The Pursuit of a Cholesteatoma by Harvey Cushing: Staged Approach to a Complex Skull Base Tumor

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Abstract

Objective The evolution of neurosurgical techniques during Harvey Cushing’s practice was immense. The authors illustrate this evolution using archived historical records from Harvey Cushing.

Setting Historical patient records retained by the Cushing Center at Yale University Department of Neurosurgery.

Design The authors present the case of one of Cushing’s patients with a cholesteatoma.

Results Cushing’s surgical treatment of a cholesteatoma extending into the skull base is an example of his meticulous documentation and accelerated surgical techniques.

Conclusions This case demonstrates how neurosurgical techniques advanced in the management of complex skull base tumors via a staged approach through the middle and posterior fossae at a time long before the development of modern skull base surgery.

Keywords ► skull base surgery  ▶ Harvey Cushing  ▶ neurosurgical procedures  ▶ cholesteatoma

“A good paper might be written on the subject of this woman’s case under the title The Pursuit of a Cholesteatoma”

—Harvey Cushing

Introduction

The practice of neurosurgery evolved around the turn of the 20th century with the introduction of antisepsis, anesthesia, and cerebral localization.1 During this time, Harvey Cushing (1869–1939) revolutionized the practice of neurologic surgery from palliative care into a distinct discipline with significant therapeutic implications. Later, he further extended the field by implementing imaging, electrocautery, and radiation that form the core of the modern practice of neurosurgery.2

The evolution of neurosurgical techniques during Cushing’s practice was immense. We present the case of a patient of Cushing with a cholesteatoma to illustrate this phenomenon. This case demonstrates how neurosurgical techniques evolved in the management of complex skull base tumors via a staged approach through the middle and posterior fossae. In addition, Cushing was impressed enough with this case to suggest the title of our article, per the quote from Cushing at the start of this article.

First Two Hospital Admissions

The following notes and text, until the Discussion section, are from Harvey Cushing’s original notes archived in Cushing’s patient records stored in the Cushing Center at the Yale University Department of Neurosurgery.

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History
In December 1926, the patient was admitted in the hospital with left facial numbness, first time noticed about the left upper lip/C24 15 months prior to this admission. This paraesthesia slowly spread to involve the left side of her face. There was diminution of hearing of the left ear. X-rays showed increased intracranial pressure without any localizing signs. Dr. Horrax found tortuous retinal veins with absent optic cup. The laboratory findings and lumbar puncture results were non-contributory. Dr. Horrax suspected a cerebello-pontine angle tumor. The patient was discharged without treatment.

In May 1927, the patient was readmitted with the same symptoms, but slightly exaggerated. The neurological examination showed a possible partial anosmia on the left side, marked hyperaesthesia on the left side of the face, absent left corneal reflex, protrusion of the tongue to the right with a fine tremor, a lack of sensation of taste on the left side of her tongue, absent abdominal reflexes, and exaggerated deep tendon reflexes. She believed she did not receive “enough attention” in the hospital and insisted on going home and was discharged four days after entry without treatment.

After discharge, the patient did fairly well and noticed no new symptoms until three months before her third admission.

Third Hospital Admission

Summary of Positive Findings (→ Fig. 1)
December 7, 1928
Dr. Schreiber

Subjective
1. Increasing numbness of left face, and diplopia for 4½ years.
2. Frequent dizzy spells without loss of consciousness. Three weeks ago had one with loss of consciousness.
3. Impairment of memory and cerebration.
4. Difficulty in voiding.

Objective
1. Complete impairment of left trigeminus.
2. Left external rectus palsy.
4. Increased right elbow jerk, and left knee kick.
5. Crusting lesion of left nares.
6. Keratitis of left eye.
7. Suggestion of aphasia.

IMPRESSION: Meningioma of left temporal fossa much in the same position as that of (the name of another patient) who was just discharged from C-2.

December 8, 1928
Dr. Schreiber

Seen in O.R. by Dr. Cushing. “Very likely temporal fossa meningioma.”

Special Note
December 22, 1928
Dr. Cushing

This was a most interesting, encouraging, and suggestive operation in a case which offered very little more than an exploration with perhaps removal of some fragments of a meningioma. The diagnosis of a meningioma involving the Gasserian envelopes was a natural one, in view of the patient’s symptomatology. The operation was relayed with Dr. Horrax who turned down the usual flap of the magnified ganglion type for operations in this region.

