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REVIEW

Classifications of hydrocephalus based on Walter Dandy and his paradigm

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Abstract

Purpose The purpose of this review is to assess the early work of Walter Dandy leading to a paradigm or model that led to the first classification of hydrocephalus and resulted in the development of treatments.

Methods The modern understanding of hydrocephalus begins with the works of Walter Dandy. The purpose of this review is to discuss what was changed in the second decade of the 20th century and how the outcome is useful today. As a result of his experiments during that time he was able to recognize the role of the choroid plexus in the production of cerebrospinal fluid (CSF) within the cerebral ventricles. He then identified the role of obstruction blocking the flow of CSF from the ventricles to the absorption of CSF to the systemic vascular. As a result of those findings he showed that there were two forms of hydrocephalus and therefore the first classification of hydrocephalus into obstructive hydrocephalus and communicating hydrocephalus. Very soon after the publication of the experiments there was general agreement of this work by neurosurgeons working on hydrocephalus. The findings published in "experimental hydrocephalus" became a paradigm useful for all or the vast percentage of those neurosurgeons.

Results Dandy was the first to create a classification of hydrocephalus into obstructive and communicating hydrocephalus. He developed treatments for hydrocephalus such as removal of the choroid plexuses that remained in use until effective valved shunts became available in the 1950s. Essentially all subsequent classifications begin with this paradigm.

Conclusion Over time there have been new classifications primarily focused on specific uses. It is important that classifications in the sciences be reviewed periodically to include new findings and new ideas. Since the expectation that hydrocephalus can be treated or even cured new classifications tend to focus on the physics of CSF, the choice of treatment and the outcome in specific subgroups. These thoughts should be seen as additions to the paradigm.

Keywords Paradigm · Hydrocephalus · Classification · Anomaly

Modern history of hydrocephalus

In the second decade of the twentieth century, Dr. Walter E Dandy (neurosurgeon at Johns Hopkins) together with the help of Dr. Kenneth D Blackfan (a pediatrician) developed a dog model of hydrocephalus related to points of obstruction. In 1919, Dandy published the results of a decade of his research on the nature of hydrocephalus and his thoughts for potential possibilities of treatment [1]. In the first experience of the model, a cotton ball was placed at the bottom of the aqueduct of Sylvius which blocked the flow of CSF and resulted in distention of the lateral and third ventricles as well as the aqueduct itself. This form of hydrocephalus Dandy called "obstructive hydrocephalus." Prior to this experiment, there was a great degree of skepticism as to whether or not hydrocephalus was caused by blockage of CSF flow.

The experiments in this manuscript led to significant understanding most of which are still appropriate today.

1. CSF is formed by the choroid plexus.
2. CSF is formed within the ventricles.
3. CSF is absorbed within the cortical subarachnoid space.
4. The sole communication between the ventricular system and the subarachnoid spaces is through the foramina of Luschka and the foramen of Magendie.
5. Phenolsulphonephthalein placed in the ventricle will prove whether the CSF is present in the subarachnoid space or not.

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(a) If there is no dye in the subarachnoid space, the hydrocephalus is obstructive.
(b) If there is dye found in the spinal fluid the hydrocephalus is communicating.

6. What Dandy called communicating hydrocephalus does not mean that the CSF is blocked at the point of absorption but is blocked prior to getting to the distal cortical subarachnoid space.

The term communicating hydrocephalus has been misunderstood since then. It did not mean that there was a failure of terminal loss of absorption at the venous system. In 1960, Ransohoff and his team tried to make it clear by changing the nomenclature to be that Dandy's obstructive hydrocephalus be changed to intraventricular obstructive hydrocephalus and what Dandy's communicating hydrocephalus to be named extraventricular obstructive hydrocephalus, but the change has not been generally used [2].

Based on these experiments, there were definitely two forms of hydrocephalus. From these observations, Dandy then developed further experiments to find potential treatments based on the points of obstruction. For patients with obstructive hydrocephalus (specifically situation where dye within the ventricle could not be found in the spinal subarachnoid space), Dandy performed what was essentially a third ventriculostomy. These procedures involved an opening *by performing a* subfrontal craniotomy and removing one of the optic nerves to open into the third ventricle. These procedures had high morbidity and were abandoned. Dandy felt that this procedure could be done using an endoscope. This procedure was first performed by Mixter in 1923 [3].

