and radiological criteria.

Keywords: Advanced hydrocephalus, Cranial circumference, Psychomotor development, Ventriculoperitoneal shunt, Postoperative mortality

INTRODUCTION

Hydrocephalus is a common childhood neurosurgical disease [1-5]. It is one of the most common paediatric affections observed in neurosurgical practice in Benin [6-8]. The principal mode of discovery is a macrocephalus. The consultation is very late and the resulting excessive macrocephalus gives a particular socio-cultural connotation to these children who become the object of all negative interpretations [6]. These socio-cultural considerations, combined with the advanced stage of diagnosis and therapeutic vicissitudes, finaly make all hope of a successful outcome illusory [9-11]. The management of hydrocephalus thus becomes, as soon as the children are admitted, a delicate preoccupation about therapeutic choices in the face of parents' expectations [11-13]. Despite the population campaigns conducted since 2008, the proportion of advanced hydrocephalus cases admitted to our hospital is still considerable. Is it possible to provide a surgical response to an affection whose socio-cultural perception condemns the patients concerned in advance? Can surgical management improve the future of these children and facilitate their social integration? The objective of this study was to report our experience of the management of advanced Child hydrocephalus and the future of these children over a

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ten-year period from 2008 to 2018 in the neurosurgery department of Parakou Teaching hospital in Benin.

PATIENTS & METHODS

It was a retrospective, transversal and descriptive study. It was performed at the Neurosurgery Unit of the Departmental Teaching Hospital of Borgou and Alibori (CHUD-B/A) in Benin. This hospital is the reference Centre for the northern region of Benin. There is no scanner device in the hospital. However, this examination can be performed in another hospital located 6 km from the reference centre. The neurosurgery unit is housed in the general surgery department. This unit does not have an endoscopy column. The ventriculoperitoneal shunt (VPS) insertion is therefore the only option for surgical management of hydrocephalus. Shunts are ordered in advance as they are not available in the hospital's pharmaceutical stocks. Patients are accountable for all costs related to their care. There is no service to manage patients who do not have resources.

All cases of hydrocephalus treated in patients aged one month to fifteen years admitted to the unit from 2008 to 2018 have been identified. Cases of advanced hydrocephalus managed during this period were considered. The diagnosis of "advanced" hydrocephalus has been based on clinical and radiological records. These were chronic clinical intracranial hypertension, head circumference, impaired psychomotor function, the importance of ventricular dilation associated with a cerebral cortex reduced to a cortical mantle or almost disappeared. Clinical status of children, postoperative outcomes, complications, term survival and outcomes of children were the variables studied. The follow-up period was 1 to 10 years. The immediate postoperative period was considered to be the period from the first to the thirtieth day (D1- D30) postoperatively. Cases of hydranencephaly, tumoural hydrocephalus have not been included. Cases of advanced congenital hydrocephalus of incidental discovery at birth have been excluded.

RESULTS

During the study period, 291 cases of hydrocephalus were identified among the childhood neurosurgical pathologies. Among these cases, 153 (52.6%) were considered to be advanced hydrocephalus. These children were referred by families, charities or referred from a health centre in respectively 22 (14.4%), 95 (62.1%) and 36 (23.5%) cases.
The mean age of the children was 9.37 ± 16.79 months with a extreme of 3 months and 11 years. These children were divided into 73(47.7%) boys and 80(52.3%) girls with a sex ratio of 0.9. A medical history of a recent infection or hospitalization was found in 44 (28.7%) children. Operative spinal dysraphy was found in 15 (9.8%) children. The mean head circumference observed in these children was 61±8.75 cm with ranges of 49cm and 76cm. **Figure 1** shows the morphological condition of these children at admission. The main signs of examination observed in these children were reported in **Table 1.**

Figure 1. Macroscopic aspect, of children on admission: **a)** Voluminous macrocephalus with frontal prominence, a "sunset" looks and abnormal visibility of the peripheral veins **b)** straight infant with outstretched arms and closed fists with decerebral rigidity witnessing chronic intracranial hypertension.

The diagnosis was performed using head ultrasound (transfontanel) in 71 (46.4%) cases and brain CT-scan in 82 (53.6%) cases. From 2008 to 2010, it was exclusively ultrasound in 50 (32.7%) cases, from 2010 to 2013 an ultrasound and a brain CT-scan in 21 (13.7%) cases and 35 (22.9%) cases respectively, and finally from 2014 to 2018 the diagnosis was exclusively made using brain CT-scan in 47 (30.7%) cases. **Figure 2** shows the brain CT-scan

Table 1. Distribution of signs or symptoms observed in children admitted for advanced hydrocephalus.

