CYSTICERCOSIS CEREBRI IN THE PHILIPPINES

A Report of Two Cases

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A review of Philippine literature on cases of cysticercosis in general showed that in 1927, Africa and Sta. Cruz (1) reported two cases among Filipinos. Up to that time only three cases had been reported: the first case, a living subject, was reported by Guerrero (1) and the two others, both autopsy cases from more than 12,000 autopsies performed by the Department of Pathology and Bacteriology of the College of Medicine of the University of the Philippines from 1907 to 1926, were reported by Hammack (1912) and Sta. Cruz (1926) respectively (1).

The first case reported by Africa and Sta. Cruz (1) had cysticercosis of the brain, the hypophysis and the skeletal muscles; this patient had died of acute cardiac dilatation. The other had had generalized muscle cysticercosis; involvement of the central nervous system could not be determined since the family of the deceased had not granted permission for the brain to be opened.

Philippine medical literature from 1927 to the present has been silent on any further case reports of cysticercosis among Filipinos. Whether this is due to a real absence of the infection, or more probably, due to a lack of awareness of its possible existence, remains a matter of conjecture.

This paper presents two cases of cerebral cysticercosis among Filipinos.

Case 1: A.R., 18 years old, female, single, born in Tacloban, Leyte and a resident of Manila for the past two years, was admitted to the Philippine General Hospital on July 11, 1959 for loss of consciousness. She had had 2 similar episodes in the past, once two years before admission and again three months before this admission. The seizures were characterized by upward turning of the eyeballs and tonic-clonic convulsions, with fits of violence, crying and shouting; each attack usually lasted from 4 to 5 hours.

Less than an hour before admission she was found lying flat in the bathroom in tonic convulsions, with blood-tinged, frothy salivation. On admission she was unconscious with a pulse rate of 60/min., a respiratory rate of 24/min., and a blood pressure of 70/40. The eyeballs were fixed downward and to the right. The pupils were 2 mm. wide, sluggishly reactive to light. There was blood in the nose and mouth. The neck was not rigid. Heart and lungs were essentially normal. The abdominal reflexes were absent. The upper extremities were alternatingly flaccid and spastic, the lower extremities were tonically rigid with spontaneous Babinski on the left and positive on the right on stimulation. She was then given Sodium Luminal, grain II. Two hours after admission she was semiconscious and responded to pain stimulus. Ocular right disc could be visualized. The extremities were constantly moving except for the right upper limb The skull X-ray was interpreted as negative.

Two hours later her temperature was 38.7 C., pulse rate 120/min., respiratory rate 32/min., and blood pressure 100/76. Other findings were slight pallor, positive corneal reflexes, absence of blinking when an object is suddenly brought near the eyes and negative Babinski.

Urine was reddish yellow, turbid, had four plus albumin and one plus sugar. There were 500 red cells per high power field, 5 to 10 pus cells per high power field. Blood Kahn, Wassermann and VDRL were all negative.

On the second hospital day she was noted to be restless and with right hemiparesis. Her temperature was 38.5 C. until the next day, when she became cyanotic and had cold clammy perspiration. The pulse became imperceptible and the blood pressure was zero. The heart rate was very rapid. The right pupil was 6 mm., the left 5 mm., both non-reactive to light. Ten minutes later the patient was pronounced dead.

At autopsy the brain weighed 1291 grams and showed flattening of the gyri and shallow sulci, all evidences of brain edema. There was also slight herniation of the cerebellar tonsils into the foramen magnum. Multiple cysts averaging 0.5 cm. in diameter were found all over the brain: there was one in the right inferior frontal gyrus, another in the superior frontal gyrus, and smaller ones distributed all over the left cerebrum. The cysts appeared to be distributed without any particular relation to blood vessels. On opening one of the cysts, it was found to be well circumscribed from the surrounding brain tissue and contained a node. Subsequent histologic examination confirmed the diagnosis of cysticercosis cerebri.

