Neurosurgical Forum LETTERS TO THE EDITOR

Hydrodynamic hydrocephalus in nongalenic arteriovenous fistula

TO THE EDITOR: I read with interest the article by Morales-Gómez et al.² (Morales-Gómez JA, Garza-Oyervides VV, Arenas-Ruiz JA, et al: Hydrocephalus in a patient with an unruptured pial arteriovenous fistula: hydrodynamic considerations, endovascular treatment, and clinical course. *J Neurosurg Pediatr* 19:307–311, March 2017). I salute the authors on their report concerning the rare development of hydrodynamic hydrocephalus in nongalenic cerebral (or pial) arteriovenous fistula. I coined the term “nongalenic cerebral arteriovenous fistula” as part of our 1992 report.¹ Among our 13 cases up to 1992 and the 35 cases reported by others, the case with hydrocephalus was considered obstructive due to varices at the tentorial incisura. In 1994, however, I encountered a case of hydrocephalus (Fig. 1 left) in which the aqueduct was clearly open (Fig. 1 right). There was evidence of venous hypertension with stenosis at the sigmoid sinus (Fig. 2) and reversed flow in the superior sagittal sinus. Considering the rarity of this scenario, one might postulate that venous outflow stenosis is an added requirement for hydrodynamic hydrocephalus to develop in such cases.

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FIG. 1. Images obtained in a 20-month-old male with increased head circumference, developmental delay, and cranial bruit. **Left:** Axial T1-weighted MR image shows marked hydrocephalus and mesial left occipital nongalenic malformation. **Right:** Sagittal T1-weighted MR image shows the aqueduct is patent.

FIG. 2. Lateral common carotid artery injection, late venous phase, showing severe stenosis at the left sigmoid sinus–jugular vein transition.

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Disclosures

The author reports no conflict of interest.

Response

We were pleased to read and discuss with interest the letter by Dr. Lownie and thank him for his interest in our case. We also thank the Editor for allowing us the opportunity to respond. The relationship between dural venous sinus stenosis and hydrocephalus has not been fully elucidated, though correcting venous outflow obstruction may not be enough to effectively treat patients with congenital

hydrocephalus.² It seems that outflow stenosis plays a role in the development of hydrodynamic hydrocephalus, but it is not clear if, in high-flow arteriovenous fistulas, the stenosis is a consequential phenomenon of autoregulation, which worsens the venous hypertension.¹ In our case there was also venous hypertension, sigmoid sinus stenosis, and reversed flow.

The practical relevance at this point in the treatment of hydrodynamic hydrocephalus should be a careful and judicious selection of shunting reserved for refractory or life-threatening cases.

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Ventricular catheter tip proximity to choroid plexus is a key factor in shunt failure

TO THE EDITOR: We read with interest the article by Whitehead et al.⁴ (Whitehead WE, Riva-Cambrin J, Kulkarni AV, et al: Ventricular catheter entry site and not catheter tip location predicts shunt survival: a secondary analysis of 3 large pediatric hydrocephalus studies. *J Neurosurg Pediatr* 19:157–167, February 2017). The study provided a significant amount of data analysis that is beneficial to the neurosurgical literature; however, the title of the article, stating "not catheter tip location," is misleading. Choroid plexus is known to be one of the primary reasons for proximal catheter tip obstruction.³ There is even a specific surgical technique published on how to remove choroid plexus when it is obstructing a ventricular catheter tip via the Seldinger technique with a guidewire and cautery.^{2,3} In 2005 we performed a study very similar that of Whitehead et al. analyzing proximal shunt failures in anterior (frontal) versus posterior (parietal) approaches with

the hypothesis that the posterior approach would have a higher failure rate based on the longer trajectory.¹ We were surprised to discover that the approaches had no statistically significant difference but rather the proximity of the catheter tip to the choroid plexus was the main factor for failure.¹ The results of Whitehead et al. demonstrated that the posterior approach did have a significantly higher failure rate in comparison to anterior approaches, but a very important factor was excluded from the data analysis.

In describing their data collection in the *Methods* section, Whitehead and colleagues state the "relationship to choroid plexus was divided into catheter tips touching the choroid plexus, tips not touching the choroid plexus, and tips completely not in the ventricle (≥ 2 cm of the proximal tip in the brain/cistern)." In the *Results* section, however, they state that the variable of proximity of catheter tip to choroid plexus was excluded from analysis due to interobserver variability. The authors go on to hypothesize in the *Discussion* section that catheters that enter the ventricle from a posterior approach enter through the walls of the ventricle and may be more likely to rest on the ventricular floor or choroid plexus, and this may result in earlier obstruction. If this was actually the case, that the posterior approach catheter tips were closer to the choroid plexus, then it would contradict the title of their manuscript. Whitehead et al. go on to state in the *Conclusions* that unexpectedly they found that entry site selection has a greater effect on shunt survival than ventricular catheter tip placement. We would venture to say that most neurosurgeons would agree that outside of shunt infection the next most common concern for failure when placing a ventricular catheter tip is its proximity to the choroid plexus. Contrary to our findings in 2005, Whitehead et al. found that the anterior (frontal) approaches had a lower failure rate than posterior (parietal) approaches, but again this could all be due to the important excluded variable.¹ What if when reanalyzing the posterior approach data it is discovered that the catheter tips in the posterior approaches were closer to the choroid plexus or that anterior approaches were further away from choroid plexus? This would totally change the conclusions.