For the treatment of communicating hydrocephalus, he assumed that it would not be possible to get the CSF to a point of absorption, and therefore, he felt that removal of the choroid plexuses was the treatment of choice. He would first remove the choroid plexuses from the frontal ventricles, and if that was not sufficient, he would also do a removal from the third and fourth ventricles. He suggested that this procedure would be best done using an endoscope, and this was an important form of treatment until the general use of valve regulated valves in the 1950s [4].

With the improved understanding of hydrocephalus, new treatment being used, and a great deal of research ongoing, there was a general acceptance of the work of Dandy**.** As a result, his paradigm became widely accepted as it is today.

Paradigm

What is a paradigm in science? Thomas Kuhn in his critical book related to the history and philosophy of science *The Structure of Scientific Revolutions* spent a great deal of the conversation on the concept of paradigm [5]. His definition of paradigm in science was "The standard illustration of various theories in their conceptual, observational and instrumental applications." In other words, it is a model that is accepted by a community involved in a specific area of science. The community that values the paradigm according to Kuhn "is revealed in textbooks, lectures and laboratory exercises." The highest levels of communities will have their own journals, their own meetings to discuss the issues, and at the highest level will have boards that specifies the member of the community. In the case of hydrocephalus, the community of scientists includes neurosurgeons, neurologists, neuroradiologists, neuroscientists, and engineers as well as others studying hydrocephalus. It is also often discussed and taught to students, patients, and caregivers. In hydrocephalus, the fact that the work of Dandy is the paradigm of science in hydrocephalus is supported by the fact that all of the classifications of hydrocephalus discussed here are based on Dandy's work [6–14].

Classification of hydrocephalus

While finishing my residency at Case Western in Cleveland, I had the opportunity to work with the School of Engineering at Case Institute which was a wonderful opportunity. The plan was to develop a mathematical model with the engineers at Case with then to test it over the physics utilizing what I know now as the Dandy paradigm. The model led me to understand and challenge the physics of CSF, and the interest in a classification was needed [17]. In 2000, Professor David McLone asked me to write a book chapter on the classification of hydrocephalus, and I believe I have been working on that since then [10]. In 2008, with the help of the artists at the Barrow Neurological Institute in Phoenix, I put together a *publication* attempting to develop a definition of hydrocephalus and a classification [11]. I spent a significant amount of time with the engineers and the artist and came up with a plan to find a consensus of these issues. The artist's concept of the classification made it easier to get others to understand what was happening and what we were trying to get through. You now see it as Fig. 1. Over the next 2 years, a group of hydrocephalus scientists met and talked and came up with a consensus. It was then discussed at the ASPN, ISPN, and other involved groups. In 2011, after a high percentage of pediatric neurosurgeons had discussed and accepted, with some changes, the classification was published [12].

Over the next decade, the model worked well in the lab and in the operating room. During that time, it was clear that there were some problems with the presentation and the figure. New information needed to be entered. It is clear

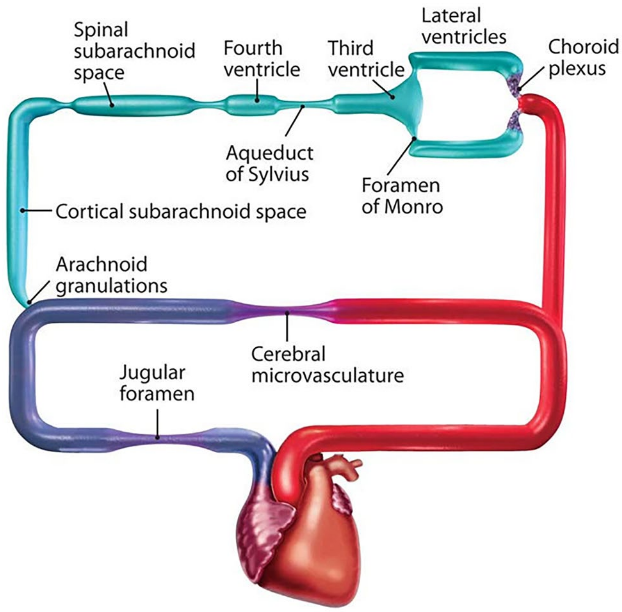

Fig. 1 Intracranial hydrodynamics represented as a circuit diagram with a parallel pathway and cerebral blood flow (with permission from the Barrow Neurologic Institute)

that the lymphatic route for CSF absorption does not show well enough in the model. There is minimal CSF absorbed in the spinal canal, so it is now shown as a cistern. One of the problems with the model as shown has to do with the very rare hydrocephalus with no blockage of the CSF before it gets to the veins or lymphatics. This is excess production of CSF by choroid plexus tumor or hyperemia. I have not been able to figure out how to put that in the model. The new model is now Fig. 2.

