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January 1995, Volume 36, Number 1 183 Subarachnoid Neurocysticercosis with Occlusion of the Major Intracranial Arteries: Case Report Case Report

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ABSTRACT: CYSTICERCOSIS IS THE most common parasitic disease affecting the central nervous system. Stroke is a recognized complication of neurocysticercosis, occurring in 2 to 12% of cases, mostly in the form of small lacunar infarcts. We report a case of hemiparesis and aphasia in a 51-yearold Hispanic woman, which was secondary to complete occlusion of the left internal carotid and bilateral anterior cerebral arteries. Magnetic resonance imaging demonstrated the presence of enhancing subarachnoid material surrounding these occluded cerebral arteries, providing antemortem, noninvasive documentation of the inflammatory meningeal cysticercotic reaction that was presumably responsible for the occlusive arteritis causing the cerebral infarction. This represents the third reported case of internal carotid artery occlusion and the first reported case of anterior cerebral artery occlusion secondary to neurocysticercosis.

<u>KEY WORDS</u>: Cysticercosis; Hydrocephalus; Stroke

Cysticercosis is the most common parasitic disease affecting the central nervous system. Seizures, hydrocephalus, and stroke are the usual presenting complications, with stroke or focal neurological deficit as the primary manifestation of neurocysticercosis (NCC) in 2-12% of cases ^(5,7-9,12, 14,15,17). Most instances of cerebral infarction associated with NCC involve occlusive arteritis in small perforating vessels, resulting in lacunar infarcts ^(1,4-8,15). Only six cases of cerebral infarction from the occlusion of a major intracranial artery in patients with NCC have been reported--four cases of middle cerebral artery (MCA) occlusion ^(13,16) and two cases of internal carotid artery (ICA) occlusion ^(11,17).

We report the third case of complete occlusion of the ICA resulting from inflammatory arteritis of subarachnoid cysticercosis. In the same patient, we demonstrate, for the first time, bilateral occlusion of the A2 segments of the anterior cerebral arteries (ACA) from an adjacent inflammatory subarachnoid lesion in the interhemispheric fissure. The magnetic resonance imaging (MRI) characteristics of these NCC inflammatory cysts surrounding the arteries in the subarachnoid space are detailed.

CASE REPORT

A 51-year-old right-handed Hispanic woman presented to the emergency room at our institution with a history of increasing weakness on the right side and difficulty with speech occurring over several days. She had recently moved from Guadalajara, Mexico, where she had been diagnosed with NCC 6 weeks before admission to our facility. At the time of the initial diagnosis, she was observed to have a mild right hemiparesis. A computed tomographic (CT) scan revealed mild-to-moderate hydrocephalus, but no evidence of cerebral infarction (*Fig. 1A*). She was treated with albendazole, and her hemiparesis improved.

At admission to our institution, she reported a 4day history of an increased right hemiparesis and a new aphasia. She denied any headache, nausea, or emesis, but did report a fever that reached 101°F 4 days before admission. Her medical history was negative for hypertension, cardiac disease, or diabetes, and there was no significant family history of ischemic cerebrovascular disease.

At admission, her temperature was 37.0°C, her heart rate was 72, her blood pressure was 100/60, and her respiratory rate was 16. Her general physical examination was normal, including bilateral normal carotid pulses without bruits.

A neurological examination revealed an alert Hispanic woman with a nonfluent aphasia. She was unable to name objects or repeat words and could only follow simple one-step commands. Her pupils measured 3 mm and were reactive; her visual fields were full: and her ocular movements were intact. She was observed to have lower right facial weakness, consistent with a left hemispheric central facial palsy. Her motor power was 5/5 on the left, 2 to 3/5 in the right arm and 3 to 4/5 in the right leg. Her deep tendon reflexes were slightly increased on the right, and the patient exhibited a right Babinski reflex. Her sensation to light touch and pin prick was intact bilaterally. Her electrolyte, glucose, and cholesterol levels were normal, as was her complete blood count; her erythrocyte sedimentation rate was 30, and the cerebrospinal fluid cysticercosis immunoblot assay was positive.

A CT scan on the day of admission (*Fig. 1B*) demonstrated increased hydrocephalus, compared with the CT scan from Mexico obtained 1 month earlier (*Fig. 1A*). Scattered small calcified lesions were seen throughout the cerebral parenchyma. There were enhancing hypodensities in the left MCA distribution (*Fig. 1B and C*), consistent with subacute infarcts, and nonenhancing low density in the left frontal white matter. Enhancing material in the region of the subarachnoid ICA and the proximal ACAs was also noted (*Fig. 1D*).