	Effectif	Percentage (%)
Increase in head circumference	153	100
Frontal prominence	153	100
Craniofacial dysmorphism	153	100
Enlarged anterior fontanel	150	98
Ocular abnormalities	147	96.1
Disjunction of the skull sutures	148	96.7
Psychomoteur retardation	104	68
Rigidity / axial hypotonia	100	65.4
Turgescence of the epicranial veins	96	62.7
Undernutrition	91	59.5
Bedridden child	62	40.5

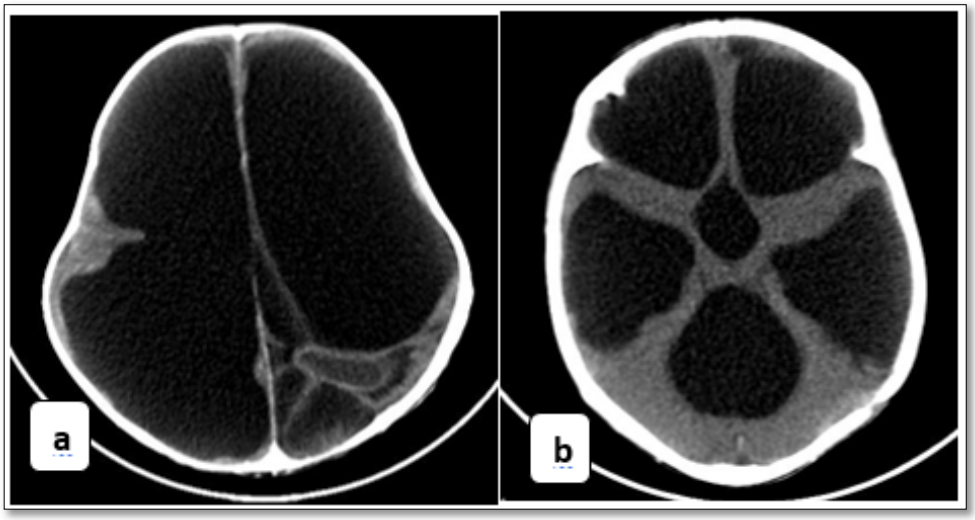

Figure 2. CT scan of ventricular dilation observed in children considered to have advanced hydrocephalus. **a)** To be noticed is the importance of ventricular dilation and the near disappearance of cerebral parenchyma **b)** Significant dilation of the 4th ventricle and temporal horns of the lateral ventricles.

appearance of the cerebral parenchyma and ventricular dilation observed at admission.

Among the 153 children, 116 (75.8%) underwent ventriculoperitoneal shunt (VPS) insertion. The shunt used were all non-adjustable and medium pressure. The motives for the 37 patients (24.2%) who did not undergo surgery were reported in **Table 2**. Concerning the patients who underwent surgery, the immediate postoperative course was indicated in **Table 3**. The post-operative complications (32.8%) observed were divided into infections, shunt migration, shunt disconnection and cranial depression in respectively 19 (16.4%), 5 (4.3%), 4 (3.4%), 10 (8.6%) cases. A shunt revision was successfully performed in all 9 cases of malfunction. Of the cases of infection observed during hospitalization, 15 (78.9%) were successfully treated and 4 patients died. Post-operative mortality was 16.4% (n=19 cases). A ten-year follow-up was carried out on the 97 children discharged from hospital. Regarding the long-term future of children, over the cumulative period of 10 years, 28 (28.9%) patients were lost to follow-up and 32 (33%) died. These deaths occurred over a period of 6 to 36 months after

Table 2. Distribution of the different motives for not performing surgery on 37 patients for whom an operative indication has been given.

	Number	Percentage (%)
Operated patients	116	75.8
Non-operated patients	37	24,2
Motives		
Anesthesia	15	9.8
Family decline	7	4.6
Financial default	6	3.9
Death before surgery	9	5.9

Table 3. Distribution of postoperative results of the 116 children who underwent VPS.