In addition, there were found multiple cysts in the lungs, averaging 1 cm. in diameter. These turned out to be those of *Paragonimus westermani.*

Case 2: F.T., 60 years old, male, married, born in Tuguegarao, Cagayan and a resident of Tondo, Manila, was admitted to the Philippine General Hospital on December 29, 1960 for loss of consciousness. In the past he had complained of frontal, occipital and hemicranial headaches. A month previous to admission he had an attack of unconsciousness, and involuntary movements of the extremities were then observed. He was then hospitalized at the North General Hospital where his systolic blood pressure was found to be 200 mm.Hg. The diagnosis given then was: Cerebral thrombosis secondary to arteriosclerosis; essential hypertension; senile dementia; urinary tract infection. After discharge he had several other episodes of involuntary movements of the head and the

extremities. Three days prior to admission there was a similar recurrence, and a few hours before admission he could no longer talk. Thereafter, he lost consciousness.

Pulse rate on admission was 140/min., respiratory rate 36/min., and blood pressure 140/110 mm.Hg. There were involuntary clonic movements of the right upper extremity every one to two seconds. The head which was turned to the left was in clonic motion toward the same side. There were no cranial deformities or lesions. The left eye was half-closed, the right open; both were unblinking. The corneal reflexes were absent. Pupils were equal (3 mm. each) and reactive to light. The neck was slightly rigid. There were cutaneous ulcerations at the trochlear and coccygeal regions. There were 2 cm. firm subcutaneous nodules on the upper extremities; these nodules were free from the overlying skin but attached to the underlying tissues. The deep tendon reflexes were absent. Babinski and its modifications were negative.

Routine laboratory examinations were essentially negative, except urinalysis which showed plenty of red cells, some white cells, and occasional casts. Funduscopy showed hypertensive arteriosclerotic grade I fundi. A spinal tap done on the third day in the hospital yielded 12 cc. of clear colorless fluid containing 90 mg. sugar, 34 mg. total proteins and 6 cells/cu.mm.; the initial pressure was 70 mm. water.

The clonic movements of the upper extremities at the wrist and elbow joints persisted. On the 5th hospital day he was found to be deeply comatose, had poor muscle tone and evidences of vasomotor collapse. Low-grade fever was present from the 2nd day to the 4th hospital day when it rose to 38.5 C.; patient was afebrile for the next three days. The blood pressure subsequently became erratic, and was maintained between 120/80 and 140/100 mm.Hg. with intravenous fluids and Levophed. The pulse rate ranged from 98 to 108/min., the respiratory rate was 32/min. Subsequent course was progres-

sively downhill with laboratory findings suggestive of a terminal uremia.

Autopsy revealed generalized and widely disseminated cysticercosis cellulosae. Typical cystic lesions ranging from 2-5 mm. in diameter and containing a white, easily detachable parasite in the cystic cavity were found disseminated throughout all visceral organs except the spleen, liver, kidneys, adrenals, and the wall of the gastrointestinal tract. Cysts were also abundant in the skeletal muscles, diaphragm and the tongue, and also in the eyeballs. In addition. one of the cysts in the lung (right lower lobe) had been secondarily infected by bacteria with a resulting abscess. The brain was studded with cysts, histologically confirmed to be those of *T. solium.*

DISCUSSION

History: It was Panarolus (11) in 1650 who first clearly described the occurrence of cerebral cysticercosis in man. However, it was only many years later when the parasitic nature of such cysts was recognized. Excellent summaries have been published by Dizon and Smithers (11) and by Stephen and Chorobski (18).

Incidence: Since 1957 five cases of cysticercosis cerebri have been reported in the United States: three by White and co-workers (20), one by Dent (9) and the latest by Haining (13). An increase in its incidence has been noted in Poland (18) and this has been attributed to poor wartime hygiene. Similarly Bickerstaff and co-workers (5) also reported an increasing frequency of the disease in England and they blame the large number of persons returning from the East as the factor responsible for the apparent increase. The reports of Obrador (15), Arana Iniquez and Asenjo (2), Obrador and Ley (16), and Trelles (19); show that cysticercosis cerebri is prevalent in Mexico, Chile and Spain and Peru, respectively. Although it is said that the occurrence of this disease is greater in certain countries of the Far East (because of the greater incidence of intestinal infection with *T.*

solium in man in these areas), and although Yen-Fang and co-workers (14) detected cysticercosis cellulosae in 30.8% of 26 postmortem cases of Japanese B encephalitis (and they believe that preexistence of cerebral cysticercosis predisposes to Japanese B encephalitis, with an increase in mortality), here in the Philippines the incidence has not been high.

