COMBINED SUPRA- AND INFRACLINOID ANEURYSMS OF INTERNAL CAROTID ARTERY

REPORT OF A CASE OF UNUSUAL CONGENITAL DILATATION OF INTRACRANIAL PORTION OF CAROTID ARTERY AND INJURIES TO VISUAL, OCULOMOTOR, SENSORY, AND TASTE FIBRES

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(Received for publication March 25, 1955)

Following Sir Geoffrey Jefferson’s classic description in 1938\textsuperscript{12} of infraclinoid aneurysms of the carotid syphon it has been possible to make an exact localization of their position within the cavernous sinus by the neurological signs. Jefferson divided these lesions into three main groups. If the aneurysm is situated in the most rostral portion of the sinus, only the ophthalmic division of the trigeminus is compressed, with resulting pain and more or less hypaesthesia in the forehead and eye. When the aneurysm is posterior, the maxillary nerve is likely to be involved in addition, and when still further caudal or when it fills the entire sinus, there is sensory loss over the mandibular area and often paralysis of the motor root as well. The oculomotor nerve is nearly always involved, with resulting ptosis and pupillary dilatation. Regardless of the anteroposterior position of the aneurysm, there may be a paralysis of the trochlear and abducent nerves and a complete external and internal ophthalmoplegia. Vision and taste are not ordinarily affected by infraclinoid aneurysms, but are possible complications when the aneurysm bulges upward to press upon the optic nerve or sufficiently far back in the middle fossa to compress the greater superficial petrosal ramus of the facial nerve on the petrous ridge. This latter possibility has not been mentioned heretofore.

Upward expansion of an aneurysm in the cavernous sinus to the level of the anterior clinoid, where it may cause added erosion or tilting of this structure with compression of the optic nerve, has not often been described. The dense layer of dura mater that covers the sinus tends to confine aneurysmal expansions either to the infra- or the supraclinoid portion of the carotid trunk. Jefferson\textsuperscript{11,12,13} and Dandy\textsuperscript{6} have reviewed the world literature and recorded 25 examples of this combined syndrome, which are listed in Table 1. Four of these were in Jefferson’s personal series and 3 in Dandy’s (one other published with Heuer\textsuperscript{9}). The blindness has usually been of the monocular type. Occasionally there has been impingement on the crossing fibres from the inferior nasal portion of the retina of the opposite eye. These curved forward into the optic nerve for a short distance in front of the chiasm as they decussate. When compressed there is loss of the upper temporal
field of vision in the contralateral eye, as described by Traquair.20 One example of this defect is included in Table 1 (Jefferson, 1955, Case 2). In addition there may be a central scotoma (Dandy, 1944, Case 1; Jefferson, 1955, Case 3) or damage to the optic tract with hemianopsia (McKinney et al., 1936; Dandy, 1944, Case 1), but the aneurysm is not likely to extend this far caudally. Papilloedema has also been reported (Blessig, 1877; McKinney et al., 1936).

Fatal hemorrhage has been rare, because of the dense dural covering over the cavernous sinus, but one such complication has been recorded by Beadles (Nettleship’s case, in which exsanguination took place through the nose). Aneurysms in the carotid syphon may rupture into the cavernous sinus, giving rise to an arteriovenous communication. As this type of lesion causes visual damage by another mechanism, we have not included any cases of this sort.

Most of the aneurysms giving rise to the combined visual, oculomotor, and sensory syndrome were studied before the days of angiography. Only 17 have been operated on and but 8 of these were exposed intracranially. Jefferson’s first patient, whose lesion was verified by intracranial exploration, developed a hemiparesis and died a month later. Postmortem examination revealed a huge aneurysm, almost completely thrombosed, which filled the middle fossa and compressed the temporal lobe. Another patient was submitted to carotid ligation in the neck with satisfactory improvement, but the 2 others were not operated upon. Dandy carried out combined cervical and intracranial occlusion of the internal carotid in 2 patients, with little improvement in the one whose clinical result is mentioned. He performed a subtemporal decompression in his third patient and in another reported with Heuer.9 Three other aneurysms were exposed at intracranial operation by Reinhardt,17 Zollinger and Cutler,23 and Gardner.7 The first 2 patients died, but Gardner’s survived despite gross bleeding on opening the aneurysm, which was controlled by packing and cervical ligation.

In view of the rarity of large infraclinoid aneurysms that bulge upward out of the cavernous sinus to involve the optic nerve, and of the lack of surgical experience in dealing with them, it seems worth while to report the following case, especially as it demonstrates the added neurological sign of loss of taste, which has not been observed heretofore.