	Number	%
Good evolution	7	6
Complications	38	32.7
No improvement	52	44.8
Death	19	16.4

discharge from hospital. The overall post-operative mortality rate thus increased from 16.4% to 44% over a period of 3 years. No motor acquisition was observed in 26 (26.8%) children who remained bedridden and completely dependent. Among these children, two (2) cases of annal shunt migration were observed. A valve revision was performed for these two children. An increase in cranial dysmorphism was observed in 15 (15.5%) children. **Figure 3** shows the short and long-term evolution among children undergoing surgery. A partial autonomy, partial language acquisition and the possibility of communication with their family and friends were observed in 7 (7.2%) children. Complete autonomy was observed in 4 (4.1%) children. Among these autonomous children, one child goes to school in a charity center. The recapitulative evolution of the management of these cases of advanced hydrocephalus has been reported in **Table 4.**

DISCUSSION

Hydrocephalus is a common condition in children and has been the subject of scientific research by several authors [9,13-16]. In Benin, the child's hydrocephalus is the object of a particular perception. It is located in a heavy socio-cultural context of suspicion where the mother of a child suffering from hydrocephalus is suspected of witchcraft or adultery. This condition is sometimes perceived as a malediction or reincarnation of a "God". The social consequence of this perception is the banishment, exclusion or abandonment of these children. The management of these children is therefore a way of averting the bad fate for the survival of the community. It is therefore a question of providing a surgical and modern response to "medieval" considerations.

Several studies have been performed concerning the etiologies and management of this condition [7,12]. The findings resulting from medical and surgical management have made it necessary to reflect and define a therapeutic strategy, although difficult to formalize.

Often deplored by several authors [11,13,15,16], the long delays in consultations, the brain condition of children at admission, their general presentation and finally the conditions of diagnosis and management justified the results reported by these authors. This late consultation and delay in diagnosis has the consequence of a "monstrous" hydrocephalus that the authors of this study have reclassified as "advanced hydrocephalus". Over a period of 10 years, the long-term evolution of this category of children in hospital with socio-cultural conditions and considerations in Benin was assessed.

The problem of hydrocephalus and in particularly, the so-called "advanced" hydrocephalus is complex. Indeed, defining advanced hydrocephalus is a nosological problem.

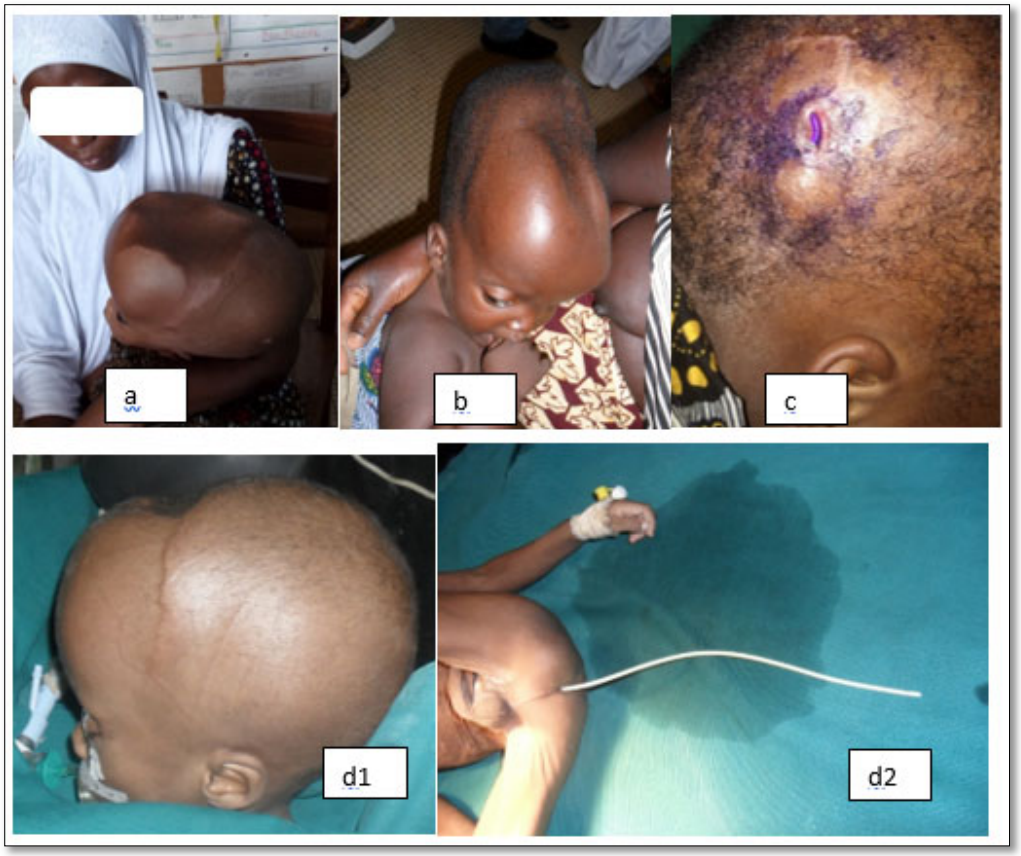

Figure 3. The short- and long-term evolution among operated children. **a**) Significant postoperative hyper drainage with a cranial defect indicating the importance of the disjunction of the skull sutures before surgery. **b)** Post-operative scaphocephaly (These are the children seen in **Figure 1** other form of over drainage (cranial depression). **c)** cranial skin infection with externalization of the catheter. **d1)** Cranial deformation and overlapping skull bones associated with annal migration **(d2)** in a same child.