The patient was admitted to the surgical intensive care unit and began receiving dexamethasone intravenously, 10 mg every 6 hours; gentle volume expansion with intravenous administration of 25% albumin, 25 gm every 8 hours, was initiated. She also began receiving praziquantel, 900 mg orally, three times a day. The morning after admission, a cerebral angiogram was performed that revealed complete occlusion of the left ICA at the level of the anterior choroidal artery (*Fig. 2A-C*). Bilateral occlusion of the A2 segments of the ACAs was noted on the right ICA injection (*Fig. 2D*), and on late arterial phase films, the right ACA distribution demonstrated retrograde filling from the right MCA collaterals. After the angiogram, a ventriculoperitoneal shunt was placed for treatment of the hydrocephalus. After ventriculoperitoneal shunt placement, more aggressive volume expansion was performed and the patient demonstrated improvement with respect to her hemiparesis and aphasia. An echocardiogram was normal.

After the ventriculoperitoneal shunt placement, an MRI scan with gadolinium was obtained that revealed enhancing subarachnoid material surrounding the left ICA from its entry into the subarachnoid space to its bifurcation. The expected flow void of the ICA was diminished in diameter proximally and absent throughout the remainder of its course in the subarachnoid space (Fig. 3A-D). Enhancing material was also seen in the interhemispheric fissure in the region of the A2 segments of the ACAs (Fig. 4). The left MCA distribution infarcts enhanced as they did on the CT scan. The ventricles were slightly decreased in size, compared with the CT scan obtained before the placement of the ventriculoperitoneal shunt. During the ensuing weeks, the treatment with dexamethasone was tapered slowly and changed to prednisone; the patient was then weaned from the prednisone after the completion of a 4-week course of praziquantel.

At a 2-month follow-up, the patient demonstrated significant neurological improvement and was able to name objects and to follow three-step commands. Her right hemiparesis had improved significantly, with her gait now normal, but she continued to demonstrate a moderate weakness in her right hand. A follow-up MRI scan revealed a normal ventricular size and persistent signal change in the region of the left MCA infarct.

DISCUSSION

With the increasing numbers of immigrants to the United States from regions in which cysticercosis is endemic, it is imperative that we familiarize ourselves with the diverse clinical syndromes of NCC. Most physicians with an interest in neurological disorders, especially those in the West and Southwest, readily recognize the classic aspects of CT scans in patients with NCC and are quite familiar with the presentation and management of patients with hydrocephalus or seizures associated with NCC. However, in patients with ischemic cerebrovascular events, NCC is seldom considered in the differential diagnosis; concern for patients developing a stroke during the course of treatment for NCC, as occurred in this case, rarely arises.

In large series of patients with NCC, the reported incidence of stroke ranges from 2 to 12% ^(5,7-9,12,14,15, 17). Conversely, in endemic regions, cysticercosis has been considered as the cause of stroke in 2 to 7% of

cases in large stroke series ^(1,3). Alarcon et al. ⁽²⁾ found cerebral cysticercosis to be an independent risk factor for stroke in a group of patients under the age of 65 in Ecuador. However, with stroke defined simply as the sudden onset of focal neurological deficit, some patients have been diagnosed with an NCCassociated stroke without any evidence of anatomical cerebral infarction by radiographic studies or autopsy $^{(1,3,12)}$. In some patients, a focal neurological deficit may be explained by the presence of a large intraparenchymal NCC cyst and the resulting mass effect, with no indication of true cerebral infarction ^(12,15,17). Of the cases with clear documentation of cerebral infarction, small lacunar infarcts are decidedly the most common cerebrovascular complication of NCC (1,4-8,15).

Only six cases of stroke associated with occlusion of a major intracranial artery in a patient with NCC have been reported (Table 1); four involved the MCA (13,16), and two involved the ICA (11,17). We report the third case of complete occlusion of the ICA from cysticercotic arteritis. This is the first such case involving the ACA. Presumably, occlusion of these arteries has occurred as a result of the intense inflammatory reaction associated with the adjacent cysticerci in the subarachnoid space. A dense inflammatory exudate is believed to surround the arteries of the circle of Willis in patients with basilar meningitis secondary to cysticercosis. The vessel walls are then invaded by inflammatory cells, resulting in an occlusive endarteritis, with thickening of the adventitia, fibrosis of the media, and endothelial hyperplasia (7,10,13,17).