CASE REPORT

MGH U-857750. Chester P., aged 16 years, entered the hospital in June 1954. Otherwise in good health, he had noticed a gradual loss of vision in his left eye over the preceding year. One week prior to admission a severe pounding left temporal headache developed which prevented sleep. The next day, as the headache subsided somewhat, a sense of numbness spread over the entire left trigeminal area, the left eyelid drooped, and all movements of this eye were lost. There were also further reduction in vision of the left eye and loss of taste on the left side of his tongue. He did not become nauseated, vomit or have a stiff neck.

Examination. Blood pressure was 120/62; temperature was normal. There were two small naevi on the left upper lip and temple. The left eye seemed slightly more
### TABLE 1

Jefferson's and Dandy's summaries of infraclinoid aneurysms that extended beyond cavernous sinus and thereby caused damage to optic nerve.*

<table>
<thead>
<tr>
<th>Author</th>
<th>Date</th>
<th>Sex</th>
<th>Age</th>
<th>Visual Defect</th>
<th>Other Clinical Findings</th>
<th>Site of Aneurysm</th>
<th>Operation</th>
<th>Result</th>
</tr>
</thead>
</table>
| Holmes          | 1901 | M   | 16  | L. eye blind               | L., supraorbital pain & numbness, Paralysis III, IV & VI | L. cavernous sinus
|                 |      |     |     |                           |                              | Autopsy          | 0               | Died            |
| Adams           | 1909 | M   | 56  | R. eye blind               | Neuralgia V, I & II, Paralysis III, IV & VI | R. cavernous sinus
|                 |      |     |     |                           |                              | Autopsy          | 0               | Died            |
| Blessing        | 1877 | M   |     | L. eye blind, High-grade choke | Exophthalmos, L., III palsy | Internal carotid
|                 |      |     |     |                           |                              | Carotid ligation | Died 35th day    |                 |
| Caermbak        | 1902 | F   | 59  | Blind L. eye, recent       | VI palsy 6 yrs, Neuralgia brow & face 4 yrs. Ptosis L. recently | Aneurysm L. internal carotid, Autopsy | 0               | Died            |
| Beadles         | 1907 | M   |     | L. amblyopia 18 yrs. L. optic atrophy | 18 yr. history facial pain & ptosis. L. ophthalmoplegia | L. internal carotid
|                 |      |     |     |                           |                              | in cavernous sinus communicating with nasopharynx | 0               | Died from nasal haemorrhages |
| Reinhardt       | 1913 | M   |     | Blind L. eye               | L. III palsy, complete, Pain L. eye & forehead | Large aneurysm
|                 |      |     |     |                           |                              | middle fossa at autopy | 2               | Died 7th day     |                 |
| Heuer & Dandy   | 1916 | M   |     | L. eye blind               | Paralysis V, III, IV & VI | L. cavernous sinus
|                 |      |     |     |                           |                              | Autopsy          | 0               | Died            |
| Greedel         | 1921 | M   |     | L. optic atrophy 8 yrs. L. eye blind | L. ophthalmoplegia; slight exophthalmos. X-ray: dilated sella | L. internal carotid
|                 |      |     |     |                           |                              | size of child's fist | Transphenoidal op. | Death from haemorrhage |
|                 |      |     |     |                           |                              | Autopsy          | 0               | Died            |
| Brunetti        | 1931 | F   | 63  | Failing vision, R.         | R. ptosis & partial ophthalmoplegia, Pain in eye & forehead | X-ray diag.: calcification region R. internal carotid & ophthalmic arteries | 0               | Recovery        |
|                 |      |     |     |                           |                              | size of hen's egg R. middle fossa compressing optic nerve, Autopsy | 0               | Dead            |
| McKinney, Acree & Soltz | 1936 | M   |     | Slight pallor L. disc. R. normal, Temporal constriction L. visual field | L. ptosis & ophthalmoplegia | X-ray diag.: calcification in aneurysm | 0               | Recovery, Pain improved with carotid compression |
| McKinney, Acree & Soltz | 1936 | F   |     | Marked pallor L. disc. blind L. eye 2 yrs. | Ophthalmoplegia, partial L. hypoesthesia V. I | Unverified. Suggestive X-ray | 0               | Recovery, Sympathetic improvement |
| McKinney, Acree & Soltz | 1936 | F   |     | R. papilloedema; L. normal, Blind R. eye | Ophthalmoplegia, R. Hypoesthesia V. 1 & 3 | X-ray diag.: calcification in wall of aneurysm | 0               | Recovery, Unimproved |
prominent and he could open the lids only a few millimeters. The pupil was dilated to 7 mm. Both direct and consensual reflexes to light were lost. Paralysis of all the ocular movements was complete. Sensation was absent over all three trigeminal divisions with added paralysis of the motor root on this side. Examination of the eyes by Dr. David G. Cogan revealed a visual acuity of 5/200 on the left, 20/30 on the right. The disc on the left showed no papilloedema, but a severe degree of optic atrophy. The visual field on the right was normal in extent, but on the left there was a suggestion of an eccentric scotoma. The left eye, which was very slightly propsected, could easily be pressed back without resistance and there was no conjunctival engorgement. In addition to the left-sided visual loss, which soon progressed to nearly total blindness, and the paralysis of the 3rd, 4th, 5th, and 6th cranial nerves, the boy had lost sense of taste over the anterior portion of his tongue on the involved side. There was no impairment of facial movement or of the remaining cranial nerves. There were no other neurological abnormalities.

