Bacterial aneurysms of the intracavernous carotid artery

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Six cases of bacterial intracavernous carotid artery aneurysms of extravascular origin secondary to cavernous sinus thrombophlebitis are reported along with a review of 12 similar cases collected from the literature. Of the authors' six cases, there were three children and three adults. Meningitis was found in five patients. All patients received prolonged antibiotic therapy. Spontaneous resolution of the aneurysm occurred in one patient, thrombosis of the internal carotid artery in another, and progressive enlargement of the aneurysm was seen on sequential angiography in the other two. Evidence of associated arteritis was present in all of the patients. Carotid ligation for persistent ophthalmoplegia was carried out in two patients, of whom one had a giant aneurysm and the other progressive aneurysm enlargement. The results of treatment were good in all cases. The authors believe that carotid arteriography is obligatory in cases of cavernous sinus thrombophlebitis in which ophthalmoplegia persists despite adequate antibiotic therapy.

KEY WORDS: bacterial aneurysm, intracavernous aneurysm, cavernous sinus thrombophlebitis, ophthalmoplegia, carotid ligation

Since Osler's description of an aortic aneurysm arising secondary to bacterial endocarditis, the term "mycotic aneurysm" has been spuriously used to describe any aneurysm arising due to an infectious process involving the arterial wall. This misnomer has prevailed in the literature for many years, with the term "mycotic" being used in recent reports of true mycotic (fungal) aneurysms. The designation "bacterial intracranial aneurysms" is now used for aneurysms that result from bacterial infection and the term "mycotic" is reserved for fungal (true mycotic) aneurysms.

Aneurysms of the intracavernous portion of the carotid artery constitute 5% of all intracranial aneurysms, and are reported to represent 20% to 25% of the lesions producing cavernous sinus syndrome. Bacterial intracavernous carotid artery aneurysms of extravascular origin are rare. To our knowledge, only 12 such cases have so far been reported in the English literature. In an extensive review, Brihaye encountered two more cases reported in the French literature. We report six such cases of which three were in children and three in adults. To date there is no documented case of bacterial intracranial carotid artery aneurysm of embolic origin, and to the best of our knowledge no patient with fungal (true mycotic) aneurysm of the intracavernous carotid artery has been reported.

We wish to report our experiences in the management of six cases of bacterial intracavernous carotid artery aneurysms of extravascular origin. Based on our cases and a review of the literature, we suggest a regimen for treatment of these rare vascular lesions.

Case Reports

Case 1

This 2½-year-old girl was brought to the hospital in a state of altered sensorium of 24 hours' duration. One month before admission she was treated elsewhere for left supraorbital cellulitis, with drainage of pus and systemic antibiotics. Three days before admission to our hospital she developed high fever with vomiting and diarrhea.

At admission, she was comatose with spontaneous decerebration. She had bilateral papilledema and ophthalmoplegia. A scar was present over the left upper eyelid. Initial laboratory data revealed a hemoglobin of 8.8 gm%, a total white blood cell (WBC) count of 30,900/cu mm, and an erythrocyte sedimentation rate (ESR) of 93 mm/1st hr.

A clinical diagnosis of left frontal abscess with meningitis was made. Left carotid angiography on the day of admission revealed a 4 x 3-mm aneurysm of the left intracavernous carotid artery. There was evidence of
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FIG. 1. Case 1. Left carotid angiograms. Left: On admission an aneurysm is seen arising from the intracavernous portion of the carotid artery, with narrowing of the supraclinoid segment and marked stretching of the anterior cerebral artery. Right: Seven months after admission the aneurysm has disappeared and the narrowing of the supraclinoid segment of the internal carotid artery has resolved.

arteritis involving the supraclinoid segment and ventricular dilatation (Fig. 1 left), which was confirmed at ventriculography. A brain abscess was excluded. Ventricular cerebrospinal fluid (CSF) revealed a protein level 28 mg/dl, a glucose concentration of 80 mg/dl, and a WBC count of 55/cu mm (neutrophils 65% and lymphocytes 35%). External ventricular drainage was established. Lumbar puncture yielded CSF containing 400 WBC's/cu mm (neutrophils 60% and lymphocytes 40%), 220 mg/dl of protein, and 15 mg/dl of glucose. As the CSF culture grew no organisms, the patient was placed on broad-spectrum antibiotic therapy, anticonvulsant agents, and steroids. She showed steady improvement, and a ventriculoperitoneal (VP) shunt was inserted when the CSF analysis returned to normal.