Recent classifications

As I did before, for the first classification attempt, I went to PubMed looking for papers with the words hydrocephalus and classification since 2006 to the present. Over that time, there were 449 hits. I read the title of all of them, the abstract of about 20 and decided, because of time and space to limit a discussion to four that were most important to the discussion.

Going through the PubMed search, it became clear to me that it is very difficult to understand what a classification is especially related to hydrocephalus but also more importantly in science. A Google search using the words classification and science led to 436,000,000,000 hits. Google did attempt to help in understanding the interaction of classification and science. The most important discussion of the role of classification was from *Science* in 1974 [15]. In the work of Kuhn related to the philosophy of science, it seems essential that in science, it is necessary that the majority of the community understand what is stated in the classification and how can it be used for teaching, medical management, or experimentation. For my work on classification, I was looking to create a process that would lead to both understanding of the biophysics of hydrocephalus and lead to focused treatments. Going through other discussions of classification, I found four that needed discussion. What did the author or authors expect from the outcome of the new classification?

1. Tully and Dobryn did a thorough assessment of the causes on hydrocephalus in infants. They used the definition from the International Hydrocephalus Imaging Working Group. “Hydrocephalus is an active distention of the ventricular system resulting from inadequate passage of cerebrospinal fluid from its point of production in the ventricles to its point of absorption to the systemic circulation.” The discussion dealt primarily to genetics and problems that began within the uterus or soon after birth. The classification is done well with the intention that it will lead to prospective trials of treatment and outcome [14].
2. Professor Oi updated his extensive discussion of the classification and has now given it the name of “Multi-categorical hydrocephalus classification.” In this process, he identifies three subjects: patient, CSF, and treatment. Each of these subjects has 10 categories. Overall, there are theoretically 72,576,000 patterns of hydrocephalus. This system has been experienced in centers in Japan. With a vast database, this thorough classification has a potential to better understand complex problems of hydrocephalus [8, 16].
3. Professor Thomale has published a “practical approach for a complex disease” [13]. This new classification is based on seven factors related to hydrocephalus stated to being critical to the understanding of hydrocephalus. The seven factors are pulsation, CSF production, major CSF pathways, minor CSF pathways, CSF absorption, venous outflow, and respiration. It is unclear *who is to use the* classification. It seems that at least 4 of the factors are controversial. It is possible that this classification relates to areas that require further experiment or control trials. How are neurosurgeons who care for patients, especially infants with hydrocephalus to use the classification. Some of the factors are difficult to identify. It would be better if the function of this classification were more clear [17].
4. Milan and colleagues from Copenhagen have developed the ASPECT system for clinical use. This classification is essential a well-thought-out check sheet for maintaining data over time for patients with hydrocephalus. It is created so as to be useful by patients to make certain that they have the information of how the hydrocephalus has affected them. It is especially valuable when patients