Other studies of interest included lumbar puncture with the initial pressure 100, and clear colourless fluid containing 1 lymphocyte and a protein of 30 mg. Roentgenograms of the skull revealed a striking thinning and upward displacement of the left anterior clinoid, but no other evidence of bony erosion or calcification. The optic foramina on the two sides were of normal diameter, as were the superior orbital fissures and the foramina rotunda and ovale. A left percutaneous arteriogram demon-

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**TABLE 1. Continued**

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<th>Author</th>
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<th>Site of Aneurysm</th>
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<th>Result</th>
</tr>
</thead>
<tbody>
<tr>
<td>de Vet &amp; Zeckel</td>
<td>1937</td>
<td>M</td>
<td>10</td>
<td>Blind L. eye; sight depressed on R.</td>
<td>Complete ophthalmoplegia &amp; aneurysm V. 1, 1. Wassermann +</td>
<td>Angiographic verification</td>
<td>Transfrontal exploration, Ligation internal carotid</td>
<td>Recovery</td>
</tr>
<tr>
<td>Jefferson</td>
<td>1937</td>
<td>F</td>
<td>60</td>
<td>L. eye blind</td>
<td>Anaesthesia V &amp; ophthalmoplegia</td>
<td>Unverified</td>
<td>None because of age &amp; refusal to risk surgery</td>
<td>Alive 14 yrs. later; aneurysm had calcified</td>
</tr>
<tr>
<td>Krayschil</td>
<td>1941</td>
<td>M</td>
<td>44</td>
<td>L. eye blind</td>
<td>Paralysis V, 1 &amp; 2, III, IV &amp; VI</td>
<td>L. cavernous sinus</td>
<td>Carotid ligation</td>
<td>Palsies clearing at 3 wks.</td>
</tr>
<tr>
<td>Dandy</td>
<td>1944</td>
<td>F</td>
<td>52</td>
<td>R. eye 20/50 with central scotoma</td>
<td>Partial V. Complete paralysis III, IV &amp; VI</td>
<td>R. cavernous sinus</td>
<td>1. Threads passed through aneurysm; 2. Partial carotid occlusion. 3. Cervical &amp; intracranial occlusion</td>
<td>Not stated</td>
</tr>
<tr>
<td>Jefferson</td>
<td>1945</td>
<td>F</td>
<td>64</td>
<td>L. eye blind. ? R. upper temporal field loss R. eye</td>
<td>Anaesthesia V &amp; ophthalmoplegia</td>
<td>Arteriogram: aneurysm filling cavernous sinus</td>
<td>None because of age &amp; arterioleisis</td>
<td>Died 2 yrs. later of another cause</td>
</tr>
</tbody>
</table>

* Arteriovenous fistulae are not included.
strated, as expected, a huge aneurysm extending from a dilated foramen lacerum throughout the length of the cavernous sinus, and then upward behind and above the anterior clinoid (Fig. 1). The supraclinoid portion of the carotid was dilated all the way back to its bifurcation at the circle of Willis. There was no suggestion of a sac, but a long fusiform dilatation of the carotid throughout its entire intracranial extent.

1st Operation. Preliminary testing by carotid occlusion was tolerated well. Therefore the carotid bifurcation was exposed under deep cervical plexus block on June 15, 1954. The common carotid artery was tightly compressed by a tantalum clip and the external branch doubly tied with silk ligatures. Unfortunately our electronic manometer was out of order and pressures in the internal carotid distal to the clip could not be ascertained. Nevertheless it was obvious that there was quite a forceful distal pulsation, which persisted even with the addition of a rubber-covered clamp placed temporarily 1 cm. above the tantalum band to make certain that it was effectively closed. Blood was aspirated from the segment of artery between the two clamps, which then remained collapsed. After an hour’s observation without ill effect the incision was closed.