Right carotid angiography carried out a month after admission was essentially normal except for findings suggesting mild ventricular dilatation. Orbital venography demonstrated bilateral thrombosis of the cavernous sinus and ophthalmic veins, with a cluster of abnormal venous channels (Fig. 2). The patient improved slowly, with the return of ocular movements to normal without any neurological deficit. Repeat left carotid angiography 7 months later showed complete disappearance of the aneurysm (Fig. 1 right). At the last evaluation, 3 years after discharge from the hospital, the child was attending school without any detectable neurological deficit.

Case 2

This 5½-year-old boy presented with a history of continuous fever for 36 days, and headache, vomiting, and altered sensorium for 18 days. The sudden onset of bilateral proptosis and diplopia was noted 12 days before admission. He was admitted to a local hospital where a lumbar puncture yielded cloudy CSF with abundant pus cells, a glucose level of 60 mg/dl, and a protein value of 40 mg/dl. Culture of the CSF grew Staphylococci. A diagnosis of pyogenic meningitis was made and antimeningitic therapy was started. Although his fever subsided, he was referred to us for persisting clinical signs of meningitis and left-sided ophthalmo-plegia.

At admission, the patient was afebrile, but signs of meningeal irritation were present. He had bilateral mild proptosis with total ophthalmoplegia on the left side. Ocular fundi were normal. Lumbar CSF analysis revealed 2500 WBC's/cu mm (polymorphonuclear cells 95% and lymphocytes 5%) with a glucose level of 38 mg/dl and a protein content of 86 mg/dl; CSF culture was sterile. Chest x-ray films revealed multiple bilateral staphylococcal lung abscesses. His meningitis and proptosis improved following treatment with gentamicin and cloxacillin for 3 weeks. However, ophthalmoplegia remained unchanged. Left carotid angiography revealed a giant 32 × 25-mm aneurysm of the intracavernous carotid artery, with gross arteritis (Fig. 3). Right carotid angiography was normal and showed excellent filling of the left cerebral vessels on cross-compression. Left internal carotid artery (ICA) ligation in the neck was carried out 2 months after the onset of ophthalmoplegia. When the patient was seen 5 months after carotid ligation, his ophthalmoplegia had improved, and, except for mild adductor paresis, he had no other neurological deficit.

Case 3

This 6-year-old girl was referred to us as a case of bilateral cavernous sinus thrombosis. Her symptoms had started with a furuncle of the nose and orbital cellulitis associated with fever and headache. A week later she developed bilateral ophthalmoplegia and right-sided facial palsy.

FIG. 2. Case 1. Orbital venogram showing bilateral thrombosis of the cavernous sinus and ophthalmic veins, with a cluster of abnormal venous channels on the left side.
FIG. 3. Case 2. Left carotid angiograms, anteroposterior (left) and lateral (right) views, showing a giant lobulated aneurysm of the intracavernous portion of the internal carotid artery.

FIG. 4. Case 3. Left carotid angiograms showing an aneurysm of the intracavernous carotid artery with narrowing of the supraclinoid segment (left), and spontaneous thrombosis of the internal carotid artery 4 months later (right).

Examination revealed altered sensorium, classical features of meningitis, bilateral proptosis with total ophthalmoplegia, papilledema, and spasticity of all four limbs. The patient required ventilatory support to relieve her respiratory distress. Laboratory studies showed a hemoglobin of 9.5 gm% with an ESR of 76 mm/1st hr. Lumbar CSF was cloudy, with 420 WBC's/cu mm (polymorphonuclear cells 80% and lymphocytes 20%), a glucose level of 30 mg/dl, and a protein content of 104 mg/dl. Culture of blood and CSF grew no organisms. Chest x-ray films showed patchy bilateral consolidation of the lungs suggestive of staphylococcal pneumonia. Left carotid angiography 1 week after admission revealed an 11 x 10-mm aneurysm of the intracavernous carotid artery, with arteritis and possible ventricular dilatation (Fig. 4 left). Right carotid angiography also showed features of arteritis involving the intracavernous segment of the ICA. Orbital venography confirmed thrombosis of the cavernous sinus and ophthalmic veins bilaterally with a cluster of abnormal venous channels.