Pathogenesis: Cysticercosis cerebri is caused by the larva of the cestode *T. solium.* Man may become infected through any one of these three ways: (1) Ingestion of feces from a person who harbors an adult tapeworm (heteroinfection); (2) Ingestion of feces containing eggs that are transferred from anus to mouth by unclean hands of an infected person (external autoinfection); (3) by reverse peristalsis of the small intestines by means of which eggs are carried into the stomach where they are conditioned for hatching (internal autoinfection). The larvae enter the circulation and lymphatics and are therefore widely disseminated throughout the body; in fact, they may be found in practically every organ and tissue of the body (12), particularly the skeletal muscles, the chambers of the eye and the central nervous system. It is said that the most frequent site of encystment is the brain (13, 20). In the central nervous system, the cysts may localize within the leptomeninges or brain substance; localization is soon followed by encapsulation and calcification. As long as the larvae remain alive, only minimal local inflammatory reaction is provoked. The death of the larvae produces an intense local granulomatous inflammation of the leptomeninges with foreign-body giant-cell reaction and fibrosis (9). Trelles (19) has a detailed description of the pathological gross and microscopic lesions caused by this parasite.

Classification of Cases According to the Clinical Picture: According to Stephen and Chorodbski (18), localization of the cysts in the brain may give rise to manifestations which can be classified into any one of these three groups:

Group 1: The symptoms and signs in this group are suggestive of a space-occupying lesion, especially an

expanding intracranial tumor. Of the 94 surgically treated patients (23 cases of Stephen and Chorobski and 71 review cases) 64% belonged to this group. The most common sites involved were the cerebral hemispheres (60.3%). This is the type most amenable to surgery.

Group 2: In this group there is widespread infection of the brain without localizing signs. The principal findings are those of swelling, increased intracranial pressure, papilledema, pleocytosis and elevated protein values in the cerebrospinal fluid, and mental changes. The prognosis is very poor, and operation can offer only temporary palliation through decompression (24).

Group 3: The cysts are located mainly at the base of the brain where they provoke a proliferative inflammatory reaction in the basilar meninges. Thus it is not surprising that internal hydrocephalus, nausea and vomiting, and occasionally, paresis of the cranial nerves develop. White and co-workers (20) state that in this group the opening of the anterior end of the third ventricle (lamina terminalis) by the method of Stookey and Scraff to drain the ventricular system may prove to be beneficial provided the cisterna chissmatica has not been involved.

Trelles (13) believes that intracranial hypertension, epileptic attacks and mental disorders make up the fundamental symptomatology of cerebral cysticercosis. Usually neither sudden nor progressively increasing, the characteristic intracranial hypertension is intermittent for long intervals and they may be severe. The epileptic seizures are usually of late onset and focal at the beginning. The interval between seizures may be as long as 15 years. Myoclonia and sensory auras have been frequently observed. The racemose type frequently presents as a mental disorder, e.g. amnesia, disorientation, senile dementia.

Diagnosis: Because cysticercosis cerebri produces symptoms and signs very suggestive of intracranial tumors, it is often

missed preoperatively. It should always be considered in patients living in or coming from areas where the disease is prelent. A history of passing out tapeworm segments in the stools also suggest the diagnosis. Eosinophilia, in the blood and in the cerebrospinal fluid, may be present; unfortunately, however, its presence is exceptional. The cerebrospinal fluid may also show lymphocytosis and an elevated protein content, but these merely suggest the possibility of the disease and are not pathognomonic; furthermore they are more absent than present. Another diagnostic aid is the complement-fixation test, using an alcoholic extract of *T. solium* cysticerci (20); however, a negative result does not exclude the diagnosis, and positive reactions are not conclusive because the specificity of the antigen is not absolute. According to Tolosa, this test is correct in only 60% of cases.

