Emergency suboccipital decompression for respiratory arrest during supratentorial surgery: the untold story of a surgeon’s courage in times of despair

Historical vignette

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The odyssey leading to the discovery of herniation syndromes was prolonged due to a lack of early understanding of the underlying pathophysiology. In 1896, Leonard Hill documented transtentorial pressure gradients as the intervening phenomenon involved in uncal herniation. In 1904, James Collier became the first to describe cerebellar tonsillar herniation as a “false localizing sign” often associated with intracranial tumors. During the infancy of neurological surgery, management of increased intracranial pressure and an improved understanding of brain herniation syndromes were of the utmost importance in achieving a safe technique. Harvey Cushing provided seminal contributions in understanding the pathophysiology of increased intracranial pressure and resulting cardiopulmonary effects.

Cushing believed that tonsillar herniation was a cause of acute cardiorespiratory compromise in patients with intracranial tumors. In this vignette, we describe the untold story of Cushing’s heroic attempt to treat respiratory arrest operatively during supratentorial tumor surgery with an emergency suboccipital craniectomy to relieve the medullary dysfunction that he believed was caused by compression from tonsillar herniation. This case illustrates a surgeon’s determination and courage in fighting for his patient’s life in the most desperate of times.

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Herniation Syndromes and Increased ICP: Historical Considerations

Sir Jonathan Hutchinson made one of the earliest reported observations regarding the signs of uncal herniation in 1867. He reported on two cases of unilateral pupillary dilation as the result of trauma associated with intracranial hemorrhage. His postulated pathophysiology was compression of the third cranial nerve. He wrote:

...from the position of the clot there can be little doubt that the third nerve is compressed and thus, the dilatation of the pupil is explained. These two cases, so exactly parallel, seem to supply us with a new and very valuable symptom indicative of effusion of blood in this situation.

It would take almost two more decades before further contributions to the understanding of herniation syndromes through observation of unilateral pupillary dilation would be made by William MacCormac. Nonetheless...
less, Hutchinson provided the surgeons of that time with neurological localization for trephining.

William Macewen attributed unilateral pupillary dilation to a unilateral cortical lesion. Up to this time, the connection between uncal herniation and unilateral pupillary dilation had been unclear. Macewen speculated that the cause of dilation was an "irritation and paralysis respectively of the oculomotor nerve." Pupillary dilation was considered the result of "vascular changes in connection with the cerebral lesion."11,14

In 1896, Leonard Hill’s dog experiments advanced the understanding of brain herniation syndromes. Utilizing a fluid-filled pouch to cause compression within the supratentorial space, he was able to demonstrate unequal distribution of pressure within the brain: a pressure difference between the supratentorial and infratentorial compartment. Hill concluded that this pressure differential was the result of "viscosity of the cerebral substance" and "the interposition of the falx cerebri and the tentorium cerebelli." He noted a pressure difference across two structures: the foramen magnum and the tentorial notch.9 These data established the basis for recognizing the critical points at which compression of neighboring structures may occur. Other investigators have used this data to guide their experiments related to cerebral herniation.11,14,15

Inspired by Hill’s experiments, Pierre Marie expanded the understanding of herniation syndromes in his studies of two cases of intracranial hemorrhage in 1899. In the first case, hemorrhage of the external lenticular nucleus had shifted the vermis to the right, and in the second case, a thalamic hemorrhage was compressing the left cerebellar hemisphere and causing protrusion of the cerebellar tonsils that “appeared to have engaged the occipital foramen.” In these cases, Marie referenced Hill’s conclusion that compression from a hemorrhage could cause tonsillar herniation through the foramen magnum, trapping the medulla. If the pressure was sufficient, it would compress nearby vessels and lead to bulbar ischemia.11,14

In 1900, Marie would present two additional cases: thalamic hemorrhage resulting in herniation. In response to Alquier’s conclusions, Marie stated: “in my opinion, the trauma of autopsy projects a portion of the cerebellum through the occipital foramen,” and attributed herniation to post-mortal artifact.11 Therefore, there still remained some controversy regarding brain herniation and its consequences.

