CAROTID-CAVERNOUS FISTULA: A PERPLEXING SURGICAL PROBLEM*

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Although there are several operations for correction of carotid-cavernous fistulas, both the medical literature and personal experience indicate that no method can be relied upon to bring about a cure. Moreover, there seems to be no way to determine which patient may be in the group of approximately 10 per cent in which spontaneous obliteration of the fistula will be obtained. Although we are pessimistic regarding the possibility of obtaining complete cure when confronted by a patient with pulsating exophthalmos, it seems worth while to record our experience with 14 patients with carotid-cavernous fistulas seen between 1941 and 1958 (Table 1), and to inquire into the feasibility of attempting to induce thrombosis of the fistula by lowering the systemic blood pressure by hypothermia and by lowering the cerebral blood pressure further during hypothermia by digital occlusion of the carotid arteries in the neck.

REVIEW OF LITERATURE

Various methods of treatment have been employed with success or failure by various writers1-4,5,7,10,14-16,18,20,21,23 and the following are interesting to review. Thrombosis of the carotid-cavernous fistula has occasionally been created by intermittent digital occlusion of the common carotid artery in the neck. Both successes and failures have been reported after (a) ligation of the common carotid artery, (b) ligation of the internal carotid artery, (c) ligation of both the internal and external carotid arteries, (d) ligation of the common and internal carotid arteries, (e) ligation of the common and external carotid arteries, (f) ligation of the common and external carotid arteries and the branches of the external carotid artery, (g) ligation of the internal carotid artery intracranially and extracranially with or without intracranial occlusion of the ophthalmic artery (the trapping operation), (h) the trapping operation plus ligation of the contralateral internal carotid artery, and (i) excision of the ophthalmic veins in the orbit. Hamby and Gardner9 obtained only temporary success with implantation of muscle in the internal carotid artery to block the fistula. Gurdjian8 reported complete cure by using a probe to pack a strip of muscle, 20 cm. long and 0.25 cm. in diameter, into the internal carotid artery. Jaeger11 reported cures in 2 patients treated by plac-

* Presented at the meeting of the Harvey Cushing Society, April 21, 1958, in Washington, D.C.
### TABLE 1