This case is not only interesting in itself, but illuminating to me on the ground that it shows possibly a novel way of getting at the Gasserian ganglion, absolutely free from any difficulty with bleeding, even from the meningeal artery. I think that it could be done easily enough through the ordinary ganglion opened without bone flap, though of course a bone flap would greatly facilitate the operation. Indeed, it would hardly be necessary to expose the whole ganglion as was done in this

Fig. 1 (A, B) Preoperative patient images demonstrating cranial nerve palsies. (Image courtesy of the Cushing Center at Yale University Department of Neurosurgery.)
case. I feel that if the dura is put on tension down to the region of the foramen spinosum and a small puncture is then made on each side of the meningeal artery with an electric needle, while the brain is necessarily elevated, it will be possible to put a clip easily on the vessel and then to make an opening through the dura, which will allow the immediate escape of cerebrospinal fluid. Then on increasing this opening along in the direction of the third and second divisions, it would be easily possible to elevate the brain so that with the electric needle the dura can be opened over the sensory root by electrical methods and the fibers can be picked out directly from above instead of underneath the dura, as is the usual method of procedure. As a matter of fact, this is not a vastly different operation in which the dura is reflected upward, and incision is made in such a position as to directly enter the sheath of the root. However, for our method of operating, with the patient lying down, I think it would be much easier with the patient sitting, than the operation which we do. Unquestionably, it would be with the patient lying down, I think it would be much easier sheath of the root. However, for our method of operating, incision is made in such a position as to directly enter the electrical methods and the needle the dura can be opened over the sensory root by

Operative Note
December 22, 1928
Anesthesia - Novocain

The bone flap had been turned down by Dr. Horrax without difficulty, the brain being found under considerable tension. He had made a subtemporal defect and had begun to strip the dura from the temporal fossa in the direction of the third division of the ganglion.

At this juncture I took over the procedure.

As stated, the dura was so tense that in view of the fact that the patient was not under general anesthesia, I thought in all probability there would be an intradural tumor. I stripped the dura away from the bone well down to the attachment of the 2nd and 3rd divisions and elevated the dura enough so that had there been the margin of a meningoima in this region I certainly should have palpated it. Moreover, there was no increased vascularity which speaks against a meningoima.

In view of these facts, I thought I would see if I could get fluid by nicking the dura at its point of attachment just as we would in a transfrontal exploration to expose the optic nerves. I accordingly with the hook-knife, holding the brain well upward, made a short incision and an immense amount of fluid escaped, completely lowering tension. I then increased this incision in the usual fashion by putting in a pledget of cotton to hold the brain away and then incising the dura until I had an opening in the dura corresponding practically to that shown in sketch #1 (►Fig. 2). It was then possible to elevate the temporal lobe, well protected by dura, so that I could see the base of the fossa. I was surprised to find that instead of the fossa rounding out below, as I looked in toward the region of the cavernous sinus it seemed to be distinctly bulging, and though it was bulging it was soft as though it contained fluid. Never having exposed in an operation the Gasserian ganglion from this aspect, I was at a loss to know whether this was due merely to fluid or whether it was a normal condition, and I accordingly made a small incision through what I took to be the upper envelopes of the ganglion, and came down upon unmistakable nerve tissue in which I could see the fibers running in the expected direction. Tissue, however, looked edematous, and I finally enlarged the opening, brushed the dura to the side and coagulated it so that it tended to shrink up and pucker away from the field until I had the top of the ganglion exposed about as shown in ►Fig. 2.

The ganglion had a most peculiar edematous and yellowish appearance, and on gradually picking at the fibers, I found that the fibers of the first division shredded out away from the region of the cavernous sinus and there was exposed the unmistakable mass of glistening cells of the contents of a cholesteatoma, the upper wall of which I must have broken into while shredding these fibers away. This appearance is shown in sketch #2 (►Fig. 2; please see inset II).
It then pulled the fibers of the first and 2nd divisions outward and cut it off about the foramen of exit and reflected the thin and shredded ganglion upward, as shown in Fig. 2; please see inset III). This maneuver brought into view the whole upper surface of the cholesteatoma, the cellular contents of which were first scooped out for histological study and then sucked out, leaving a perfectly clean fossa. I then started to spray the wall of this fossa, hoping that I might kill off the epithelial cells of the membrane, but in so doing, the membrane began to pucker up and I found that I could pick it up and peel it out completely from the whole fossa. This small bit of tissue has been so destroyed by the spraying that it will not be worth histological study.

As a final step in the operation, the fibers of the third division were picked up, leaving the motor root intact in its bed.

Sketch #4 (Fig. 2; please see inset IV) shows roughly the appearance of the conclusion of the operation, the opening possibly being a little too large. The motor root can be seen in the posterior part of the field, the cavernous sinus at the upper anterior part of the field, and at the place marked X, there was what appeared to be nothing but a thin membrane which apparently lay between the base of the skull and the tissues below without the presence of bone. I feared that this might overlie a cell in this region though it would be a peculiar place for a mastoid cell, and I am consequently did not open it.