The following studies of increased ICP contributed to the eventual understanding of herniation syndromes. In 1835, Joseph Francois Malgaigne studied raised ICP in rabbits by injection of fluid into the subarachnoid space. Similarly, in 1866, Ernst von Leyden recorded ICP in dogs.8 A monograph written by von Bergmann in 1899 detailed the clinical symptoms of increased intracerebral or cerebrospinal fluid pressure. Although von Bergmann attributed the symptoms of increased ICP to a reduction in cerebral blood flow, the clinical consequences of increased ICP were well described but yet to be attributed to herniation:

With further increase of the space occupying pathological mass inside the cranial cavity, deep coma follows, the pupils become dilated without reaction to light and the pulse becomes weak and fluttering. The end stage is generally circulatory and respiratory paralysis followed by primary respiratory arrest and death.16

In 1902, Cushing published results detailing vaso-motor center control of blood pressure during cerebral compression. Cushing embarked on his studies of increased intracranial tension in Berne, Switzerland where he worked in the laboratories of the distinguished physiologist and pioneer in blood pressure studies, Hugo Kroenecker. The experimental setup Cushing used was based on Hill’s work.8 By the end of 1901, Cushing published his observations of the effects of elevated ICP on blood pressure, known today as Cushing’s Reflex:

As a result of these experiments a simple and definite law may be established, namely, that an increase of intracranial tension occasions a rise of blood pressure which tends to find a level slightly above that of the pressure exerted against the medulla.1

Cushing’s laboratory work resulted in 5 other publications in 1901–1903, including a publication in 1902, which connected the effects of tonsillar herniation with cardiorespiratory failure.2–5,10 In his 1902 paper, he remembered a patient with suspected temporal lobe abscess from ottis media who presented with “profound stupor, with a subnormal temperature, a snoring respiration, a heart beat of fifty to the minute, a high-tension pulse...” indicating an increase in intracranial tension. The patient stopped his respiration upon “elevation of the temporal lobe.” Cushing describes the reason:

The medullary center, evidently suffering already from compression effects, could not endure this additional burden. On immediately releasing the pressure, and after some artificial respiratory efforts, spontaneous breathing returned.5

Although Cushing drained the abscess, respiration ceased again and was never resumed despite aggressive artificial respiration.5

In 1904, James Collier reported, “in many cases of intracranial tumor of long duration, it was found post-mortem that the posterior inferior part of the cerebellum had been pushed down and backwards into the foramen magnum and the medulla itself somewhat caudally displaced.” He described the structures as forming a “cone shaped plug” filling the foramen magnum. He postulated that tonsillar herniation was a late and false localizing sign often observed with intracranial tumors.1 The findings of Collier were reinforced the following year when Louis Alquier reported similar findings in autopsies of patients with brain tumors. He speculated that the brain tissue is “forced to migrate due to pressure effect of the tumor” resulting in herniation. In response to Alquier’s conclusions, Marie stated: “in my opinion, the trauma of autopsy projects a portion of the cerebellum through the occipital foramen,” and attributed herniation to post-mortem artifact.14,15 Therefore, there still remained some controversy regarding brain herniation and its consequences.

Cushing noted Collier’s notion of “cerebellar pressure cone” in his book, Tumours of the Nervus Acusticus and Syndrome of the Cerebellopontine Angle in 1917 (Fig. 1).6 In this book, Cushing describes his experience with a patient who underwent a posterior fossa decompression for respiratory arrest:

In one patient, whose respiration had suddenly ceased in the ward the day of admission, artificial respiration was imme-
Immediately instituted and an operation hurriedly prepared for and performed, with resumption of spontaneous breathing on the evacuation of a gliomatous cyst after fifty minutes of artificial respiration: this occurred eight years ago and the patient recovered and is still living.

Cushing believed that increased intracranial tension may lead to tonsillar herniation and medullary compression causing respiratory failure. It is this understanding that would foreshadow one of his heroic efforts to come.

In 1909, while removing a supratentorial tumor, he attempted emergency decompression of the medulla oblongata in expectation of relieving possible bulbar ischemia due to compression. The following case details illustrate his desperate attempts during desperate moments.

**Case Presentation**

In 1909, a 9-year-old boy presented with headaches, nausea, vomiting, and decreased vision. Positive findings on physical examination included skull tenderness on percussion, particularly over the frontal region. Cranial nerve examination was remarkable for weakness of the right lateral rectus muscle. Examination of the fundi disclosed a right choked disc (D = diopeter) “easily to 5D and left 6D.” There was a very marked right facial weakness. Cushing decided to proceed with a subtemporal decompression in the absence of adequate localizing signs.