Recently, it has been recognized that patients with meningeal NCC may develop a cerebral infarction during a course of anticysticercal drug therapy. In at least two reported cases of stroke associated with NCC, the cerebral infarction occurred while the patient was receiving praziquantel ^(5,7). In our case, the stroke occurred while the patient was undergoing therapy with albendazole. It is possible that the meningeal inflammatory reaction, leading to occlusive endarteritis, is intensified by the anticysticercal drug as a result of acute destruction of the parasites in the subarachnoid space and the subsequent host inflammatory reaction. It would seem prudent, as has been suggested by others ^(5,7,9), to administer corticosteroids during antibiotic treatment for meningeal cysticercosis. Some authors have recommended using steroids in conjunction with anticysticercal drugs only after patients have demonstrated clinical signs and symptoms of an increased inflammatory reaction, such as fever, headache, nausea/vomiting, seizures, or increased neurological deficit ^(8,9). We think it is appropriate to await these clinical signs and symptoms before adding steroids when treating parenchymal cycticercosis, because, in this setting, local parenchymal inflammation and edema is the likely cause of patient deterioration, and this is usually readily reversible with steroid treatment. However, in cases of subarachnoid NCC, we think that the significant risk of neurological deterioration from inflammatory arteritis, with subsequent stroke during

antibiotic therapy, warrants the routine use of corticosteroids in patients demonstrated to have extensive disease in the basilar subarachnoid space. In such patients, waiting for clinical signs of neurological deterioration before administering steroids may result in an irreversible deficit or even death from cerebral infarction. Therefore, our preference in the management of subarachnoid NCC has been the use of anticysticercal drugs along with dexamethasone at the onset of therapy.

We have reserved surgical intervention for patients in whom there is an acute obstruction to the flow of cerebrospinal fluid caused by a focal cysticercotic lesion or for patients with a significant focal neurological deficit secondary to a large subarachnoid or parenchymal cysticercus with mass effect. In these situations, surgery is the treatment of choice; for patients with evidence of residual disease, antibiotics are used, with or without the addition of steroids, according to the factors discussed above. The benefit of surgery to remove subarachnoid lesions in patients at risk for vascular occlusion is unclear. McCormick et al. ⁽¹¹⁾ reported a case of carotid artery occlusion from NĈC that was managed by direct surgical excision of a cluster of cysts from around the right ICA in the subarachnoid space. Despite the fact that the ICA was already occluded and no attempt was made to reopen it, the patient did well and recovered from the left hemiparesis, apparently as a result of improvement in the cerebral perfusion pressure that occurred by keeping the patient recumbent.

Our goal in the management of patients with NCC is to recognize those at risk for stroke before the development of neurological deficits. We have shown that a gadolinium enhanced MRI scan can be used to identify patients with extensive inflammatory basilar NCC meningitis who may be at increased risk for stroke during therapy with anticysticercal drugs; these patients should, therefore, be considered as candidates for treatment with adjuvant corticosteroids.

A review of the literature reveals only one reported case in which MRI was employed in the evaluation of a patient with a stroke related to the occlusion of a major intracranial vessel from NCC meningitis (16). In this report of a patient with bilateral MCA occlusions, terPenning et al.⁽¹⁶⁾ demonstrated the presence of a cyst in the region of the MCA bifurcation, but no gadolinium enhanced scans were included. In our case, gadolinium MRI demonstrated enhancing material in the subarachnoid space surrounding the occluded left ICA as well as an enhancing lesion adjacent to the proximal A2 segments of the ACAs. This focal subarachnoid enhancement, when correlated with the occlusion of the adjacent vessels and a positive cerebrospinal fluid cysticercosis immunoblot assay, is consistent with an inflammatory exudate associated with a cluster of NCC cysts. Thus, with enhanced MRI, we have provided clear antemortem, noninvasive documentation of the inflammatory meningeal cysticerci presumably responsible for the occlusive endarteritis causing cerebral infarction in a patient

with neurocysticercosis.