Course. Carotid occlusion was well tolerated, but, as predicted by the retrograde filling and pulsation of the internal carotid, little benefit ensued. Headache, which had been grumbling continuously prior to carotid occlusion, was lessened but not

Fig. 1. Anteroposterior and left lateral arteriograms showing the large subclinoid aneurysm on left side which filled the entire length of the cavernous sinus. It also involved the entire supraclinoid portion of the internal carotid as far distally as the circle of Willis. Even the first portion of the anterior and middle cerebrials seem enlarged. Insert shows thinning and upward displacement of left anterior clinoid.
entirely relieved. There was no improvement in the cranial nerve palsies save a slight return of sensation in the 2nd trigeminal division with a trace in the 3rd. There was also a faint return of contraction in the inferior rectus muscle.

Both the patient and his mother requested further surgery because of persistent headache and failure of improvement. Before supraclinoid ligation was considered a right-sided arteriogram was carried out, which showed no cross filling of the aneurysm and an apparently normal arterial tree on this side. It was also of interest that the patient could tolerate digital occlusion of the right carotid without any apparent ill effect.

2nd Operation. On June 24, 1954, the supraclinoid portion of the left internal carotid was exposed through a small transfrontal bone flap under local and light Pentothal anaesthesia. Spinal fluid was removed by lumbar drainage to ensure a slack brain. The supraclinoid extension of the main aneurysm in the cavernous sinus was then clearly visible. At the level of the eroded and up-tilted anterior clinoid, the internal carotid was dilated to a full centimeter in diameter and had flattened the optic nerve to a thin ribbon, as shown in Fig. 2. This dilatation extended to within a few millimeters of the origin of the anterior and middle cerebral arteries. It was possible to occlude it with a pair of dural clips, but the sac still seemed to pulsate. Finally, with careful manipulation, an aneurysm needle was slipped around

![Fig. 2. Photograph taken at time of craniotomy to show compression and flattening of optic nerve by underlying bulge of supraclinoid portion of internal carotid artery. The two dural clips partially occluding the artery can be seen beneath the right-hand edge of the ribbon retractor.](image-url)
the artery and it was securely ligated. Thereupon the tense sac collapsed. The incision was then closed.

Course. After operation the boy recovered consciousness rapidly with no evidence of aphasia or right-sided weakness. At discharge 3 weeks later, sensation was improving in the two lower trigeminal divisions, and loss of taste had disappeared. He was again able to see large objects with the left eye and the consensual pupillary response to light was recovering. He was still depressed by his drooping lid and reluctant to return to school with this deformity.

Recovery of the levator palpebrae muscle happily began within 2 days of his return home, and the recovery of ocular movements within 2 weeks. Return of abducens innervation to the external rectus was slower than that of the other extraocular muscles, but within 10 weeks lid and eye movements were nearly back to normal. By this time the consensual light reflex was also functioning well, as were the maxillary, mandibular, and motor divisions of the trigeminus. There was still a reduction in sensation over the forehead with persistent corneal anaesthesia and no direct response of the left pupil to light, although he could see large moving objects in the temporal field. The optic nerve head remained pale and atrophic. The sense of taste had recovered completely.

One year after occlusion of the aneurysm this boy is reported to be doing very well at school and without complaint save nearly complete blindness of his left eye.

DISCUSSION

This case is of interest from several points of view. Firstly, it represents an unusual type of aneurysm. It was not a simple saccular or berry-shaped defect arising at the crotch of a bifurcating vessel—in this instance the opthalmic and internal carotid arteries. Instead it was an irregular fusiform expansion which extended for several centimeters, from the foramen lacerum nearly to the end of the carotid artery. In fact, even the horizontal primary portions of the anterior and middle cerebrials appear somewhat enlarged in the anteroposterior arteriogram shown in Fig. 1. The widest part of the expanded vessel was its intracavernous portion. Moreover, this was obviously not an example of the usual fusiform arteriosclerotic aneurysm. We have the impression that these congenital fusiform dilatations, which we have seen on both the carotid and basilar arteries, are special examples of a congenital arterial malformation which probably exists from the time when the vasculature of the brain first develops. In support of this idea is the fact that our patient had also two cutaneous naevi within the field supplied by the left carotid artery. This type of aneurysm may gradually expand, as in our case, in which the supraclinoid portion compressed the optic nerve and the infraclinoid portion the 4th, 5th, and 6th cranial nerves, presumably injuring the greater superficial petrosal branch of the facial as well.