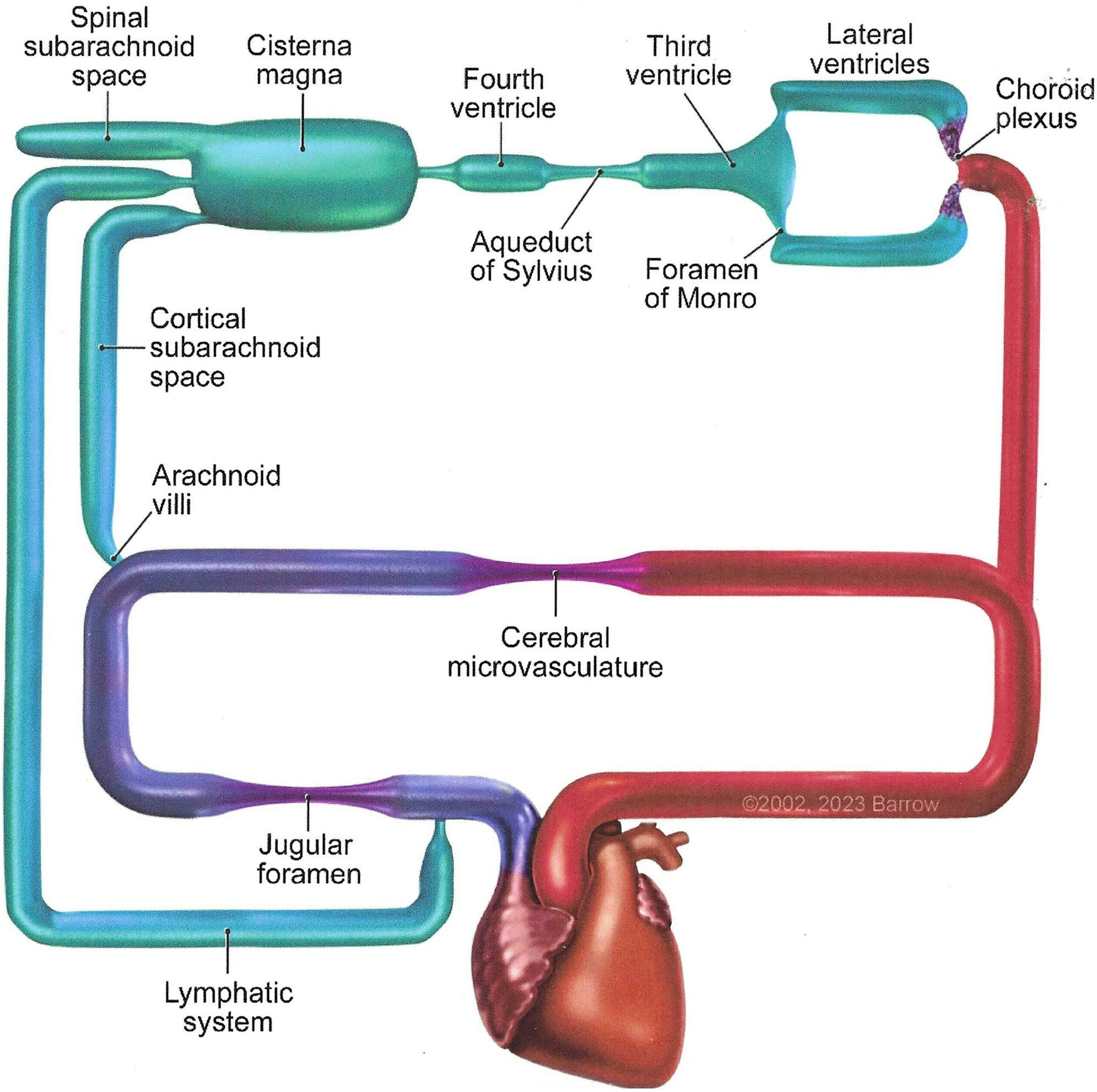

Fig. 2 A slightly different diagram (with permission from the Barrow Neurological Institute)

are seeing new caregiver or transferring from pediatric to adult management or for needed information in times of crisis [7].

Anomaly: learning through observation

The Structure of Scientific Revolutions by Thomas Kuhn is thought to be the most important book on the philosophy and history of science of the twentieth century [18]. One of the most important messages of the book relates to anomaly in science. Anomaly occurs when an experiment in science or surgery ends up with an outcome that is unexpected and is not easily understood by the theories that are expected by the community (paradigm). Kuhn states that scientists generally know what to expect when testing a theory. They are looking through the eye of a paradigm or model that is widely accepted in the community. Over time, there are findings that add to the knowledge of the community. In the science of hydrocephalus, for instance, the finding that CSF is produced across the ependymal walls was not part of the initial paradigm of

Dandy. It did not however have a significant challenge to the paradigm. Kuhn postulated that a change in the paradigm or paradigm shift required the finding of an observation that could not be explained by that paradigm. An unexpected observation of that importance is rare, but it is these anomalies that lead to new understandings.

"Discovery commences with the awareness of anomaly, i.e. with the recognition that nature has somehow violated the paradigm induced expectations that govern normal science." An anomaly will make a physician or scientist take a new look of the paradigm. I tend to become excited when dealing with an anomaly. It is important to understand how to deal with one.

