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### Original Article

# An algorithmic approach to the management of unrecognized hydrocephalus in pediatric candidates for intrathecal baclofen pump implantation

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#### **Abstract**

**Background:** Complications of intrathecal baclofen (ITB) pump implantation for treatment of pediatric patients with spasticity and dystonia associated with cerebral palsy remain unacceptably high. To address the concern that some patients may have underlying arrested hydrocephalus, which is difficult to detect clinically because of a low baseline level of neurological function, and may contribute to the high rates of postoperative cerebrospinal fluid leak, wound breakdown, and infection associated with ITB pump implantation, the authors implemented a standardized protocol including mandatory cranial imaging and assessment of intracranial pressure (ICP) by lumbar puncture prior to ITB pump implantation.

**Methods:** A retrospective case series of patients considered for ITB pump implantation between September 2012 and October 2014 at Seattle Children's Hospital is presented. All patients underwent lumbar puncture under general anesthesia prior to ITB pump implantation and, if the opening pressure was greater than 21 cmH<sub>2</sub>O, ITB pump implantation was aborted and alternative management options were presented to the patient's family.

**Results:** Eighteen patients were treated during the study time period. Eight patients (44.4%) who had ICPs in excess of 21 cmH<sub>2</sub>O on initial LP were identified. Eleven patients (61.1%) ultimately underwent ITB pump implantation (9/10 in the "normal ICP" group and 2/8 in the "elevated ICP" group following ventriculoperitoneal shunt placement), without any postoperative complications.

**Conclusions:** Given the potentially high rate of elevated ICP and arrested hydrocephalus, the authors advocate pre-implantation assessment of ICP under controlled conditions and a thoughtful consideration of the neurosurgical management options for patients with elevated ICP.

**Key Words:** Arrested hydrocephalus, cerebral palsy, CSF leak, intrathecal baclofen pump, hydrocephalus, spasticity

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#### **INTRODUCTION**

Intrathecal baclofen (ITB) delivery is one of the most common and effective treatments for spasticity and dystonia associated with cerebral palsy; however, complications of ITB pump implantation continue to be unacceptably high, with some reports citing a combined risk of infection and malfunction exceeding 40%. Despite these challenges, caregivers and practitioners overwhelmingly express satisfaction with baclofen pumps because of their ability to improve ease of care, patient comfort, and quality of life. The onus rests on the neurosurgical community to introduce innovations that minimize the incidence of complications with this increasingly routine procedure.

In many patients with cerebral palsy and spastic quadriparesis, arrested hydrocephalus may concurrently but may be difficult to clinically detect because of many factors including, in some, the limited ability to communicate symptoms associated with hydrocephalus. Albright et al. (2005) demonstrated elevated intracranial pressures (ICP; range: 22–41 cm H<sub>2</sub>O) by lumbar puncture (LP) in 23 of 24 (96%) children with cerebral palsy and ventriculomegaly.[1] In recognition of this prior work and based on our institutional experience with ITB pump implantation, we theorize that unrecognized intracranial hypertension promotes postoperative cerebrospinal fluid (CSF) leaks, increasing the risk of wound breakdown and infection after ITB pump implantation. To detect patients with elevated ICP, a LP was performed on all candidates prior to pump implantation. If opening pressure was found to be elevated (>21 cmH<sub>2</sub>O) in the setting of controlling the end tidal CO2, a clinical pause was introduced to determine whether CSF diversion was necessary or recommended prior to pump implantation. We review our experience and suggestions for management of elevated ICP in the setting of baclofen pump insertion with a view toward reducing the complication of CSF