The wound was left absolutely dry and the cavity was filled with salt solution. Flap replaced and closed securely in position, the larger part of the closure being taken over by Dr. Horrax.

(Dr. Cushing)

Pathology Report
December 22, 1929
Dr. Eisenhardt

Examination of tissue by supravital technique shows numerous crystals, calcium deposits and debris. No epithelial cells identified (Fig. 3).

Impression: Cholesteatoma
The patient had an uneventful postoperative course.

Discharge Note
January 3, 1929
Dr. Schreiber
Excellent recovery. Keratitis of the left eye completely healed. Dizziness much improved. Told to wear frosted glass over left eye (Fig. 4).

Fourth Hospital Admission
Summary of Positive Findings
November 2, 1929
Dr. Oldberg

Subjective
1. History of ataxia and numbness of the left side of the face 2½ yrs. duration preceding an operation performed at this hospital on Dec. 22, 1928.
2. History of low left temporal bone flap on Dec. 22, 1928, disclosing and extirpating cholesteatoma of left Gasserian ganglion.
3. Remission of many of preoperative symptoms for 6 mos. following this operation.
4. History of recurrence and exaggeration of vertigo and ataxia, with development of dysarthria and dysphagia for 3–5 mos.

Objective
1. Low left temporal bone flap scar, well healed, with soft decompression.
2. Questionable slight bilateral chronic choked disc, with slight secondary atrophy on the right.
3. Nystagmus and left abducens paresis.
4. Right lower facial weakness of central type.

Fig. 3  Resected tumor. (A) Gross specimen. (B) Histology. (Images courtesy of the Cushing Center at Yale University Department of Neurosurgery.)
5. Bilateral moderate diminution of auditory acuity.
7. Ataxia, most marked in Romberg and staggering gait.

IMPRESSION: Caudal extension of cholesteatoma, originally arising in left Gasserian ganglion.

Special Note
November 8, 1929

Dr. Cushing

A good paper might be written on the subject of this woman’s case under the title The Pursuit of a Cholesteatoma.

If I remember the story correctly, she came in here a year or two ago with pain in the left trigeminal region associated with hypesthesia. I assumed naturally enough that she probably had a meningioma of the trigeminal sheath. At the operation, I came down upon a trigeminal nerve which was greatly thinned out and pushed upward by an underlying cholesteatoma about as big as a marble. I cleaned this tumor out as I thought thoroughly and completely but in the process endeavored to save what I could of the trigeminal nerve. My mistake probably lay in so doing for if I had sacrificed the nerve I might have seen that tumor was passing down through the dural canal for its sensory root or perhaps better, was squeezing up from below, though I did not then consider this possibility.

She kept having trigeminal pain for a long time which mystified me and I regretted that I had not divided her sensory root.

I had lost track of the woman until I saw her a short time ago when it was perfectly evident to me that she had definite cerebellar symptoms and I consequently assumed that they must be due to the subtentorial extension of this old growth. She had no chocked disc, to be sure, and if I remember correctly, no pressure symptoms but very definite and to me unmistakable incoordination. I consequently without any great enthusiasm, suggested that I might look in behind and see if we would find some of the growth but I anticipated finding if anything a small lesion with which I could scarcely expect to deal.

The operation was relayed with Dr. Horrax.

Operative Note
November 8, 1929

Anesthesia-Novocain

Suboccipital Exploration; Disclosure of Hard Cholesteatoma Extending up Toward the Incisura and Down to the Foramen Magnum, through the Center of Which Passed the Trigeminal Nerve. Extensive Radical Removal of this Growth with the Larger Part of its Capsule. A few fragments that were adherent to the side of the pons and to the vertebral artery were left in situ.

Dr. Horrax had made a primary suboccipital exploration without opening the dura when I entered the field, the exposure having been made at my suggestion well off on the left side and without extensive exposure on the right. The dura was somewhat tense and on opening it the cerebellum was protruding so that I felt almost certainly there must be a tumor. Patient at this time began vomiting and there were occasional attacks of vomiting during the rest of the procedure. I fortunately succeeded in getting an abundance of fluid and was able to elevate the left cerebellar hemisphere and by good luck met with no anchoring veins. I retraced the cerebellum sufficiently as that I could get a view well up above the 8th nerve and there I saw a small, minute glistening point, unmistakably a cholesteatoma. I clipped this vein and then divided electrically, fortunately without accident. It was then possible to brush the cerebellum in this region away from the tumor

Fig. 4 (A, B) Postoperative patient images after the first stage of her operation. (Image courtesy of the Cushing Center at Yale University Department of Neurosurgery.)
The 7th and 8th nerves were divided long enough to allow the cerebellum to drop back in its position. Closure was performed by Dr. Horrax.