*Results of treatment of 14 patients with carotid-cavernous fistula*

<table>
<thead>
<tr>
<th>Case</th>
<th>Age</th>
<th>Admitted Mo./Yr.</th>
<th>Trauma</th>
<th>Duration</th>
<th>Treatment</th>
<th>Result</th>
<th>Follow-up</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>35</td>
<td>8/41</td>
<td>Yes</td>
<td>7 yrs.</td>
<td>Trapping</td>
<td>Improved</td>
<td>6 mos.</td>
</tr>
<tr>
<td>2</td>
<td>36</td>
<td>3/44</td>
<td>Yes</td>
<td>6 yrs.</td>
<td>Ligation of internal and external carotid</td>
<td>Improved</td>
<td>6 mos.</td>
</tr>
<tr>
<td>3</td>
<td>42</td>
<td>6/44</td>
<td>Yes</td>
<td>60 days</td>
<td>Repeated digital occlusion of carotid</td>
<td>Cured</td>
<td>13 yrs.</td>
</tr>
<tr>
<td>4</td>
<td>42</td>
<td>12/45</td>
<td>No</td>
<td>8 yrs.</td>
<td>Ligation of internal carotid</td>
<td>Cured</td>
<td>12 yrs. Died, heart disease</td>
</tr>
<tr>
<td>5</td>
<td>46</td>
<td>2/46</td>
<td>No</td>
<td>6 days</td>
<td>Trapping</td>
<td>Cured</td>
<td>12 yrs.</td>
</tr>
<tr>
<td>6</td>
<td>23</td>
<td>11/46</td>
<td>Yes</td>
<td>80 days</td>
<td>Ligation of common carotid elsewhere failed. Trapping 14 mos. later in 1 stage by first clipping internal carotid intracranially followed by ligation in neck</td>
<td>Improved</td>
<td>12 mos.</td>
</tr>
<tr>
<td>7</td>
<td>59</td>
<td>3/50</td>
<td>No</td>
<td>54 days</td>
<td>Ligation of common carotid and sympathectomy</td>
<td>Cured</td>
<td>3 yrs.</td>
</tr>
<tr>
<td>8</td>
<td>49</td>
<td>3/51</td>
<td>Yes</td>
<td>7 yrs.</td>
<td>None</td>
<td>Died</td>
<td>Necropsy: rupture cavernous sinus with massive intracranial hemorrhage</td>
</tr>
<tr>
<td>9</td>
<td>31</td>
<td>11/55</td>
<td>Yes</td>
<td>4 mos.</td>
<td>Ligation of internal and common carotid</td>
<td>Cured</td>
<td>12 mos.</td>
</tr>
<tr>
<td>10</td>
<td>25</td>
<td>1/56</td>
<td>Yes</td>
<td>6 mos.</td>
<td>Trapping, intracranial ligation of ophthalmic artery, ligation of orbital vessels, hypothermia</td>
<td>Improved</td>
<td>14 mos.</td>
</tr>
<tr>
<td>11</td>
<td>18</td>
<td>9/56</td>
<td>Yes</td>
<td>50 days</td>
<td>Trapping</td>
<td>Cured</td>
<td>13 mos.</td>
</tr>
<tr>
<td>12</td>
<td>20</td>
<td>10/56</td>
<td>Yes</td>
<td>66 days</td>
<td>None</td>
<td>Died</td>
<td>Necropsy: rupture cavernous sinus with massive intracranial hemorrhage</td>
</tr>
<tr>
<td>13</td>
<td>38</td>
<td>11/56</td>
<td>No</td>
<td>9 mos.</td>
<td>Ligation of common carotid</td>
<td>Improved</td>
<td>17 mos.</td>
</tr>
<tr>
<td>14</td>
<td>32</td>
<td>6/57</td>
<td>Yes</td>
<td>38 days</td>
<td>Trapping plus intracranial ligation of ophthalmic artery</td>
<td>Cured</td>
<td>8 mos.</td>
</tr>
</tbody>
</table>
ing muscle in the internal carotid artery in the neck after preliminary intracranial ligation of the carotid artery. A silver clip attached to the muscle was demonstrated to be at the site of the fistula in one of the cases. The muscle was propelled to the fistula by retrograde blood flow from the external carotid artery assisted by a small ureteral catheter. Browder\(^3\) successfully implanted muscle into the cavernous sinus. Arterial plugging has been attempted by injection of paraffin into the carotid artery in the hope that it would obliterate the arteriovenous fistula.\(^6\) Parsons and associates\(^17\) reported a spontaneous cure after angiography, which was attributed to the irritating vasoconstricting effect of the dye. Singleton\(^22\) injected a 50 per cent solution of glucose and later sodium morrhuate into the dilated supraorbital vein; this caused a severe inflammatory reaction but resulted in a cure. Potter\(^19\) reported a case of spontaneous cure after a period of severe hemorrhage and syncope. He suggested that hypotension might be beneficial in causing spontaneous thrombosis.

RESULTS OF TREATMENT

There is a false impression among the medical profession that the rate of cure after ligation of one or more of the arteries in the neck for carotid-cavernous fistula is high. The fact is that the rate of cure with this type of operation is less than 50 per cent. Only 27 per cent of our patients were cured by ligation of cervical vessels. There is another false impression that the trapping operation cures almost all patients. Only 3 of our 6 patients having trapping operations were completely cured.