The patient was treated with systemic gentamicin, cloxacillin, steroids, and anticonvulsant drugs. She showed steady improvement, and a VP shunt was placed 2 months after admission for communicating hydrocephalus, when the CSF analysis returned to normal. Repeat bilateral carotid angiography 4 months later showed spontaneous thrombosis of the left ICA (Fig. 4 right) with good cross-circulation from the right side. She was discharged from the hospital, as her ophthalmoplegia also showed progressive improvement.

Case 4

This 18-year-old woman developed a furuncle over the right periorbital region, followed by swelling in that area, fever, and headache 1½ months before admission to another institution. The periorbital abscess was drained surgically and she was treated with broad-spectrum antibiotic therapy. While receiving antibiotic therapy she developed diplopia and right ophthalmoplegia, and also had a generalized seizure. She was then referred to us for persisting right ophthalmoplegia following cavernous sinus thrombophlebitis.

At admission the patient had a total right ophthalmoplegia with proptosis and diminished right corneal sensation. Ocular fundi were normal. There were no clinical features of meningitis. Lumbar puncture revealed clear CSF with 2 lymphocytes. Right carotid angiography demonstrated an irregular intracavernous carotid artery aneurysm, measuring 13 x 12 mm, with features of arteritis (Fig. 5 left). Although she showed slow but progressive improvement from her ophthalmoplegia on prolonged antibiotic therapy, repeat carotid angiography 6 weeks later (Fig. 5 right) surprisingly showed marked enlargement of the aneurysm, which now measured 19 x 17 mm. The patient declined surgical intervention. When she was last seen as an outpatient 6 weeks following her second angiography and discharge from hospital, she was asymptomatic with full ocular movements, but her right pupil remained larger and poorly reactive.

Case 5

This 32-year-old woman was referred to us with a diagnosis of right cavernous sinus thrombosis. She had undergone drainage of an abscess on the right side of her face 12 days before admission to our service. She presented with fever, headache, vomiting, diplopia, and swelling of the right side of her face.

At admission she was toxic, emaciated, and had all the clinical features of meningitis. Subconjunctival hemorrhage was present on the left side. She had bilateral lateral rectus palsy. Initial laboratory data revealed a hemoglobin of 8 gm%, a peripheral WBC count of 13,650/cu mm, with 90% polymorphonuclear cells and 10% lymphocytes, and an ESR of 141 mm/1st hr. Lumbar puncture yielded cloudy CSF containing 1120
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WBC's with 98% polymorphonuclear cells and 2% lymphocytes. Although the CSF culture grew no organisms, culture of the sputum grew *Klebsiella* and *Escherichia coli*. Chest x-ray films showed patchy consolidation and pleural effusion on the right.

The patient was put on antimeningitic antibiotic therapy. Right carotid angiography after admission showed an irregular aneurysmal dilatation with partial thrombosis of the intracavernous portion of the carotid artery and proximal arteritis (Fig. 6). However, the distal vessels were normal. The patient then developed right total ophthalmoplegia and left hemiparesis. Orbital venography demonstrated bilateral cavernous sinus and intraorbital venous thrombosis. Her pleural effusion and lung consolidation improved following aspiration and antibiotic therapy. Repeat lumbar puncture after 3 weeks of therapy yielded normal CSF. During her hospital stay her right ophthalmoplegia and left hemiparesis showed a steady improvement.

Case 6

This 59-year-old man presented with fever, retroorbital pain on the right, and drooping of the right eyelid, followed by total ophthalmoplegia of 12 days' duration.