Whitehead et al. performed a very thorough study on proximal shunts and the numerous variables that could lead to failure; it would be very interesting if they could take the excluded data on proximity to choroid plexus and have one neuroradiologist review all the data to determine if this was a significant contributor to the overall failure rate. We had the same exact hypothesis 12 years ago in our study—that frontal approaches would have a lower failure rate due to the shorter distance—and were surprised with our final results in that the entry site had no significant effect, but rather the proximity of the tip to the choroid plexus was the key to failure.¹ The overall analysis provided by Whitehead et al. is very helpful, although the conclusions from the study could be substantially strengthened and/or totally altered by including the proximity to choroid plexus data.

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Disclosures

The authors report no conflict of interest.

Response

We thank Dickerman and Reynolds for their comments regarding our paper; we are certainly familiar with Dr. Dickerman's previous work in this area.¹ Dickerman and Reynolds raise the concern, however, that the title of our manuscript is misleading and that proximity of the catheter tip to the choroid plexus may be the most important factor determining shunt survival. They also suggest that we should repeat our analysis using the catheter relationship to choroid plexus variable despite our finding that this variable cannot be reliably determined from the images in our data set. We were in no way trying to be misleading in the title of the manuscript and still believe that the title reflects the results of the clinical study we performed.

Dickerman and Reynolds may be correct about the choroid plexus being a key factor in the determination of shunt survival. This has been stated in the literature and supported anecdotally for many years. Our research, however, was an attempt to measure and quantify the effect this has on the risk of shunt failure. Does placing the catheter away from the choroid plexus reduce the risk of failure by 5% or 50%? We decided to address this question because we still have not identified a surgical technique for accurate shunt placement.^{2,3} Both the endoscopic shunt insertion trial and the HCRN (Hydrocephalus Clinical Research Network) ultrasound study failed to place catheters accurately. We decided that before devoting more resources toward the problem of accurate shunt placement, it was necessary to clearly define the target and measure the magnitude of the effect of accurate placement.

Unfortunately, our experiment showed that it is difficult to determine the catheter tip's relationship to the choroid plexus using routine postoperative scans. When we showed the scans to two independent observers, they frequently disagreed about whether the catheter tip touched

the choroid plexus or not. It can be difficult to see the catheter tip and/or the choroid plexus, depending on the imaging modality, the orientation of the images, and the slice thickness.

If a variable cannot be reliably measured, one cannot determine its effect on outcome. Having only one observer record the variable, as suggested by Dickerman and Reynolds, does not solve the problem. Other groups conducting the same experiment would have trouble reproducing our results. Until a reliable means of determining this variable is found, its exact effect on shunt survival cannot be measured.

In the *Discussion* section of our paper, we do try to explain the difference in shunt survival between anterior and posterior shunts based on proximity of the catheter to the choroid plexus. We hypothesize that a catheter with its long axis pointing at the choroid plexus from an anterior approach may not become obstructed as quickly as a catheter lying in the choroid plexus from a posterior approach. This is not a contradiction because we do not deny that proximity to the choroid plexus may play a role, we were just unable to quantify it in our study. We also think that entry site plays a more significant role in determining this relationship than previously thought because of the effect it has on catheter orientation relative to the surrounding structures.

Finally, Dickerman and Reynolds may not have found entry site to be an important factor in their study for a variety of reasons. First, their patient population was very different from ours and included adults (overall age range 1 month–80 years, mean age 19 years in the anterior shunt group and 31 years in the posterior shunt group), patients with normal pressure hydrocephalus, and patients undergoing shunt revision procedures. Our study includes only pediatric patients undergoing first-time shunt insertions (age range newborn to 18 years, mean 1.5 years). It is possible that the effect of entry site may not be as significant or as easy to detect in their population where the shunt failure rate is lower. Second, their sample size is relatively small ($n = 117$), making it more difficult to detect an effect. Third, data on the relationship of the catheter tip to the choroid plexus were not collected; in fact, no data on the catheter tip location were collected or used in the analyses.

We stand by our title. In our paper we precisely defined what we meant by ventricular catheter location. We evaluated a variety of catheter tip placement variables in an effort to define the best target, and none of them significantly lowered the risk for shunt failure. As we emphasized in our manuscript, we believe that more work needs to be done to identify modifiable risk factors for shunt failure. We are not recommending a practice change based on the analyses performed in our paper. However, we have used this study to justify a randomized controlled trial comparing anterior to posterior entry site (The CSF Shunt Entry Site Trial, clinicaltrials.gov: NCT02425761); the trial is currently accruing subjects.

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