#### **ILLUSTRATIVE CASE**

Prior to establishing a formal protocol for screening of occult hydrocephalus, our center recorded rates of complications after pump implantations that were in line with published occurrence rates. One such case illustrates the need for ICP interrogation prior to intrathecal pump implantation. A 17-year-old male patient with a history of hypoxic ischemic injury with resultant spastic dystonic quadriplegia and severe developmental delay functioning at the Gross Motor Function Classification System (GMFCS) level V was recommended for baclofen pump implantation for ease-of-care considerations by the Rehabilitation Medicine team. No intraoperative

complications were encountered at the time of baclofen pump implantation. The catheter was placed through a paraspinal transfascial approach. ICP was not measured at the time of pump implantation. In the postoperative period, the patient developed a tense pseudomeningocele at the operative site that ultimately resulted in wound breakdown and a frank CSF leak. Approximately 2 weeks after the initial surgery, a wound revision was performed. After exploring the patient's lumbar pseudomeningocele, we observed CSF egress around the intrathecal catheter. A purse-string suture was placed around the intrathecal catheter that effectively eliminated intraoperative evidence of the CSF leak. Following this surgery, there was temporary improvement in the appearance of the patient's lumbar wound, however, over the following 2 weeks, the patient's lumbar pseudomeningocele recurred and, again, a frank CSF leak was noted. A second, more extensive wound revision was performed at that time in an attempt to definitely address the CSF leak; this procedure consisted of an additional catheter site purse-string suture, a blood patch, and local paraspinal muscle advancement flaps. Unfortunately, the CSF leak persisted, and a decision was made to remove the patient's baclofen pump system in its entirety 5 days later. Upon removal of the lumbar catheter, ICP was measured via lumbar puncture and found to be 38 cmH<sub>2</sub>O (pCO<sub>2</sub> of 32 mmHg). No prior history of hydrocephalus was noted in review of the patient's history. In a delayed fashion, the patient underwent placement of a right frontal ventriculoperitoneal (VP) shunt. Three months after VP shunt placement, the patient again underwent baclofen pump insertion. Intraoperative measurement of ICP at the time of pump insertion was 8 cmH<sub>2</sub>O (pCO<sub>2</sub> of 38 mmHg) in the setting of a working VP shunt. After baclofen pump reinsertion, no postoperative pseudomeningocele and no wound healing concerns developed. The patient's family noted a dramatic improvement in the patient's tone, with appropriate titration of the patient's intrathecal baclofen. Of note, the patient's family did not note any improvement in functional status or cognition after VP shunt placement; the patient remained nonverbal and dependent for eating, dressing, and mobility, as was the case at his preoperative baseline.

#### **MATERIALS AND METHODS**

The case series presented is a retrospective review of patients considered for ITB pump implantation between September 2012 and October 2014 at the Seattle Children's Hospital. Patients were deemed candidates for ITB pump implantation after being seen by both the Rehabilitation Medicine and Neurosurgery teams. Head computed tomography (CT) or brain magnetic resonance imaging (MRI) was performed on all patients preoperatively to evaluate ventricular size

and configuration. Frontal and occipital horn ratios were calculated as a means of estimating ventricular size. [8] At the time of this study, we were offering many patients pump placement based on clinical criteria without a pre-implantation baclofen test dose. Regardless of the family's decision to proceed with a test dose versus moving directly to pump implantation, the procedure began with a LP, performed with the patient placed in a lateral decubitus position under general anesthesia; opening pressure was measured with the patient's end-tidal pCO, kept at approximately 36 mmHg. If the opening pressure on LP was greater than 21 cmH<sub>2</sub>O, we proceeded with planned baclofen test dose administration, but planned ITB pump implantations were aborted and management options were presented to the patient's family [Figure 1]. These options included CSF diversion followed by delayed ITB pump implantation, ophthalmologic examination with repeat LP to confirm the findings of elevated ICP, no additional therapy with close clinical follow-up, or referral for a second opinion.

#### **RESULTS**

Twenty patients were considered for ITB pump implantation during the period of the study. Of these, 18 patients ranging in age from 4 to 20 years met the criteria for inclusion in the study [Table 1]. Two patients considered for ITB pump implantation during the study period were excluded. In one case,

the patient's ITB pump was implanted outside of the study protocol as preoperative cranial imaging was not obtained. The second patient excluded was an 8-year-old female patient with tetraplegia and spasticity secondary to a cervicomedullary pilocytic astrocytoma who had undergone partial resection and radiation therapy. She was known to be at risk for increased ICP preoperatively and underwent placement of a VP shunt and ITB pump implantation during a single anesthetic session.