The pathology of this group of aneurysms deserves careful study. It is quite apparent from reading the case reports summarized in Table 1 that the gross pathology has not been adequately described. In addition, few histological reports are available. Even arteriographic evidence is lacking because so many were reported before this procedure came into common use. One cannot determine how many represent the usual berry-type aneurysm or the fusiform arteriosclerotic dilatations. In the end this can be decided only
by determining the exact point of origin of the aneurysm, its form, and also whether the typical defect in the media and internal elastic lamina, which is to be found in most saccular aneurysms, is present.

Secondly, this case report describes an additional operative experience and one in which an unusually good view of the intracranial lesion was obtained. Such opportunities have been rare. Jefferson, in his 4 cases of cavernous sinus aneurysm with supraclinoid extension, had the opportunity to carry out an intracranial exploration in only 1, Dandy in 2 others. In Case 6 of Jefferson’s 1937 paper, the aneurysm was not treated by proximal ligation because this 60-year-old woman did not wish to accept the risk of hemiparesis. Fourteen years later he reports (personal communication) that the aneurysm “has calcified and her visual state remains stationary. She died in 1951 or ’52 of cancer.” In the other patient mentioned in this paper the diagnosis was made at operation and the extent of the huge aneurysm in the middle fossa was thoroughly examined post mortem.

In his 1955 report, concerning the first patient with neurological signs very similar to ours (Case 2), Jefferson stated that “the pains were not very severe, and that fact combined with her age (64) and the presumably arteriosclerotic nature of her aneurysm were deciing factors against ligaturing her carotid.” She was subsequently lost track of, but it is known that she died 2 years later, “from some cause other than aneurysm.” His other patient (Case 3), which resembles the subject of this report save for incomplete blindness (central scotoma) and incomplete trigeminal paralysis, made a very satisfactory recovery after common carotid artery ligation. Jefferson comments that she is “a good example of the shrinkage of even a large aneurysm after proximal ligature of its main vessel.”

Dandy was a strong advocate of “trapping” infraclinoid aneurysms by ligating the carotid proximally in the neck and clipping it intracranially in addition. Generally he did not wait to see if proximal ligation alone would suffice. In our patient the forceful retrograde filling and continued pulsations of the sac after proximal ligation, as well as the persistent headache and blindness advancing to totality, made early intracranial clipping unavoidable.

A third and unique feature in the subject of this report was the associated loss of taste, a sign that has not been previously reported. Examination of Fig. 1 shows that this aneurysm could well have stripped the dura mater upwards on the medial anterior surface of the petrous ridge, thereby causing injury to the greater superficial petrosal nerve as it leaves its canaliculus and runs medially under the Gasserian ganglion. Schwartz and Weddell have observed that elevation of the dura mater in this region at the time of retro-gasserian neurectomy is often followed by loss of taste in the anterior two-thirds of the tongue. On investigation they have found that this nerve may be an accessory taste pathway, carrying gustatory fibres from the lingual to the facial nerve, and that the usual route via the chorda tympani may be absent. This seems to be the logical explanation for loss of taste perception in the lingual area in this individual and its rapid recovery after the aneurys-
mal pressure was reduced. A possible finding that is not in full agreement with this theory is the fact that neither the continuous nor reflex secretion of tears was reduced in the left eye. These autonomic secretory fibres also are contained in the greater superficial petrosal nerve, and we have found that reflex lachrymation is abolished after the nerve has been cut (see White and Sweet, p. 443). However, there is reason to believe that the larger myelinated sensory fibres are more susceptible to pressure than small unmyelinated autonomic axones, which may have accounted for the differential paralysis of the nerve.

It is a pity that this boy did not come to neurosurgery at an earlier date before optic atrophy became so far advanced. The cause of his visual loss would doubtless have been apparent in time to save his vision, either by radiological evidence of pressure on the anterior clinoid or by visualization of the aneurysm by arteriography, long before compression of the other cranial nerves made the diagnosis obvious.

SUMMARY

Aneurysms of the carotid syphon within the cavernous sinus have long been recognized by their compression of the branches of the trigeminal, 3rd, 4th, and 6th cranial nerves. This leads to pain and varying degrees of hypaesthesia in the face and paralysis of the ocular and masticatory muscles. While both external and internal ophthalmoplegia may become complete, visual changes are rare.

Monocular blindness can result only if the infraclinoid aneurysm extends upward above the anterior clinoid to compress the optic nerve. This is a rare complication described in only 25 previous cases.

An additional case is reported with added unilateral loss of taste, probably caused by injury to the greater superficial petrosal nerve.

Proximal ligation of the common and external carotid arteries in the neck and intracranial clipping of the internal carotid gave satisfactory relief, save for continued blindness from long-standing compression and atrophy of the optic nerve.

REFERENCES

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