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REFERENCES: ⁽¹⁻¹⁷⁾

- 1. Alarcon F, Hidalgo F, Moncayo J, Vinan I, Duenas G: Cerebral cysticercosis and stroke. **Stroke** 23:224-228, 1992.
- 2. Alarcon F, Vanormelingen K, Moncayo J, Vinan I: Cerebral cysticercosis as a risk factor for stroke in young and middle-aged people. **Stroke** 23:1563-1565, 1992.
- 3. Barinagarrementeria F, Cantu C: Neurocysticercosis as a cause of stroke. **Stroke** 23:1180-1181, 1992 (letter).
- 4. Barinagarrementeria F, Del Brutto OH: Neurocysticercosis and pure motor hemiparesis. **Stroke** 19:1156-1158, 1988.
- 5. Barinagarrementeria F, Del Brutto OH: Lacunar syndrome due to neurocysticercosis. **Arch Neurol** 46:415-417, 1989.
- Barinagarrementeria F, Del Brutto OH, Otero E: Ataxic hemiparesis from cysticercosis. Arch Neurol 45:246, 1988 (letter).
- Del Brutto OH: Cysticercosis and cerebrovascular disease: Review. J Neurol Neurosurg Psychiatry 55:252-254, 1992.
- Del Brutto OH, Sotelo J: Neuroscysticercosis: An update. Rev Infect Dis 10:1075-1087, 1988.
- 9. Ernest MP, Reller LB, Filley CM, Grek AJ: Cysticercosis in the United States: 35 cases and a review. **Rev Infect Dis** 9:961-979, 1987.
- Escobar A: The pathology of neurocysticercosis, in Palacios E, Rodriguez-Carbajal J, Taveras JM (eds:) *Cysticercosis of the Central Nervous System*. Springfield, Charles C Thomas, 1983, pp 27-54.
- McCormick GF, Giannotta SL, Zee C-S, Fisher M: Carotid occlusion in cysticercosis. Neurology 33:1078-1080, 1983.
- 12. McCormick GF, Zee C-S, Heiden J: Cysticercosis cerebri: Review of 127 cases. Arch Neurol 39:534-539, 1982.
- 13. Rodriguez-Carbajal J, Del Brutto OH, Penagos P, Huebe J, Escobar A: Occlusion of the middle cerebral artery due to cysticercotic angiitis. **Stroke** 20:1095-1098, 1989.
- 14. Scharf D: Neurocysticerosis: Two hundred thirty-eight cases from a California hospital: **Arch Neurol** 45:777-780 1988.
- 15. Sotelo J, Guerrero V, Rubio F: Neurocysticercosis: A new classification based on active and inactive forms. Arch Intern Med 145:442-445, 1985.
- 16. terPenning B, Litchman CD, Heier L: Bilateral middle cerebral artery occlusions in

neurocysticercosis. Stroke 23:280-283, 1992.

 Torrealba G, Del Villar S, Tagle P, Arriagada P, Kase CS: Cysticercosis of the central nervous system: Clinical and therapeutic considerations. J Neurol Neurosurg Psychiatry 47:784-790, 1984.

COMMENTS

This is a report of a rare complication of subarachnoid neurocysticercosis and stroke. Cysticercosis, especially subarachnoid cysticercosis, should be considered as a cause of ischemic cerebrovascular events in areas where there is an increasing influx of immigrants from the endemic areas. This report provided an excellent discussion on the treatment of subarachnoid cysticercosis. Surgery is indicated when the cerebrospinal fluid pathway is obstructed or when a large cyst is compressing on vital structures such as the brain stem. From our experience, gadolinium enhanced magnetic resonance images may provide valuable information in predicting whether surgery will be successful. Enhancement of the cyst wall indicates the presence of adhesion and reaction to adjacent tissue, and attempts to remove these cysts surgically may not succeed. A nonenhancing cyst disclosed by magnetic resonance images is easily removed at surgery.

Medical treatment with anticysticercal drugs is a controversial issue. As stated by the authors, patients with meningeal cysticercosis may develop a cerebral infarction during a course of anticysticercal drug therapy. We concur with the authors that the significant risk of neurological deterioration from inflammatory arteritis, with subsequent stroke during antibiotic therapy, warrants the routine administration of corticosteroids in patients demonstrated to have extensive disease in the basilar subarachnoid space. We believe medical treatment merely accelerates the degeneration of cysticercosis cysts. Whether patients will benefit from such treatment in the long run requires a well-controlled prospective study.

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Levy et al.'s report brings up a frequent clinical situation in which Latin American neurosurgeons are involved everyday: circulatory complications of subarachnoid neurocysticercosis resulting from inflammatory and scarring reactions in major or minor vessels. Because it is such a common situation in our environment, we are used to living with it, so we have scarcely felt the need of publishing such an important issue. The relevance of this report is that it puts in "black and white" a very common and usually deleterious complication of this disease, with the example of a very characteristic and well-documented case.