Among the 18 patients included in the study population, 8 (44.4%) had an ICP in excess of 21 cmH<sub>2</sub>O on initial LP. Nine of the 10 patients with ICP ≤21 cmH<sub>2</sub>O underwent ITB pump, implantation and there were no complications in this group. The family of the one patient (#8) in the "normal ICP" group who did not undergo ITB pump implantation ultimately decided to pursue an alternative procedure (selective dorsal rhizotomy) for spasticity management. One patient (#13) who was found to have an elevated ICP of 23 cmH2O by LP prior to a planned ITB pump insertion subsequently underwent a second LP to confirm the finding and, on repeat procedure, was found to have an ICP of 24 cmH<sub>3</sub>O. There was a mild positive correlation between preoperatively measured frontal and occipital horn ratios on cranial imaging and measured opening pressures on sedated LP  $(R^2 = 0.038)$  [Figure 2].

Two of the 8 patients in the "elevated ICP" group (#1, #16) ultimately underwent ITB pump implantation and, in both cases, VP shunt placement was performed

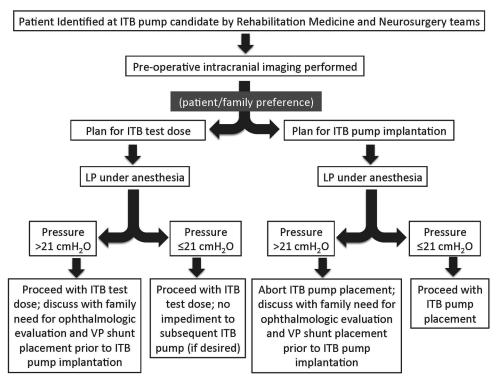


Figure 1: Flow chart demonstrating the clinical protocol employed for patients identified as candidates for intrathecal baclofen pump implantation

Table 1: Overview of clinical data

atient	Age/Sex	Spasticity	Motor	Frontal and		Planned	$ICP(cmH_2O)$	pCO <sub>2</sub> (mmHg)	Outcome
		Etiology/Clinical Details	Function (GMFCS)	Occipital Horn Ratio	Preoperative Imaging Findings	Procedure			
1	6/M	Hypoxic/ ischemic (presumed sudden infant death syndrome)	V	0.49	Diffuse cystic encephalomalacia	TD	36	35	VP shunt placed 10 days later; Baclofen pump implanted 3 months later without complication
2	20/M	Unknown	V	0.38	Mild prominence of extra-axial CSF spaces	TD	14	20	Pump Implanted 1 month later without complication
3	7/M	Lennox-Gastaut Syndrome, cortical blindness, pachygyria, microcephaly	V	0.48	Diffuse pachygyria; microcephaly	TD	22	32	Family deferred baclofen pump implantation (tone improved with tes dose but overall clinical status/ comfort not felt to be improved by family)
4	4/M	Kernicterus	V	0.40	Mild prominence of extra-axial CSF spaces	TD	29	38	Minimal response to test dose (pum not implanted); no papilledema or optic nerve atrophy by fundoscopic examination
5	11/M	Anoxic brain injury secondary to attempted hanging	V	0.38	Unremarkable	PI	18	37	Pump implanted without complication
6	19/M	Leukodystrophy of unknown origin	V	0.44	Absence of subcortical U fiber myelination evident by MRI imaging	TD	21	35	Pump implanted 2 weeks later without complication
7	13/F	Cerebral palsy	V	0.33	Unremarkable, typical ventricular anatomy	PI	15	38	Pump implanted without complication
8	5/M	Cerebral palsy	IV	0.42	Periventricular leukomalacia	TD	14	35	Family deferred implant (ultimately elected to pursue selective dorsal rhizotomy for spasticity management)
9	13/M	Mitochondrial cystopathy and leukodystrophy	V	0.37	Multifocal T2 signal abnormality within white matter of the bilateral centrum semiovale and subcortical frontoparietal lobes	TD	16	35	Pump implanted 2 weeks later without complication

Table 1: Contd...