The huge cavity was thoroughly cleared out by irrigation and the tumor and began clipping and sucking out great cholesteatomatous masses until I had a cup large enough to put in the thumb. Even so, I had not even begun to complete the enucleation. Finally I succeeded in working in between the bundles of the 7th and 8th and the lower nerves and here again I got a large excavation. It was finally possible, by drawing out tissue with the rongeurs, gradually to begin to loosen capsule and in the course of the next two hours I succeeded in cleaning out, as far as I could tell, the entire lower capsule, meanwhile exposing the superior cerebellar peduncle to which the shell of tumor was quite definitely adherent and to get a view of the right cerebral peduncle. Some idea from this may be gained of the huge cavity. I think I may very possibly have damaged the superior cerebellar peduncle as some nerve tissue stripped away as I withdrew the capsule of the tumor from this region.

I finally was able to work underneath the 9th, 10th, and 12th and draw out the entire lower pole of tumor, fortunately without bleeding. In this procedure, the left vertebral artery, passing up toward the basilar, was fully exposed and there were 1–2 shreds of capsule which were slightly adherent to it that I did not quite dare to remove. The 5th nerve apparently ran directly through the center of the tumor, though I did not encounter it or see it until I had begun cleaning out above the 8th nerve in the early stages of the operation. I finally divided the 5th nerve and merely left a tag of it hanging on the fossa as it entered its canal just below the incisura of the tentorium. The huge cavity was thoroughly cleared out by irrigation and the cerebellum allowed to drop back in its position. Closure was performed by Dr. Horrax.

(Dr. Cushing)

Pathology Report
November 8, 1929
Dr. Eisenhardt

Examination showed numerous cholesterin crystals, the edges of which were quite distinctly outlined.

Impression: Cholesteatoma.

November 9, 1929
Dr. Oldberg

Condition excellent. Left facial somewhat more pronounced than preoperatively.

Dysphagia slightly more pronounced.

Lumbar puncture by Dr. Cushing. 33 cc of bloody fluid removed under slightly increased pressure.

November 12, 1929
Dr. Oldberg

Seen by Dr. Cushing. Cast removed.

November 26, 1929
Dr. Oldberg

Walking about with assistance. Her ataxia seems improved. Speaks more distinctly.

Discharge Note
November 30, 1929
Dr. Oldberg

Patient today discharged home. Her dysarthria and dysphagia are much improved as is the ataxia. Has gained much strength and will shortly be able to perform domestic duties. To wear goggles and have basic care for anesthesia I consider

Follow Up Note
March 11, 1930
Dr. Cushing

The patient’s husband reports that she continues to have pain and needles feeling in the lower jaw and a little pain shooting up on this side. There is continued buzzing in the ear. Continues to wear a shield. The eye remains clear but there is an internal squint. She is able to walk up and down stairs alone by holding on to the rail. She sleeps soundly twelve hours a night and eats well. Still has some difficulty in swallowing. On the whole, I think her condition is as good as could be expected.

The last letter from the patient’s husband was received on October 9, 1931, with the report of her death after a fall. No autopsy was done.

Discussion

Advances in the field of neurosurgery at the time of Harvey Cushing are documented for treatment of pituitary tumors, anterior skull base lesions, meningiomas, and acoustic neuromas.4,5 Cushing’s contributions to the management of skull base tumors have rarely been reported.6 His surgical treatment of a cholesteatoma extending into the skull base is an example of his meticulous documentation and his accelerated surgical techniques. Cushing’s surgical approach to the cranial vault lesions was previously presented through five cases including a malignant tumor of basal meninges, a case of basal cell carcinoma, and three cases of osteoma.6 This case
demonstrates the evolution of neurosurgical techniques during a staged approach to the skull base before the development of modern skull base surgery.

The complexity of this case and its management by Cushing, despite the absence of modern imaging, reveals Cushing’s deep knowledge of localization. Despite the importance of technical expertise, the intellectual aspect of surgery was a priority for Cushing. As he once said, “I would like to see the day when somebody would be appointed surgeon somewhere who had no hand, for the operative part is the least part of the work.”

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Notes
Harvey Cushing’s original patient’s hospital records included in this article were in part published in a book coedited by the senior author: Cohen-Gadol AA, Spencer DD, eds. The Legacy of Harvey Cushing—Profiles of Patient Care. New York, NY: Thieme/American Association of Neurosurgeons; 2007. All other contents are original and have not been previously published elsewhere. The copyright for the patient records is retained by the Cushing’s Center at Yale University Department of Neurosurgery. The senior author has been given permission to use the images and patient records.

Conflicts of Interest
The authors have nothing to disclose.

References
6 Pendleton C, Raza SM, Gallia GL, Quiñones-Hinojosa A. Harvey Cushing’s approaches to tumors in his early career: from the skull base to the cranial vault. Skull Base 2011;21(4):271–276