Table 4. Recapitulative data on 153 cases of advanced hydrocephalus followed over a 10-year period from 2008 to 2018.

	Effectifs	**%**
Patients non opérés	37	24.2
Operated patients	116	75.8
Deceased patients*	51	44
Lost-Patients*	28	24.1
Partial motor acquisition*	7	6.0
Complete autonomy*	3	2.6
Child attending school*	1	0.9
No improvement*	26	22.4

The literature review reports anatomical or etiopathogenic definitions of hydrocephalus [2,9,17,18]. Even with classification efforts, the issue was not seen in an evolutionary light [1,17]. It is mentioned in the series published in Africa [9,12,13,15,16,18] and sometimes in Europe a late consultation, a delay in diagnosis, children

received with a significant increase in the head circumference or in a deteriorated condition in general. The consequence is an unpredictable evolution or a reserved prognosis upon admission.

To materialize the concept of a so-called "advanced" hydrocephalus, we have chosen major criteria. Doing so is a prognostic element and can help to define more accurate predictive factors for good or poor prognosis in future studies [16,19,20]. In Africa, and particularly in Benin, child hydrocephalus suffers from a harmful reputation based on ancestral socio-cultural considerations [6,11]. The lack of knowledge of this condition and its mode of revelation through a craniofacial transformation has made late consultations and therapeutic mistakes inevitable. This would explain the late delays in consultations reported in several studies [9,10,16,18]. Although the lack of financial resources is very often the reason given, it cannot be held solely responsible for late consultations as observed in African studies. This economic reason was the alibi that seems to obscure the socio-cultural considerations and impact of traditional hydrocephalus therapy in Benin. In a study carried out in 2011 in Parakou [7], it was already observed that among a population of 57 children admitted for hydrocephalus, 35% of families had linked their delay in consultation to socio-cultural considerations and 15.8% had first consulted a traditional therapist. Certainly 39% of the families had no monthly income. At this advanced stage at the time of the consultation, only 12.3% of children had a relatively preserved cerebral parenchyma compared to 87.7% patients whose cerebral parenchyma was severely laminated or non-existent. Santos et al. [13] in Tanzania reported a late consultation of children reporting children received in poor condition and with an average head circumference of 51±6.3 cm. This head circumference is similar to the 50.7cm reported by Kinasha, Kahamba and Semali [16]. However, these heads circumferences already described as "monstrous" and reported by these authors were lower than the average reported in our study (61±8.75 cm with extremes of 49 cm and 76 cm). These authors did not mention the state of children's cerebral parenchyma and the importance of ventricular dilation. This is due to diagnostic conditions ; researcher [13] noted that 22.4% of children who had surgery had no imaging examination and only 1.6% had brain CT. Gathura, Poenaru, Bransford and Albright [10] over a larger series of 574 patients and a shorter two-year follow-up reported 59% of bad outcomes for all complications combined and noted that the results observed were satisfactory in only 10% of children with a head circumference greater than 60 cm at the time of management. This observation is corroborated by the work of Biluts and Admasu [18] in Ethiopia concerning a series of 114 children operated on with hydrocephalus and in whom VPS was performed. A high rate of complications and deaths was observed in children with a head circumference greater than 50 cm. The difference in complication rates observed was statistically significant between children with a head circumference of less than 50 cm and those with a head circumference greater than 50 cm (p=0.028). In this population of children, 60% of whom had received CT-Scan or brain MRI, the quantified assessment of brain parenchyma would have made a significant contribution to the notion of advanced hydrocephalus. Macrocephaly and head circumference seem to be the main indicators of late consultation without ever mentioning the notion of advanced hydrocephaly.

The challenges of treating hydrocephalus in children are many [20-23]. Several circumstances can compromise the short- and medium-term results of VPS performed for a hydrocephalus. These are both biological and material factors. The prognosis depends not only on the etiology but also and above all on the timing of diagnosis and treatment [24-27]. In our study, all VPSs were performed with non-adjustable medium pressure valves. Over the 10-year period, three fundamental observations can be deduced from our study:

- A very high mortality rate in the short and long term, even if we ignore patients who have been lost to follow-up,
- The high cumulative rate of complications,
- The strong lack of psychomotor improvement among surviving operated children **(Table 4)**.