Patient	Age/Sex	Spasticity Etiology/Clinical	Motor Function	Frontal and Occipital Horn Ratio	Additional Preoperative Imaging Findings	Planned Procedure	ICP(cmH <sub>2</sub> 0)	pCO <sub>2</sub> (mmHg)	Outcome
10	10/5	Details	(GMFCS)			TD	20	27	D. and involved
10	12/F	Cerebral palsy (prematurity w/ intraventricular hemorrhage)	V	0.40	Periventricular leukomalacia	TD	20	37	Pump implanted 3 months later without complication
11	6/F	Cerebral palsy (prematurity)	IV	0.45	Periventricular leukomalacia	PI	10	35	Pump implanted without complication
12	10/M	Cerebral palsy (hypoxic event at birth)	V	0.55	Diffuse cystic encephalomalacia; colpocephaly	PI	>21	32	Pump implantation aborted; consideration for pre-pump VP shunt placement pending
13	6/M	Cerebral palsy (prematurity)	IV	0.51	Periventricular leukomalacia	PI	23	38	Pump implantation aborted; consideration for pre-pump VP shunt placement pending; no papilledema by fundoscopic examination; finding of elevate ICP confirmed by second LP demonstrating an opening pressure of 24 cmH <sub>2</sub> O (pCO <sub>2</sub> 36 mmHg)
14	15/M	Traumatic brain injury~6 months prior requiring right hemicraniectomy	V	N/A*	Right craniectomy defect w/ underlying right frontal and occipital encephalomalacia	PI	10	38	Pump implanted without complication
15	9/M	Cerebral palsy (prematurity w/ intraventricular hemorrhage)	V	0.45	Periventricular leukomalacia	PI	18	36	Pump implanted without complication
16	18/M	Cerebral palsy (near drowning accident as an infant)	V	0.37	Mild prominence of extra-axial CSF spaces	PI	32	36	Pump implantation aborted; CSF leak from lumbar puncture site 10 days post-op; subsequently underwent VP shunt placement; baclofen pump placed 5 months later without complication

Table 1: Contd...

Patient	Age/Sex	Spasticity Etiology/Clinical Details	Motor Function (GMFCS)	Frontal and Occipital Horn Ratio	Preoperative	Planned Procedure	ICP(cmH <sub>2</sub> 0)	pCO <sub>2</sub> (mmHg)	Outcome
17	12/M	Cerebral palsy (prematurity)	IV	0.35	Unremarkable, typical ventricular anatomy	TD	26	36	No papilledema by post-operative ophthalmologic examination; plan for VP shunt placement to be followed by baclofen pump implantation in a delayed fashion
18	7/F	Cerebral palsy (hypoxic ischemic injury associated with severe fetomaternal transfusion syndrome)	V	0.55	Severe encephalomalacia of the posterior aspects of the cerebral hemispheres bilaterally with associated dilation of the occipital horns of the lateral ventricles	TD	23	36	No papilledema by post-operative fundoscopic examination

TD: Baclofen test dose, Pl: Baclofen pump implantation, GMFCS: Gross Motor Function Classification Scale; \*Frontal and occipital horn ratio could not be calculated at time of pump insertion given persistent left hemicraniectomy bone defect

prior to ITB pump implantation. Patient #16 required VP shunt placement to manage a CSF leak from the LP site created at the time of the LP. In a 3-week follow-up after shunt placement, there was no difference in the patient's cognition compared with his baseline. Five months after VP shunt placement, the patient underwent placement of an ITB pump without complication. In patient #1, a 6-vear-old boy with a seizure disorder and diffuse cystic encephalomalacia on imaging, a test dose LP revealed an opening pressure of 36 cmH<sub>2</sub>O, and placement of a VP shunt was offered. After VP shunt placement, the patient demonstrated a significant reduction in seizure frequency, with a decrease from 10-15 seizures per day to 4-5 seizures per day. However, no cognitive or functional improvement was noted after shunt placement. The remaining 6 patients with elevated ICP either decided to forgo baclofen pump placement and proceed with ophthalmologic evaluation and possible repeat LP at a later date or opted for a referral to another provider for consideration of implantation using standard implantation guidelines.

#### **DISCUSSION**

Intrathecal baclofen delivery has become an effective treatment for spasticity, however, it continues to be associated with a high risk of complications and morbidity. [6] Postoperative wound breakdown and CSF

leak, which occur in up to 15% of cases, [2] may require complex wound revisions and, in some cases, temporary or permanent CSF diversion to facilitate healing. These complications often lead to hardware contamination necessitating removal and prolonged treatment with antibiotics. We propose that elevated ICP, even in the setting of meticulous surgical technique, raises the risk of postoperative CSF leak; therefore, identification and potential treatment of unrecognized hydrocephalus may mitigate the rate of complications by avoiding implantation in high-risk patients.