Over a 50-year career, I have been faced with a number of anomalies and, over that time, have learned the steps it takes to understand the effect of an anomaly to the paradigm. If the paradigm cannot explain what has happened, the effect is a revolution and requires a change in the paradigm or in other words requires a "paradigm shift."

Based on Dandy's paradigm as seen in the classification now seen in Fig. 2, I will briefly discuss the management of a difficult anomaly I dealt with over almost 2 decades.

Setting out to find the answer to an anomaly may be called "learning from observation."

This diagram is an artist's demonstration of our sense of the model originally published in 2008. It was effective in our experiments of CSF flow and potential points of blockage of the flow leading to hydrocephalus. After using it for this purpose for over 10 years, it became clear that there were a few problems with the demonstration although not severe ones. CSF leaves the fourth ventricle into the cisterna magna. From there, it disperses to the spinal central canal, the spinal subarachnoid space, and the cortical subarachnoid space. CSF in the spinal subarachnoid space may enter into the spinal cord itself through the perivascular spaces and eventually into the central canal if there is a blockage of flow out of the central canal by a tumor or trauma [19].

The spinal subarachnoid space is seen as a cistern. The lymphatics are indeed important for the absorption of CSF [20–22]. It is known that CSF follows the olfactory nerves through the skull base to mix with the lymphatics of the nose. Some studies have also shown the passing of CSF following around the optic nerves.

Case 1 As far as I can remember, the first of these anomalies in my career occurred when still a resident on call. I saw a 6-year-old boy in the emergency room with a severe headache. A CT scan was obtained, and the ventricles were small (Fig. 3).

Fig. 3 CT scan of a 6-year-old boy with severe headaches showing slit ventricles and an open cistern with collapse of the left lateral ventricle collapsed around the ventricular catheter

The diagnosis by radiology was that the shunt was working well, and he was referred for pain management. In the next 2 weeks, he was back in the ER with the same story and outcome except before he came the third time he had seen an ophthalmologist who found severe papilledema. The ventricular catheter was not working and was replaced with a higher opening pressure valve. I did not understand what happened. Why did the ventricles not get larger? This problem stayed with me for the next 20 years when a second anomaly led me to understand it. Actually, the problem of slit ventricles with extreme increase intracranial pressure is not that uncommon, but there is extreme skepticism as to whether it is real or not [23].

An anomaly does not, of itself, prove a paradigm is not working, but it requires thinking through what has happened and an attempt to understand. Setting out to find the answer to an anomaly may be called "learning from observation." An observation can lead to a paradigm shift if the paradigm cannot deal with the new finding. Some examples of paradigm shifts include the paradigm that was accepted by the community of scientists such as what happened to Ptolemy when Copernicus was able to prove that planets including the earth were circulating around the sun and that the earth itself rotated as the cause of seasons. His findings were not published until his death, and very few scientists accepted it for another 50+years. The proof of the heliocentric universe Kuhn would call that a scientific revolution. It could not become an addition to the Ptolemaic paradigm.

Epiphany and learning through observation are close brothers. Both of these thoughts assume that something new has arisen. With epiphany that something new is thought usually simple and striking. In science, the epiphany is rarely the end of the story. In science, the epiphany often comes as a result of an anomaly. In her book *Slow Looking: the Art and Practice of Learning Through Observation*, Shari Tishman creates a strategy for answering the questions brought up by observation of something you do not understand. The methodology of learning through observation is shown in the chapter on learning through observation in science [24]. Prior to the Renaissance in general and the printing press in particular, learning through observation was limited to farmers and fishermen.