At admission he had periorbital edema on the right with total ophthalmoplegia. The rest of the neurological examination was normal. His hematological profile revealed a total WBC count of 23,000/cu mm with marked polymorphonuclear pleocytosis and an ESR of 100 mm/1st hr. The CSF obtained at lumbar puncture showed 460 WBC's/cu mm (polymorphonuclear cells 60% and lymphocytes 40%), a glucose content of 51 mg/dl, and a protein level of 167 mg/dl. A diagnosis was made of right cavernous sinus thrombophlebitis and meningitis, and he was put on a course of systemic antibiotics. Right carotid angiography performed 1 week after admission revealed a 17 × 12-mm lobulated intracavernous carotid artery aneurysm with marked proximal arteritis (Fig. 7 left pair). Orbital venography demonstrated thrombosis of the right cavernous sinus. Left carotid angiography was normal.

As the clinical features and CSF findings failed to respond to routine antimeningitic therapy, anti-tubercular treatment was instituted, to which the patient responded well. However, his ophthalmoplegia remained unchanged. Repeat right carotid angiography 2 and 4 months later demonstrated progressive enlarge-
Discussion

Bacterial intracranial aneurysms constitute 2.5% to 6.2% of all intracranial aneurysms reported recently.3,19 Karsner grouped these aneurysms into three types on the basis of pathogenesis: aneurysms of "embolic origin" secondary to bacterial endocarditis are the commonest.3,4,10 Aneurysms of "extravascular origin," formed as a result of extension of infection from a neighboring septic focus, although less common, have been encountered secondary to meningitis, cavernous sinus thrombophlebitis, osteomyelitis of the skull, and sinus infections.3,4,17,19,20,23,25,26 The third group that occurs in the absence of an obvious inflammatory focus elsewhere in the body has been called "primary or cryptogenic" mycotic aneurysms. The location of the carotid artery within the cavernous sinus makes it vulnerable to infection from septic cavernous sinus phlebitis, resulting in varying degrees of arteritis.12,23,25,26 Weisman, in an autopsy study of a case of cavernous sinus thrombophlebitis, noted infiltration of polymorphonuclear leukocytes into the adventitia and media with marked intimal proliferation, suggesting focal arteritis of the intracavernous portion of the carotid artery. Bacterial infections are known to invade the arterial wall from neighboring or contiguous inflammatory foci.1,14,17,23 Arterial thrombosis may result from diffusion of toxin or involvement of the intima from the adjacent inflammatory process.14,23 The infection may produce disintegration and weakness of the vessel wall, resulting in rupture or formation of an infectious aneurysm.16 Some authors believe that inflammation affects the adventitia first and then spreads inward, finally attacking the internal elastic membrane, as seen in cases of infectious aneurysms secondary to meningitis and oesteomyelitis of the skull.17,20,23

To our knowledge, only 12 cases of bacterial intracavernous carotid artery aneurysms secondary to cavernous sinus thrombophlebitis have been previously reported in the English literature.1,2,7,8,11,13,22,23,25 The addition of our six cases raises the total number to 18 (Table 1). Of these, the aneurysms were bilateral in three. Ten patients, including three of ours, were children. In the present series, although the aneurysms were unilateral, bilateral cavernous sinus thrombosis was present in two patients. A septic focus leading to cavernous sinus thrombophlebitis could be demonstrated in all of the cases. At admission, four of our patients had altered sensorium in addition to focal neurological deficits; meningitis was found in five and septicemia in one. Despite adequate prior antibiotic therapy, the causative organism could be isolated in three. However, long-term antibiotic therapy was continued in all the patients. Staphylococcus had been the commonest organism isolated in nine of the total 18 cases, including two of ours; however, no organism could be grown in the remaining nine cases.