In this series, 8/18 (44.4%) pediatric patients with spasticity and dystonia being considered for ITB pump implantation had ICPs in excess of 21 cmH2O as assessed by LP. While alarming in isolation, the observed rate of occult hydrocephalus in our series is actually considerably lower than the 96% rate of occult hydrocephalus observed by Albright et al. in their 2005 study of 24 pediatric patients with cerebral palsy.[1] Notably, only patients with ventriculomegaly were included in the Albright study, whereas all patients deemed to be ITB pump candidates were included in the present study. Although any correlation between ventricular size and ICP is very weak in the cerebral palsy patient population, the inclusion of all patients irrespective of any preoperative concern for elevated ICP in this study may account for the disparity in findings. Similar to the results of Albright et al.,[1] the elevated ICPs found in this study must be

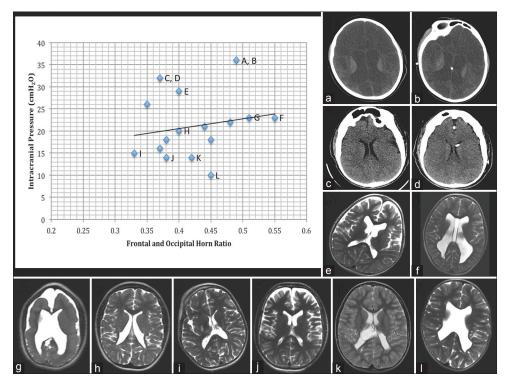


Figure 2: Scatter plot of preoperative frontal and occipital horn ratios and measured opening pressures on sedated lumbar puncture (LP) with selected imaging. Only absolute intracranial pressure (ICP) values were plotted; therefore, patient #12 (who had a recorded opening pressure of >21 cmH<sub>2</sub>O) was not included in the plot or linear regression analysis. In addition, patient #14 was not included in the plot or linear regression analysis because his hemicraniectomy bone defect precluded reliable measurement of a frontal and occipital horn ratio. The line of best fit is y = 22.19x + 11.674; R<sup>2</sup> = 0.038. Letters next to select scatter plot points correspond to the imaging studies presented in panels a–l.(a,b) Pre- and post-ventriculoperitoneal (VP) shunt placement noncontrast head computed tomographies (CTs), respectively, for patient #1.(c,d) Pre- and post-VP shunt placement noncontrast head CTs, respectively, for patient #16. In both patients who underwent VP shunt placement, a modest reduction in ventricular volume was noted after shunting. The remaining panels demonstrate preoperative T2-weighted magnetic resonance imagings (MRIs) for patients #4 (e), #18 (f), #13 (g), #10 (h), #7 (i), #2 (j), #8 (k), and #11 (l). Note the heterogeneity of ventricular anatomy. The presence of periventricular leukomalacia is common in the cerebral palsy patient population; it is particularly pronounced in patients #13 (g) and #11 (l). Significant variability is also noted with respect to the volume of extra-axial cerebrospinal fluid (CSF) spaces, with generous extra-axial CSF seen in cases with elevated (#4, e), borderline (#13, g), and normal (#2, j) CSF opening pressures

interpreted with an understanding of the limitations inherent in LP opening pressure measurements obtained at a single time point, although the one patient in our study who underwent a second LP to confirm the finding of an elevated ICP of 23 cmH<sub>2</sub>O was found, on repeat procedure, to have an ICP of 24 cmH<sub>2</sub>O. An additional limitation, also shared by the Albright *et al.* study, is the use of volatile anesthetics for general anesthesia. We acknowledge the potential for modest ICP elevations with the use of these agents; [9] however, we believe that achieving optimal patient positioning and relaxation, particularly when working with developmentally delayed pediatric patients with spasticity and dystonia, is the paramount determinant of achieving an accurate opening pressure by LP.