In 1551, an extraordinary physician in Italy, Lusitanus Amatus, who was held in great esteem by kings and popes published books called the *Centuria.* There were 7 of these books in all. Each had 100 medical case reports [25]. These had an important effect in Renaissance medicine [24]. Step 1 was that you have noted something that you had not expected (an anomaly). Second, what was found must be maintained so anyone observing it would have seen the same answer. For this important aspect of the process, the printing press was essential. The most important issue relates to the facts that all of his observations were printed in a special way. What was actually observed was printed in Latin to let it be known that he saw something and that anyone else who saw this thing would see the same and know what was seen was not to be changed. In discussion of anomalies or patients seen with a new disease or injury, the second thing that must be done is there need to be a theory as to what had happened. This theory is not therefore certain. To emphasize this essential aspect of the process, the theory was printed in italics. Using our little boy to start this discussion, he had headaches, he had a shunt, and he had papilledema. Then, the child went into surgery, and a shunt revision treated him. The third is the most important, and I will discuss that in a few examples below. The observer needs to have a theory of what happened [24]. Amatus made that clear by writing the theories or your thoughts about what had happened were written in italics to make certain that it was different. *It cannot be "truth" but it could suggest analyses*, *or experiments to find out*. I had no idea at the time I saw him why this happened. It took 20 years to get the answer. I learned a great deal from that case, and I think it might have been the last time I actually gave up on finding the answer.

Soon after this experience, I developed a relationship with the engineering school at Case Western Reserve. This was an amazing opportunity. Professor Wen Ko was the director of electrical engineering and computer science and worked on implantable ICP monitors and electric stimulation of paraplegic patient. The final person involved was Professor Howard Chizeck of systems and design engineering. We would send a half a day on Wednesdays to review the physics of CSF and hydrocephalus. They taught me engineering, and I taught them the importance from a medical point of view. Our model was based on an electric circuit with the anode being the left ventricle and the cathode was the right atrium. Obviously, this process is depended on the Dandy paradigm.

Case 2 is a patient I first met about 20 years after the boy. The patient was 16 months of age. She was first seen elsewhere at the age of 6 months and found to have a rapidly enlarged head circumference and irritability. At that point, she was documented to have achondroplasia (Figs. 4 and 5).

Over the next 8 years, she had been treated with a VP shunt which failed multiple times. Each failure included severe headaches, no ventriculomegaly, and collapse of the ventricle with the catheter. What was happening? *Her hydrocephalus was due to venous hypertension that was the cause of hydrocephalus in the first place. Her problem related to failure of absorption of CSF from the cortical subarachnoid space and a higher pressure in the cortical subarachnoid space than in the ventricle.* The problem here is well recognized in patients with idiopathic intracranial hypertension or pseudotumor cerebri where the problem is severe headaches and severe headache due to high intracranial venous hypertension. She was treated with a catheter

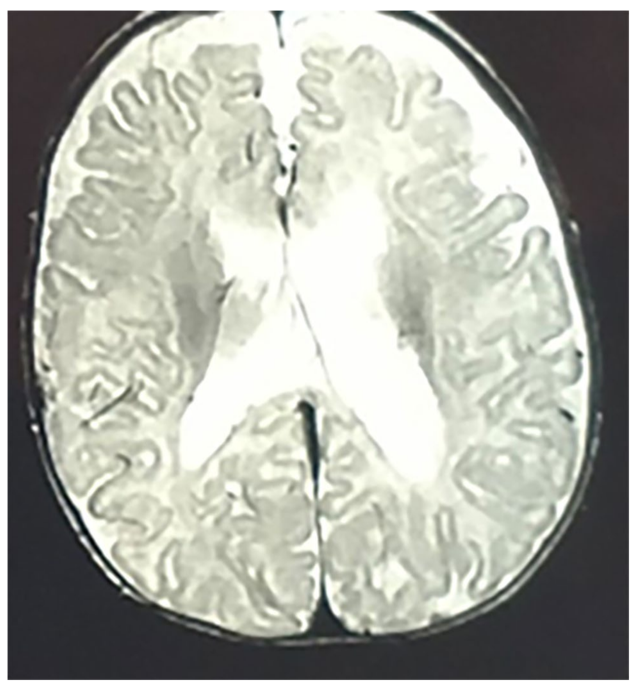

Fig. 4 T2 weighted MRI of a 6-month-old child showing excess CSF in the ventricles as well as in the cortical subarachnoid space