SUGGESTED PLAN OF TREATMENT

After the neurologic status has been determined, an electroencephalogram and bilateral arteriograms should be made. Digital occlusion of the carotid artery may be tried for a few days or weeks in the hope of producing thrombosis of the fistula. However, we believe that application of a removable clamp to the ipsilateral internal carotid artery should not be delayed by a trial of digital occlusion if the vision in the protruding eye is in jeopardy. If occlusion of the internal carotid artery does not result in a cure, the trapping operation with intracranial clipping of the carotid artery, as well as intracranial occlusion of the ophthalmic artery, if feasible, should be performed. Since there is abundant collateral circulation through the twelve branches of the ophthalmic artery, intracranial ligation of this artery is unlikely to cause blindness. Some difficulty may be encountered in exposing the ophthalmic artery, since it usually leaves the internal carotid artery beneath the optic nerve. However, the optic foramen can be unroofed sufficiently for gentle retraction of the optic nerve medially in order to clip the underlying artery. If the ophthalmic artery originates well within the cavernous sinus, the orbital roof may be opened and the artery may be clipped within the orbit. If trapping, with or without ligation of the ophthalmic artery, fails to provide a cure, muscle should be packed into the carotid
artery distal to the ligature in the neck (we have had no experience with this).

**HYPOTHERMIA AS A FORM OF TREATMENT**

In the occasional case in which the fistula persists, hypothermia can be induced in the hope of producing thrombosis. It is even possible that hypothermia could eliminate the need for intracranial operation in some patients. With hypothermia of 86°F (30°C) it has been shown that in a healthy person of middle age or younger, both common carotid and vertebral arteries can be occluded for 6 minutes with relative safety. The period of safety, however, is variable from person to person, and the best determinant is the electroencephalogram, which may dictate shortening or allow prolongation of the estimated safe period of occlusion by the appearance of delta activity. Lougheed and associates simultaneously occluded both carotid and vertebral arteries for 14 minutes and 25 seconds in a patient without adverse results while using hypothermia with a rectal temperature of 75°F to 78°F.

In one patient, who was not cured by the trapping operation and ligation of the ophthalmic artery, we tried hypothermia unsuccessfully but the experience justifies comment since we found no reports in the literature of utilization of this method. In this case (Case 10) the patient became severely hypotensive when the body temperature reached 92°F. Body cooling was discontinued at 90°F but the temperature drifted down to 85.5°F. Blood pressure and peripheral pulsations were unattainable for 45 minutes. During this period of hypotension the systolic orbital bruit could not be heard. At no time during the procedure was delta activity recorded in the electroencephalogram. The patient was given 50 mg of ephedrine with resultant elevation of the systolic blood pressure to 10 mm Hg above the normal preoperative range without return of the orbital bruit. Consequently, it was assumed that thrombosis of the fistula had occurred and, for this reason, digital occlusion of the contralateral carotid artery in the neck was not done. However, 1 hour after the blood pressure and body temperature returned to normal the systolic bruit could be heard, but it was much lower than before the procedure and the patient, who could hear it before hypothermia was induced, was unable to hear it afterwards. Would complete cure have been obtained if prolonged digital occlusion of the carotid artery had been performed as planned? This patient has agreed to return for another trial of hypothermia with digital occlusion of the carotid artery.

**MECHANISM OF CURE**

The process by which carotid-cavernous fistulas are cured is not clearly understood but in 2 of our patients (Cases 3 and 6) cure was obviously the result of thrombosis of the cavernous sinus. One patient (Case 3) treated himself with digital occlusion for several months when he had sudden pronounced increase in the proptosis with simultaneous disappearance of the bruit. He was treated at a hospital with pressure dressings and ice on the eye.
for several days before the eye returned to normal. The other patient (Case 6) had an alarming but temporary increase in the proptosis several days after the trapping operation. In the other patients cured without increase in proptosis it must be assumed that blockage of the fistula occurs by some such method as thrombosis of the cavernous portion of the carotid artery.

SUMMARY

Carotid-cavernous fistula is a perplexing therapeutic problem characterized by both successes and failures, as illustrated by the 14 personal cases presented in this paper as well as the reports in the literature. Two of our 14 patients died from rupture of the cavernous sinus before they could be treated. One patient was cured merely by repeated digital occlusion of the carotid artery. Ligation of the internal carotid artery in the neck cured another. One was cured by ligation of the common carotid artery alone and another was improved. One was cured by ligation of the internal carotid artery followed by ligation of the common carotid artery. One patient was cured by ligation of the internal carotid artery followed by ligation of the external carotid artery. Two patients were cured by the trapping procedure and one was improved. The trapping procedure was done in another patient by ligation of the intracranial segment of the internal carotid artery immediately before ligation of the carotid artery in the neck to keep the fistula from robbing the ipsilateral hemisphere of blood. Trapping plus ligation of the ophthalmic artery was performed on 2 patients with cure in one and improvement in the other. The latter patient was treated further with hypothermia with additional improvement.