Except for a single case of bilateral aneurysms observed at autopsy by Barker,2 the diagnosis of intracavernous carotid artery aneurysms was established at angiography in all previously reported cases. This study was performed for evaluation of persisting ophthalmoplegia following cavernous sinus thrombophlebitis, despite prolonged antibiotic therapy. In all of our cases, the aneurysms were also detected at angiography, carried out for suspected brain abscess in one patient (Case 1) and failure to achieve clinical improvement within 4 to 6 weeks in the other five patients. The indications for angiography in patients with cavernous sinus thrombophlebitis and the incidence of bacterial aneurysms in patients with infective cavernous sinus syndromes are not yet defined. Computerized tomography (CT) may help differentiate orbital cellulitis from cavernous sinus thrombophlebitis.25 Tomita, et al., were able to demonstrate a contrast-enhancing mass lesion in the cavernous sinus on CT, which was found to be a large aneurysm at angiography. In addition to diagnosing aneurysms, angiography may demonstrate features of associated arteritis or even arterial thrombosis. When patients are receiving prolonged courses of antibiotic therapy, repeat angiography is indicated to monitor the lesion in connection with the clinical course of the patient. Of our four patients in whom repeat angiography was carried out, two aneurysms revealed progressive enlargement (Cases 4 and 6), spontaneous disappearance of the aneurysm was noted in one (Case 1), and ICA thrombosis was found in the fourth (Case 2). Sequential angiography performed in seven of the previously reported cases demonstrated progressive enlargement of the aneurysm in three cases, and disappearance of aneurysm/thrombosis of the parent artery in four cases.7,11,23 Follow-up angiography thus remains a valuable tool in the management of these lesions.

In the management of bacterial intracavernous carotid artery aneurysms with ophthalmoplegia secondary to cavernous sinus thrombophlebitis, the role of sequential angiography with prolonged antibiotic therapy has been emphasized in isolated case reports.6,18,22,25,27 Conservative treatment alone with prolonged antibiotic therapy in nine of the reported patients (Table 1) achieved good recovery from ophthalmoplegia in eight, including three of ours, and the remaining patient was lost to follow-up review.17,11,23 Of these eight patients, sequential angiography carried out in six cases revealed disappearance of the aneurysm in two, thrombosis of
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### TABLE 1