Another important aspect that the authors emphasize is the increase of neurological symptoms and signs after the antiparasite treatment as a result of the liberation of enzymes when the parasites die, which should always be taken in consideration when a patient is going to be treated with drugs. This is true in any of the different locations of living parasites and may increase parenchymal, cranial nerve, or vascular signs and symptoms. This article is a good example of how to use one clinical case to remark upon the important clinical features of a disease.

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REFERENCES: ⁽¹⁾

 Madrazo I, Flisser A: Cysticercosis, in Apuzzo MLJ (ed): Brain Surgery: Complication Avoidance and Management. New York, Churchill Livingstone, Inc., 1993, pp 1419-1430.

This case report makes several points that can be emphasized. First, most centers in the United States that treat immigrants from endemic zones would most likely recognize the diagnosis of cysticercosis from the computed tomographic scan showing punctate parenchymal calcifications. However, it would not be generally recognized by the average practitioner in the United States, neurologically oriented or not, that a stroke might be the result of cysticercosis. This report admirably documents such a case, and the images also show the extent of cysticercosis in the subarachnoid space around the sella. There is nothing specific in this appearance on the magnetic resonance imaging scans, computed tomographic scans, or angiograms, which could be attributable to other lesions, e.g., tuberculosis, sarcoidosis, or other inflammatory problems. One of the beauties of magnetic resonance scanning is that it not only lays out the symptom-causing abnormality, such as the deep and superficial brain enhancements secondary to the vascular occlusion, but it also goes more deeply than that, when it is appropriate, to outline the character and extension of meningeal or subarachnoid space disease. This is a major contribution, as is the better analysis of other meningoencephalitides.

It is easy to see how this patient could have developed endarteritis, which we know can be caused by other granulomatous processes, and it is just as easy to see from the images how something such as an orbital apex syndrome, for instance, could be due to this disease process.

There are a couple of interesting features, one of which is that this patient began receiving steroids concomitant with the institution of specific therapy, and, despite the steroids, worsened somewhat. However, it might be that the steroids protected against even more worsening, by virtue of the fact that the left internal carotid artery looks a great deal worse on the angiogram performed at the start of therapy than it did on the magnetic resonance imaging scan performed some months later.

A final point is the importance of simultaneously administering steroids as definitive therapy is started and especially making the distinction between parenchymal lesions and subarachnoid space lesions. The authors' reasoning is lucid and, I think, contributes to the reader's information bank. Whether others are correct in stating that parenchymal neurocysticercosis should not be treated until symptoms appear is not resolved, but that is another story.

David O. Davis

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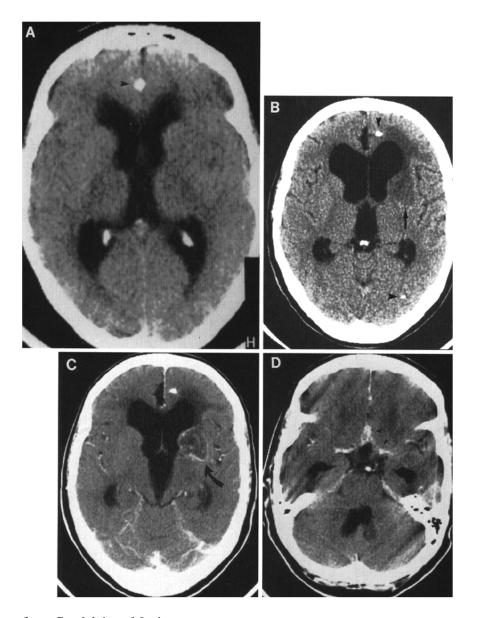


Figure 1. *A*, CT scan from Guadalajara, Mexico, demonstrates moderate hydrocephalus and calcification (*arrowhead*). *B*, CT scan at the time of admission to our facility, performed 6 weeks after the CT scan from Mexico, demonstrates progression of hydrocephalus and a new region of hypodensity involving the caudate, internal capsule, and lentiform nuclei (*straight arrow*). Also noted are scattered parenchymal calcifications (*arrowheads*). *C*, intravenous administration of contrast medium reveals enhancement around the hypodensity in the left basal ganglia (*curved arrow*), consistent with subacute infarction. *D*, diffuse enhancement of the subarachnoid space surrounding the left ICA and both ACAs is also demonstrated (*small arrowheads*).