A plan of management has been presented which we believe will be successful in the majority of cases. Initially, repeated digital occlusion of the carotid artery is tried for a few days or weeks in patients with early nonfulminating lesions. However, if the lesion is fulminating, we advocate occlusion of the internal carotid artery in the neck with a clamp that can be opened if necessary. If this procedure does not result in cure, the ophthalmic artery, if feasible, and the carotid artery should be clipped intracranially in one stage. In the remaining few patients whose fistulas persist, implantation of muscle into the carotid in the neck to block the fistula may solve the problem, but we have not done this. Finally, we believe that hypothermia under electroencephalographic control with bilateral digital occlusion of the carotid artery shows promise of creating a cure and needs further trial.

REFERENCES

6. DAWBARN, R. H. M. Cited by Martin and Mabon.13

DISCUSSION*

DR. WALLACE B. HAMBY: Unfortunately Dr. Mount’s was the only paper I had the opportunity of studying before presentation, so this discussion is somewhat extemporaneous. Multiple aneurysms and their management have interested us and perhaps have been one of the factors responsible for our preference for intracranial attack in the treatment of aneurysms. In reported series their incidence varies from 5 to 30 per cent. Angiograms have revealed multiple aneurysms in only 9 per cent of our patients. In our autopsy series, in which the vessels were subjected to better scrutiny, 25 per cent of patients dying of aneurysms have had multiple lesions. It is likely that with more refinements of angiography a higher percentage of multiple lesions will be found.

When an aneurysm is accessible and its neck permits, its simple occlusion by clip or ligature appears indicated. In many instances, however, this is technically difficult or impossible. It is for such cases that methods such as the one reported by Drs. Selverstone and Ronis are so valuable.

* In addition to the preceding papers by Dr. Mount, and by Drs. Echols and Jackson, two other papers relating to the subject were presented at this meeting: “Multiple Intracranial Aneurysms” by J. L. Poppen and C. A. Fager (see J. Neurosurg., 1959, 16: 581–589) and “Adherent Plastics for the Reinforcement of Aneurysms,” by B. Selverstone and N. Ronis.
In reference to the "loosening" of clips on cervical vessels and the apparent loosening of ligatures, being visualized later as surrounding only the intima, one is reminded of the work of Jones in the 18th century. He found that when vessels are constricted, their external and medial tunics atrophy, only the intima persisting. The loss of this tissue leaves relatively more room between intima and ligature so that the previously closed vessel again becomes patent. This also suggests the necessity of Dr. Selverstone's group determining the ultimate effect of these resins on the wall of the vessel.

The problems reported by Dr. Echols concerning carotid-cavernous fistulas are, of course, common to arteriovenous malformations in other areas as well. It might be advantageous to explore more systematically the possibilities of the method devised by Gardner of plugging the fistula by putting muscle into the vessel without interrupting the blood stream.

DR. E. H. BOTTERELL: After these distinguished essays, and Dr. Hamby's discussion, I shall confine my remarks to a brief mention of our experience with the Selverstone clamp, and with carotid-cavernous fistulae.

We've had all of the difficulties that Dr. Echols and his coworkers report regarding arteriovenous fistulae, including 2 patients who went blind after a simple carotid trapping procedure, without taking the ophthalmic artery. I, myself, believe that if I were going to trap a carotid-cavernous fistula I should, if at all possible, take the ophthalmic artery as well, so the circulation to the eye is not undermined by the persisting fistula fed only by the ophthalmic artery. This, of course, has been described by Dr. Gardner, and Dr. Hamby, and Dr. Adson did 3 such procedures many years ago.

[Slide] These are the 7 cases in which we used the Selverstone clamp in aneurysms over a period of years, and they were almost all large aneurysms. This patient had a large aneurysm which bled recurrently. She has remained well for 5 years, and her follow-up angiogram shows the cavity of the aneurysm to be about half its former size.