Although it did not establish a direct link between the children's condition at the time of management and these results, such a finding of failure is a bad message to contradict the negative perception of the child's hydrocephaly disclosed by socio-cultural considerations in Benin. The cost of treating hydrocephalus is significant in Benin as in so many sub-Saharan African countries compared to family incomes and the average cost of living [8,11,28]. However, how to assist these families if they are not received in hospital at first sight. In 2011, the mortality rate among this population of children who received VPS, all types of hydrocephalus combined, was 17%, with 55% of deaths occurring in the first month after surgery and 45% between the 1st and 6th month after surgery [12]. According to studies [29], mortality would increase over time. Thus, without having established a correlation between VPS and mortality, the authors report a survival of 70% in the first year after surgery, which drops to 40% at ten years. According to results reported by Santos et al. [13] in Tanzania, over a 70-day follow-up period, overall mortality was 14.5% in a population with an average age at surgery of 4 months. The authors finish and conclude that the head circumference, the duration of hospitalization and the type of tumor would be the risk factors for death of the operated hydrocephalus. Although in our study tumours were excluded in order to better understand the causes of morbidity and mortality related to hydrocephalus and in

particular advanced hydrocephalus, these factors corroborate our results. The importance of ventricular dilation would be a major risk factor. Advanced hydrocephalus alone can cause bedridden state, undernutrition and therefore longer hospitalization, thus increasing the risk of infection. Some authors [16] seem not to make a link between delays in management and the occurrence of complications. This interpretation can be criticized because it would mean that during the waiting period for a hydrocephalus, no physiological developments occur with regard to the evolution of hydrocephalus. Over a longé study period (40 years), another study [3] estimated the mortality rate of children who have been treated with VPS at 38%. 50% of which occurred in the first two years. This rate excludes cases of brain tumours. Mortality in relation to the equipment was 8%. In optimal condition, Acakpo et al. [30] report a mortality rate of 10%. According to the authors, this decreasing mortality is linked to patient or parent education, staff reinforcement and improved therapeutic conditions. This same observation was made by Warf [21]. These improvements did not include the physiological condition of the patients. Shunt malfunctions and infections are the most feared complications after VPS insertion [31-35]. Frequencies vary according to the studies and the environment. Karmacharya and Kumar [31] in Nepal reports à complication rate of 23% and is dominated by infections; for Piatt and Carlson [22] as well as many authors [19,24,36-38], infectious complications were the most representative and occurred in more than 50% of cases in the post-operative period. Infection control must therefore remain a daily preoccupation. This is possible when one applies oneself to respecting established standards [39].

Regardless of the stage at which hydrocephalus has been treated, it is important to focus in the short and long term on the future of children undergoing surgery. This is more crucial when it comes to advanced hydrocephalus. According to Vinchon and Dhellemmes [29], over a 20-year follow-up period, 20 to 60% of children who underwent surgery were not integrated into adulthood and reportedly have difficulty at school. These difficulties are reported to be more prevalent among young children undergoing surgery. Scientists [3] report a majority of children with autonomy although the functional status of children varies over time. Good physical condition and early diagnosis alone can explain such results. These results are identical to another study [32] which, like Paulsen, Lundar and Lindegaard [3], report adult child autonomy and enrolment in either a normal school or a class for a disabled child. The authors seem to have no anxiety about long-term social and educational outcomes but do not exclude the possibility of complications in adulthood, emphasizing the need for planned follow-up. In communities with low socio-economic conditions, it is not easy to organize follow-up for these children, who in most cases live far from the centres where neurosurgical treatment has been provided. The high wastage rate of these children is thus high, increasing the risk of death in this population prone to complications. This was reported by Biluts and Admasu [18] from 100% to 1-month follow-up to 17% at 12 months. The organization of this follow-up will allow us to have a longer perspective than those reported in the african series, whose brevity constitutes a handicap for an objective and relevant evaluation of these children [40].

CONCLUSION

Long-term survival and psychomotor development in children treated for advanced hydrocephalus are almost non-existent. It is therefore more illusory to eradicate people's harmful beliefs through surgery in the face of this childhood disease. More than ever, prevention, early diagnosis and improvement of patients' socio-economic and therapeutic conditions remain the only real solutions to prevent hydrocephalus and avoid the advanced stage. However, it is important that so-called "advanced" hydrocephalus be clearly defined by specific clinical and radiological criteria. This would help to establish criteria for the operability of this type of hydrocephalus and determine in advance the short- and long-term prognosis.

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