Bacterial aneurysms of the intracavernous carotid artery*

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Authors &amp; Year</th>
<th>Age (yrs), Sex</th>
<th>Organism</th>
<th>Side</th>
<th>Arteritis</th>
<th>Treatment</th>
<th>Course</th>
<th>Result</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Barker, 1954</td>
<td>32, M</td>
<td>--</td>
<td>BL</td>
<td>no</td>
<td>conservative</td>
<td>It thrombosed, rt ruptured</td>
<td>died</td>
</tr>
<tr>
<td>2</td>
<td>Devadiga, et al., 1969</td>
<td>1½, M</td>
<td>--</td>
<td>UL</td>
<td>yes</td>
<td>conservative</td>
<td>ICA thrombosis</td>
<td>good</td>
</tr>
<tr>
<td>3</td>
<td>Suwanwela, et al., 1972</td>
<td>6, M</td>
<td>staph</td>
<td>UL</td>
<td>yes</td>
<td>conservative</td>
<td>ICA thrombosis</td>
<td>good</td>
</tr>
<tr>
<td>4</td>
<td></td>
<td>6, F</td>
<td>staph</td>
<td>UL</td>
<td>no</td>
<td>conservative</td>
<td>aneurysm disappeared</td>
<td>good</td>
</tr>
<tr>
<td>5</td>
<td></td>
<td>10, M</td>
<td>staph</td>
<td>UL</td>
<td>no</td>
<td>conservative</td>
<td>lost to follow-up</td>
<td>---</td>
</tr>
<tr>
<td>6</td>
<td></td>
<td>33, M</td>
<td>--</td>
<td>UL</td>
<td>no</td>
<td>CCA ligation</td>
<td>--</td>
<td>poor</td>
</tr>
<tr>
<td>7</td>
<td>Adeloye, et al., 1973</td>
<td>30, M</td>
<td>--</td>
<td>UL</td>
<td>no</td>
<td>conservative</td>
<td>--</td>
<td>good</td>
</tr>
<tr>
<td>8</td>
<td>Lansky &amp; Maxwell, 1975</td>
<td>7, M</td>
<td>staph</td>
<td>UL</td>
<td>yes</td>
<td>CCA ligation</td>
<td>aneurysm enlarged</td>
<td>good</td>
</tr>
<tr>
<td>9</td>
<td>Shibuya, et al., 1976</td>
<td>42, F</td>
<td>staph</td>
<td>UL</td>
<td>yes</td>
<td>ICA ligation (neck &amp; intracranial), ECA ligation</td>
<td>aneurysm enlarged</td>
<td>good</td>
</tr>
<tr>
<td>10</td>
<td>Johnston, 1979</td>
<td>3, M</td>
<td>staph</td>
<td>BL</td>
<td>yes</td>
<td>ICA ligation &amp; wrapping</td>
<td>It thrombosed</td>
<td>good</td>
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<tr>
<td>11</td>
<td>Tomita, et al., 1981</td>
<td>2, M</td>
<td>staph</td>
<td>UL</td>
<td>no</td>
<td>ICA ligation</td>
<td>aneurysm enlarged</td>
<td>good</td>
</tr>
<tr>
<td>12</td>
<td>Eguchi, et al., 1982</td>
<td>22, M</td>
<td>--</td>
<td>BL</td>
<td>yes</td>
<td>BL STA-MCA anastomosis, ICA ligation</td>
<td>--</td>
<td>good</td>
</tr>
<tr>
<td>13</td>
<td>Rout, et al., 1984</td>
<td>2½, F</td>
<td>--</td>
<td>UL</td>
<td>yes</td>
<td>conservative</td>
<td>aneurysm disappeared</td>
<td>good</td>
</tr>
<tr>
<td>14</td>
<td></td>
<td>5½, M</td>
<td>staph</td>
<td>UL</td>
<td>yes</td>
<td>ICA ligation</td>
<td>--</td>
<td>good</td>
</tr>
<tr>
<td>15</td>
<td></td>
<td>6, F</td>
<td>staph</td>
<td>UL</td>
<td>yes</td>
<td>conservative</td>
<td>ICA thrombosis</td>
<td>good</td>
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<tr>
<td>16</td>
<td></td>
<td>18, F</td>
<td>--</td>
<td>UL</td>
<td>yes</td>
<td>conservative</td>
<td>aneurysm enlarged</td>
<td>good</td>
</tr>
<tr>
<td>17</td>
<td></td>
<td>32, F</td>
<td>--</td>
<td>UL</td>
<td>yes</td>
<td>conservative</td>
<td>aneurysm enlarged</td>
<td>good</td>
</tr>
<tr>
<td>18</td>
<td></td>
<td>59, M</td>
<td>--</td>
<td>UL</td>
<td>yes</td>
<td>CCA ligation</td>
<td>aneurysm enlarged</td>
<td>good</td>
</tr>
</tbody>
</table>

*UL = unilateral; BL = bilateral; ICA = internal carotid artery; CCA = common carotid artery; staph = Staphylococcus; ECA = external carotid artery; STA = superficial temporal artery, MCA = middle cerebral artery.

the ICA in three, and the unanticipated enlargement of the aneurysm in one, who declined surgical intervention. Thus, it may be suggested that, in patients in whom ophthalmoplegia improves, and the aneurysm remains static or becomes smaller on sequential angiography 4 to 6 weeks after admission, conservative management may be continued.

Carotid ligation for unilateral aneurysms with persisting ophthalmoplegia has been found successful in the management of these lesions.11,13,22,23,25 A recent report by Eguchi, et al.,8 of a patient with bilateral intracavernous carotid artery aneurysms treated with bilateral superficial temporal to middle cerebral artery anastomosis and bilateral carotid ligation is encouraging. Carotid ligation carried out for persisting ophthalmoplegia in spite of prolonged antibiotic therapy, in eight patients (including two of ours) achieved good results in all except one, in whom poor recovery was attributed to delay in the treatment.25 Although neurological morbidity following carotid ligation for various other lesions has been found to be as high as 20%,13 no acute complication has been encountered in the reported cases with carotid ligation for bacterial intracavernous carotid artery aneurysms. In two of our cases, carotid ligation even as long as 2 to 5 months after onset of symptoms achieved good recovery from ophthalmoplegia without any additional neurological complications. Carotid ligation is thus indicated irrespective of the clinical status of ophthalmoplegia if the aneurysm is found to be enlarging on sequential angiography.

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References


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