[Slide] This patient was operated on because blindness developed 16 years after a subarachnoid hemorrhage; the large aneurysm measured 3 by 3 cm. Her blind eye, of course, remained blind; the scotoma in the other eye has not enlarged in 2 years.

There are 3 cases of particular interest, I think. The first patient had multiple cranial nerve palsies and facial pain. The pain was uninfluenced by ligation of the common carotid artery, and was relieved by an alcohol injection by my colleague, Dr. Thomas Morley, of the gasserian ganglion. We don’t know why she died 3 years later. The second patient had a large aneurysm, and had a fatal recurrent hemorrhage 2 years following application of a Selverstone clamp. The clamp was proved to be shut at the autopsy. The third patient suffered all of the complications of an infection developing around the Selverstone clamp. We set out to remove the clamp and ended up with the vascular surgeons helping us put a graft between the innominate artery and the subclavian. Dr. Mount mentioned that 3 of his Selverstone clamps were extruded through the incision in the neck.

DR. ERNEST SACHS, JR. With regard to Drs. Poppen and Fager's excellent paper, I certainly agree completely about the matter of complete arteriography, and would like only to add the point that in the absence of demonstrating an aneurysm on bilateral carotid angiography, vertebral angiography be included, even in those patients with supratentorial signs in whom you think a vertebral aneurysm could not possibly exist. We've had such an experience in 2 cases.

My experience, in a much smaller series than Dr. Echols’, with four carotid-cavernous fistulas has been roughly similar.

I presume the reason I have been included on this panel is because of some experience with plastics, which Dr. Selverstone kindly gave me for the use of reinforcing aneurysms. I have no human cases as yet, but in our experience with animals, we certainly think that this is a useful tool which may well be applicable to these aneurysms in many situations—the notorious ones on the anterior communicating artery and at the bifurcation of the middle cerebral artery on the dominant side. Particularly, as Dr. Fager mentioned, in cases of mul-
tiple aneurysms the plastic reinforcement of the aneurysms, as Dr. Selverstone showed you, will resist the very high intravascular pressures without rupture in aneurysms that have ruptured previously.

In response to what I think Dr. Hamby said about pathology of the vessels, these plastics are resilient as well as strong. They do not reinforce the vessel as though by a metal clamp, such as the "Selverstone" clamp in the neck, but there is resiliency as well as strength.

Dr. Rudolph Jaeger: I should like to discuss Dr. Echols' interesting paper on this most devastating disorder. I have had under immediate observation 18 cases. In reviewing them, I find that the treatment has fallen readily into three groups:

Group 1 (8 cases)—Carotid ligation in the neck: Three of these patients died following this procedure: two several years later from intracranial hemorrhage and 1 immediately from ischemia of the brain. Three improved markedly but the bruit and other symptoms continued to some extent. One patient showed no improvement and lost vision in the eye on the side of the lesion. One patient could not tolerate even gradual occlusion. To sum up these 8 cases, only 3 patients were improved and results in the other 5 were unsatisfactory.

Group 2 (5 cases)—Ligation of intracranial carotid and ligation of carotid in the neck: None of these patients was cured although several seemed to be somewhat improved temporarily. Gradually their symptoms became progressively worse.