At the time, he used a tourniquet around the head to decrease scalp bleeding. The following is the note dictated by Cushing:

**Operation**

*Exploration and Decompression, respiratory failure.*

*Ether (Davis) Tourniquet*

Bone flap was turned down from the right hemisphere. The bone itself was thin, vascular, and with the sand-papery feel on its inner surface present in the previous case of Kettle. The bone was greatly thinned in patches and showed the marks of the cerebral convolutions.

The dura was exceedingly tense, and thinking that in all probability this was due to an internal hydrocephalus, the effort was made to aspirate the lateral ventricle in two places, both taps were dry.

The usual subtemporal defect was made and the dura was then opened, and protruded to a marked degree. The opening was enlarged, but the rapidly forming hernia lead to a rupture of one of the large veins traversing the wound, possibly a vein communicating between cortex and dura, and for a moment there was considerable bleeding. The bulging temporal lobe was controlled by a pledget of cotton, the child was turned on his side and a lumbar puncture was performed. The fluid was under considerable tension. About an ounce was allowed to drip away during the remainder of the procedure. The brain settled down sufficiently to allow satisfactory dural defect, no evidence of the underlying growth being present in the exposed convolutions.

The bone flap was replaced and sutured in position. As the skin sutures were being placed, respiration suddenly ceased, the needle having been withdrawn some time before from the spinal meninges.

Efforts were made to continue artificial respiration. The child was placed on his face on the cerebellar table, but no exchange of air could be obtained in that way, the chest being very soft and there being no rebound to compression.

The child was turned on its back and a tracheotomy performed and respiration kept up with bellows. The heart apparently having seized at this time. The suboccipital region was opened as in the usual cerebellar procedure in the hope that a jammed medulla might be present. There was no evidence of this however. There was complete absence of indications of the usual foraminal hernia.

Just what was the cause of the respiratory failure is uncertain—possibly thymus- or profound anesthesia.

The brain was removed through the operative openings, disclosing a large growth occupying the whole frontotemporal region.

(Dr. Cushing)

Yale Harvey Cushing Archives

**Part of the Letter from Dr. Cushing to the Referring Physician, Dr. Stickney**

The knowledge that the operation has done nothing more...
than hurry the poor youngster’s death is no special comfort, and the grief of the parents, of course, is very hard to witness, for I felt justified in telling them that decompression which I purposed to do was perfectly free from any special risk. I am sorry that they did not permit a thorough postmortem examination, but I did not care to press this. The only other fatality of this sort which I have had proof to be associated with a persistent thymus (status lymphaticus), which was a comforting thought in a way; but I do not know that this would have reconciled us anymore in the little boy’s case.

I am so sorry that this unlooked for outcome should have resulted in this patient of yours, but I hope that Mr. and Mrs. Woods will feel that everything possible was done to help the poor little chap and otherwise was inevitable.

Most sincerely yours,

Harvey Cushing

Yale Harvey Cushing Archives

Cushing’s curiosity led him to remove the patient’s brain (performed as part of the autopsy) at the end of the procedure in the operating room to discover the location of the tumor. The drainage of cerebrospinal fluid through the lumbar puncture needle may have increased the transtentorial pressure gradient and contributed to respiratory arrest. The above letter illustrates Cushing’s apologetic tone to the referring physician and describes another possible condition responsible for the boy’s demise. Based on our review of the Cushing Tumor Registry, Cushing never attempted another emergency suboccipital craniectomy for possible tonsillar herniation during surgery for a probable supratentorial tumor.

Disclaimer

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

References

3. Cushing H: Concerning a definite regulatory mechanism of the

4. Cushing H: [Physiological and anatomical observations of the influence of brain compression on intracranial blood circulation and some other related phenomenon.] Mitteilungen aus den Grenzgebieten der Medizin und Chirurgie 9:773–808, 1902 (Ger)
16. Von Bergmann E: [The Surgical Treatment of Brain Disease, ed 3.] Berlin: Verlag Von August Hirschwald, 1899 (Ger)