Patients with cerebral palsy may harbor arrested (i.e., nonprogressive) hydrocephalus; however, because of the frequent presence of developmental delay and the inability of the child to communicate symptoms, it is not a commonly pursued diagnosis in this patient population.

Unfortunately, impairments in language cognition make it difficult to detect underlying symptoms from elevated ICP in many patients. The finding of occult hydrocephalus in patients who experienced wound breakdown with CSF leak after ITB pump implantation prompted a change in our institutional protocol such that each patient is now screened preoperatively with a cranial imaging study as well as a LP under general anesthesia for assessment of opening pressure.

In our series, we noted a trend toward increased ICP by LP evaluation and frontal and occipital horn ratios; however, this correlation was quite weak. The lack of a significant correlation between ventricular size and measured ICP is consistent with Albright *et al.*, who found no correlation in this regard. We suspect that this is a reflection of the high rates of hydrocephalus *ex vacuo* seen in patients with cerebral palsy. In general, patients with ventriculomegaly in the absence of elevated ICP had evidence of cerebral volume loss on preoperative imaging such as the finding of periventricular leukomalacia. While

we still advocate obtaining preoperative cranial imaging to rule out mass lesions and/or Chiari malformations that might increase the risk of CSF sampling via LP, our data suggests that this preoperative imaging is of limited value with respect to predicting intracranial pressure in the cerebral palsy patient population. For this reason, the clinical algorithm presented relies on opening pressure by LP under controlled conditions (intubated; end-tidal CO<sub>2</sub> controlled) as our best measure of ICP.

ITB pumps confer significant benefits to patients suffering from spasticity, however, surgeons implanting these devices continue to search for ways to limit the historically high rates of associated complications. In this small series of patients, we have mitigated the potential for development of postoperative pseudomeningoceles and CSF leaks since implementing protocol presented. We believe that the assessment of ICP may help identify an especially high-risk cohort of patients for whom ITB pump implantation would likely lead to wound breakdown from CSF leak.[3] Patients who demonstrate elevated ICP on LP while intubated are offered several options in our practice. For high ICP (defined at >25 cmH<sub>2</sub>O), we generally recommend CSF diversion before we would consider placement of a baclofen pump. Patients with ICP between 22 and 25 cmH<sub>2</sub>O are offered ophthalmologic evaluation to assess for evidence of papilledema or optic nerve atrophy, and then a repeat LP performed in a delayed fashion. If the findings on the ophthalmologic examination are normal and the ICP on repeat LP is <21 cmH<sub>2</sub>O, we proceed with pump placement. Should the ophthalmologic examination reveal optic nerve injury/papilledema, a CSF diversion procedure would be encouraged as a means of preventing loss of vision, regardless of future ITB pump implantation plans.

We provide clarity to families that our protocol is not a standard practice currently in the neurosurgical management and assessment of baclofen pump patients, and we offer alternative options to families that wish to consider implantation regardless of ICP measurements. Given the potentially high rate of elevated ICP in the pediatric cerebral palsy population, we advocate for pre-implantation assessment of ICP under controlled conditions and a thoughtful consideration of the neurosurgical management options for patients with elevated ICP. Our protocol reflects our belief that elevated ICP is a risk factor for CSF leak following not just ITB pump implantation, but, any neurosurgical procedure performed that involves durotomy/dural puncture. Therefore, when LP reveals a marked elevation

in intracranial pressure we feel an ethical obligation to convey this finding to the family before proceeding with the elective procedure of pump implantation. We fully expect that the institutional protocol presented in this manuscript will be viewed as somewhat controversial and we hope that it inspires debate. We certainly acknowledge that the threshold of ICP >21 cmH<sub>2</sub>O used in our current clinical algorithm is rather arbitrary and that ICP elevations above this threshold may be tolerated by many patients without wound breakdown or resulting complications. Future work is needed to confirm that patients with elevated ICP are at significantly increased risk of postoperative complications following ITB pump implantation and should seek to identify an ICP threshold above which there is a significantly higher procedural complication rate. Given that ITB pump implantation can result in dramatic improvements in the quality of life for cerebral palsy patients, continued efforts are needed to make this complication fraught procedure safer.

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#### **Conflicts of interest**

There are no conflicts of interest.

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