Group 3 (6 cases)—Ligation of intracranial carotid followed by ligation of internal carotid in the neck, after forcing a muscle plug into the fistulous opening: All of these patients were cured completely without complications. The procedure is carried out by first clipping the carotid intracranially with Olivecrona or Jaeger type of removable clip. However, these clips were not used in the 6 cases reported here. This prevents cerebral ischemia when the carotid is ligated later in the neck. Ligation of the carotid in the neck is preceded by opening this vessel and packing the fistulous opening with one or several emboli of muscle. The first patient operated on by this procedure was in 1942 after intracranial clipping and ligation of common carotid had failed. This patient was observed during and at the end of a 7-year period and was completely well, and I have reason to believe remains so to the present date. This case was reported and the method was described before the Southern Surgical Association and documented in the Southern Surgeon, 1949, 15: 207–209. The only solution to this problem as I see it is to close the fistulous opening itself as mentioned, between the carotid and the cavernous sinus, as Dr. Echols has pointed out. All other methods have not been completely curative and many grave immediate and late sequelae have occurred. The safety and efficacy of the fistulous occlusion method has been demonstrated in the 6 cases of this third group. The three slides here demonstrate the underlying principle of vascular abnormalities associated with carotid-cavernous fistula and also a principle of specific cure by occluding the carotid-cavernous opening. The first slide presents the anatomy involved in carotid-cavernous communication. The second demonstrates the surgical maneuver of this method as it pertains to the carotid in the neck. The third slide shows two intracranial silver clips, one on the carotid and the other which identifies the muscle embolus where it is plugging the fistula.

Dr. George J. Hayes: The need to reinforce a vessel from which an aneurysm has developed was brought to our attention by the fate of one of our patients. This woman had an aneurysm arising from the internal carotid artery, with a long, narrow stalk. The stalk was clipped adjacent to the internal carotid, then cut, and the aneurysm was removed. Six years later, this woman had a subarachnoid hemorrhage, died, and at autopsy was found to have another aneurysm at the site of the original one. This is not surprising if we remember the basic defect in the vessel wall.

Following Dutton's suggestion regarding reinforcement of aneurysms with plastic material, we had the Caulk Company modify cranioplastic methacrylate to solidify more quickly. In the last year, we have had 8 occasions to use this material. Four patients had aneurysms of the middle cerebral artery at its major branching. The middle cerebral, the
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aneurysm, and the branches were all invested with plastic. Three of the patients had internal carotid aneurysms, the necks of which were clipped; then the internal carotid, the neck of the aneurysm, and the applied clip were invested with plastic. One anterior communicating aneurysm was invested with plastic. So far, these patients have all done well, but the period of follow-up at the longest is only 11½ months.

**Dr. Harry Steelman:** I should like to correct an error, which crept into print concerning Dr. Poppen's original article on treatment of aneurysms. About 4 years ago, Colonel Hayes, Dr. Hugo Rizzoli and I reported a series of aneurysms at Walter Reed Army Hospital, and in a review of the literature, through a typographical error, it was stated that 6 of Dr. Poppen's patients had died from rupture of the aneurysm after ligation of the carotid artery in the neck. Actually that is in error, and according to their slides, I believe that they have had only 2 patients who had this distressing complication. However, considering the number of patients they have treated, it is a quite respectable figure.

**Dr. J. Garber Galbraith:** I just want to make one comment about the paper of Dr. Echols and Dr. Jackson. And that is in regard to the sequence in which this trapping is done. We have been impressed with the fact that when a person does not tolerate carotid occlusion in the neck, or when carotid occlusion in the neck does not completely relieve the symptoms, it is important to do the intracranial part of the procedure first. That is, to occlude the carotid artery intracranially and the ophthalmic artery intracranially before occluding the carotid in the neck. In this way the brain is protected against back-bleeding into the fistula. In carrying it out in this sequence during the past 6 years, we have encountered 8 of these cases with no untoward results in any, by so doing, and with apparent cure in these cases to the present time.

**Dr. L. A. Mount:** I was hoping to have 100 cases to be reported when I presented this material to you. However, in the past 4 years we have done so few ligations that I've given up all ideas of having 100 cases in the near future. It has always been my impression that these aneurysms are best treated by a direct intracranial approach when there is a reasonable chance of success. I believe that the advent of hypothermia has added greatly to our chances of success by the intracranial approach.

I would like to mention that the method of using the Selverstone clamp that I have described is not that used by Dr. Selverstone. He feels that a gradual occlusion is the way to do it, and I'm sure there is some merit in what he has to say.

I would like to emphasize the need for progress or postoperative arteriography in all patients whom we have treated surgically for intracranial aneurysms, in order to see just what we have accomplished and not to fool ourselves. Even the direct intracranial approach is not a foolproof method, and we can find from doing our progress arteriogram that we have not accomplished everything we think we have.