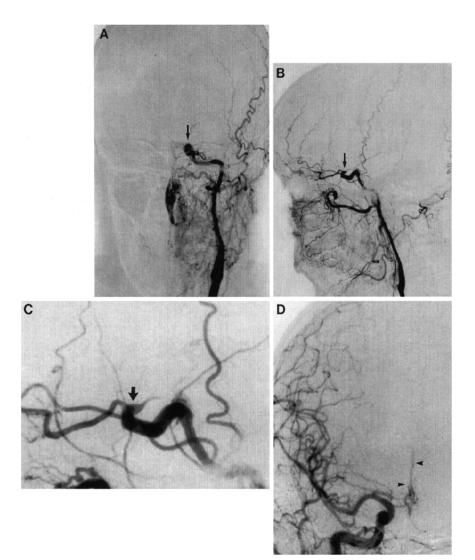


Figure 2. Anteroposterior (A) and lateral (B and C) radiographic views of the left carotid arteriogram demonstrating complete occlusion of the ICA at the level of the anterior choroidal artery (*arrows*) and a normal bulb. Anteroposterior projection of the right carotid arteriogram (D) reveals bilateral occlusion of the A2 segments of the ACAs (*arrowheads*) and stenosis of the right A1 segment.

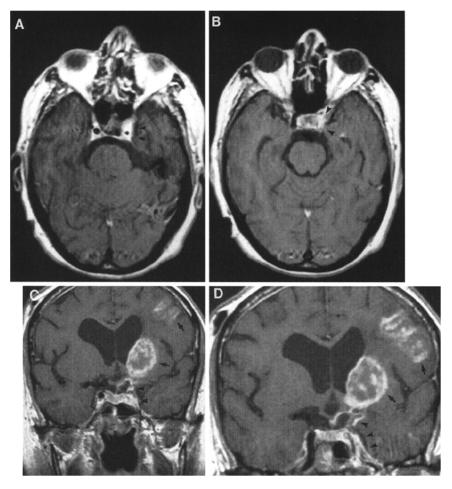


Figure 3. Axial (*A* and *B*) and coronal (*C* and *D*) MRI scans with gadolinium enhancement demonstrate diffuse enhancing subarachnoid material (*arrowheads*) surrounding the left ICA from its entry into the subarachnoid space to its bifurcation. Narrowing of the ICA flow void is seen proximally (*A*), with complete absence of the expected ICA flow void more distally (*B-D*). Note also the enhancing MCA distribution infarctions (*straight arrows*).

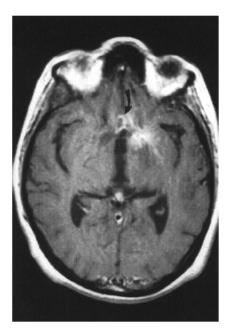


Figure 4. Axial gadolinium enhanced MRI scan reveals densely ring-enhancing structure in the anterior interhemispheric fissure (*curved arrow*) in the region of the occluded A2 segments of the ACAs.

Series (Ref. no.)	Vessel	Presentation	Outcome	Findings
McCormick et al., 1983 (11)	ICA	Left hemiparesis, headache	Hemiparesis markedly improved	Craniotomy revealed a cluster of cysticerci surrounding the ICA
Torrealba et al., 1984 (17)	ICA	Right hemiparesis, aphasia	Died	Autopsy confirmed NCC meningitis and endarteritis
Rodriguez-Carbajal et al., 1989 (13)	МСА	Right hemiparesis, aphasia, headache, emesis	Died	Autopsy revealed a cysticercus in the sylvian fissure with occlusive endar- teritis involving the MCA branches
	MCA	Left hemiparesis, headache, emesis	Hemiparesis unchanged	Open biopsy demonstrated a cysticercus in the Sylvian fissure
	МСА	Left hemiparesis, headache, seizure	Died	Autopsy revealed a basilar subarachnoid exudate with occlusive endarteritis in- volving the MCA
terPenning et al., 1992 (16)	MCA	Right hemiparesis, aphasia, seizure	Hemiparesis unchanged	Bilateral MCA occlusion. MRI revealed a cyst near the occluded MCA
Current report	ICA and ACA	Right hemiparesis, aphasia	Hemiparesis and aphasia markedly improved	MRI revealed enhancing material sur- rounding the subarachnoid ICA and an enhancing lesion in the interhemi- spheric fissure adjacent to the occluded ACAs

Table 1. Summary of Major Intracranial Artery Occlusions Secondary to Neurocysticercosis^{*a*}