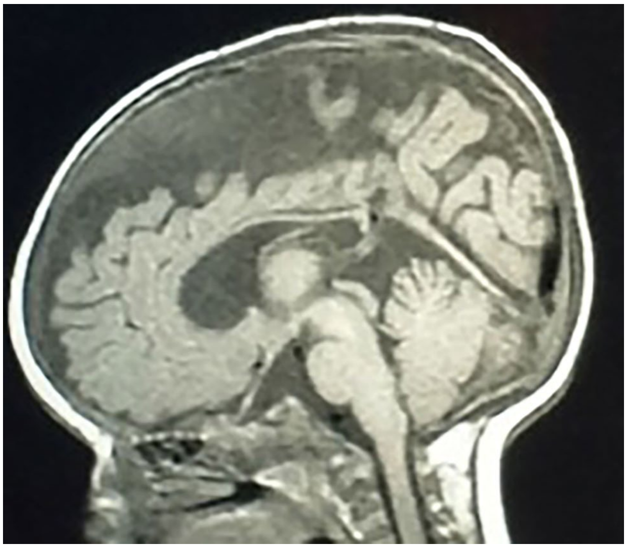

Fig. 5 T1 weighted MRI showing sagittal MRI showing excess CSF leading to ventriculomegaly but also excessive CSF in the cortical subarachnoid space

in the subarachnoid space spliced into the ventricular shunt above the valve. This procedure would lead to the ventricle and the cortical subarachnoid spaces maintaining the same pressure [26]. At that point, I had been managing patients with this problem to remove the ventricular catheter and performing a valved lumbo-peritoneal shunt [27, 28]. Unfortunately, the lumbar spine in achondroplasia made the LP shunt impossible. The patient did well for the next 6 years. At that point, she presented with low-pressure hydrocephalus (Fig. 6).

Fig. 6 CT ventriculogram showing excess CSF leading to ventriculomegaly with minimal CSF in the cortical subarachnoid space

There was flow from the cortical subarachnoid space and not the ventricle where the catheter had failed. This is evidence of the importance of a differential pressure between the cortical subarachnoid space and the ventricle. In retrospect, this would have been an ideal opportunity to perform an endoscopic third ventriculostomy. My understanding of the extraordinary importance of the trans mantle pressure was evolving at the time. Negative pressure hydrocephalus results from lower pressure in the cortical subarachnoid space than the ventricle and can be managed by opening the ventricle to the subarachnoid space using a third ventriculostomy or using two catheters proximal to the valve in a shunt.

Conclusions

More than 30 years after the inability to understand the problems I dealt with as a resident, we now have an understanding of causality. Severe intracranial hypertension will lead to hydrocephalus only in infants whose heads are still growing and whose shunt is placed in infancy [29]. In older children or adults, abnormally high pressure in the venous pressure in the intracranial sinuses is the causation of an increase in the ICP and therefore in idiopathic intracranial hypertension (pseudotumor cerebri) [30]. There are a number of conditions in babies that may lead to hydrocephalus due to high venous pressure. What they all have in common is that the calvarium does not follow the Monro-Kelle hypothesis in that the volume of the skull is not rigid. The best known and studied are achondroplasia, craniofacial conditions, and some cases of Chiari II malformation. In these situations, the trans mantle pressure difference is not between the ventricle and the cortical subarachnoid space but essentially between the ventricle and that atmospheric pressure. Shunting in infancy allows the skull to solidify. In issues like achondroplasia, the venous pressure is still high. Slit ventricles are almost certain to occur. If the venous pressure is only slightly high, the treatment could be to increase the opening of the valve and making certain that there is a device to prevent siphoning. Shunting the ventricle will always create a pressure differential with the pressure in the ventricle will be lower than in the cortical subarachnoid space. The ventricle becomes smaller because of this differential. With time, there is often noted to have a thickening of the skull itself. In these patients, the pressure in the ventricles is lower than in the cortical subarachnoid space with asymmetrical ventricles. The ventricle with the catheter collapses and CSF cannot get to a point of absorption. This condition has been called "normal volume hydrocephalus" I first heard from a talk from Peter Carmel. I had to wait until I saw the paper to understand it, but it still took a long time to realize it was a real thing [31]. More recently, a large number of children with shunts and shunt revisions at the Children's Hospital of Los Angeles looked specifically on this problem. They found that 9% developed this condition. Their median age at first shunt was 8 months [32]. It is difficult and dangerous to perform third ventriculostomy in patients with slit ventricles, and usually, it is best to place a catheter into the cortical subarachnoid space and splice it to the shunt so that both catheters are above the valve. It is very important that there never be two valves in a single patient. One of the valves *must* fail. The catheters must see the same pressures.

Author contributions I did all of the thought and writing of the paper

Declarations

Conflict of interest There are no competing interests, and there has been no funding for this paper.

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