Two diagnostic aids of great assistance are: (1) X-ray films, and (2) biopsy of subcutaneous cysts. Since both the scolex as well as the cyst wall calcify, X-ray films may show multiple small areas of calcification; these were present in a third of Obrador's patient and in 11% of those of Dixon and Hargreaves (10). Other features of the skull x-ray film which may suggest cysticercosis cerebri are: (1) dilatation of the aqueduct of Sylvius (up to 1 cm.) with absence of deviation; (2) only partial obliteration of the aqueduct or the fourth ventricle; (3) good passage of air to the fourth ventricle; and (4) presence of air around the cerebellum, which is a consequence of its atrophy (20). Biopsy of the subcutaneous cysts is the definitive way of establishing the diagnosis of cysticercosis cellulosae. Care must be taken in examining the cysts histologically since macroscopically it is impossible to distinguish between a cysticercus (caused by *T. solium*) and a coenurus caused by *Multiceps*). The distinction lies in that whereas each cysticercus cyst has only one invaginated scolex, each coenurus contains many scolices which project from the wall into the cyst cavity. Furthermore, intracranial coenuri are usually larger (3 cm. or more in diameter) (3, 4).

Although cysticercosis cerebri may be mistaken for Alzheimer's disease or Pick's disease when there are only epileptic seizures and mental deterioration associated with a slight rise in cerebrospinal fluid protein, and although basilar

adhesions with paralysis of cranial nerves may suggest meningitis, syphilitic or tuberculous, nevertheless the condition for which it is often mistaken is brain tumor. Most case reports are of this nature (7, 9, 13, 17, 20); in fact most cases are diagnosed to be posterior fossa tumors because of the prominence of cerebellar symptoms and signs. It must also be remembered that cerebral cysticercosis may present itself as a psychiatric case (8, 19).

The two cases here presented, however, most closely resemble not a brain tumor but an apoplexy. Intracranial hemorrhage, probably secondary to a ruptured angioma, was the primary consideration in the first case, and the second case was thought to be either a subdural hemorrhage or a glioblastoma multiforme which bled intracranially. In neither case was cysticercosis cerebri thought of. These two case reports therefore should remind one to consider the possibility of this disease entity not only in cases of brain tumors but also in those cases that present as apoplectiform attacks, especially where there is a history of similar episodes in the past.

It is interesting to note certain features presented by the two cases, some features being common to both cases. Both patients were admitted for sudden loss of consciousness. Both had similar episodes in the past: the first case, an 18-year old female, had 2 previous episodes, two years and then again months before her latest; the second case, a sixty-year old male, had his first attack one month before his latest. Both had in the present admission as well as in the past episodes, convulsive seizures. It is striking that both had hematuria, gross and/or microscopic. Only one case (Case 2) gave a history of headache. Only one case (Case 2) had subcutaneous nodules, which turned out to be cysticercus cysts in the muscle. The diagnosis of cysticercosis cerebri was proven histologically in both cases; in one of them it was generalized, and in the other there was an associated paragonimiasis in the lungs (Case 1).

Treatment: To date, the only form of treatment that offers hope is surgery. Male fern (oleoresin of Aspidium), the sulfas, cortisone and other adrenocorticoids have not offered satisfactory results (19). Stepien and Chorobski (18) considered

over-all results good in 50% of 94 surgically treated patients, with a recovery or improvement rate of 72.6% in Group 1 patients. In Groups 2 and 3, the benefit from surgery was brief and palliative.

SUMMARY

The current literature on cysticercosis cerebri is reviewed and two cases among Filipinos are presented.

Unlike most of the reported cases which manifested themselves like progressively growing intracranial tumors, the two cases here presented themselves as apoplectiform attacks. Both patients were admitted for sudden loss of consciousness; both gave a history of having had similar episodes in the past; both had, in the present admission as well as in the past attacks, convulsive seizures. Both had plenty of red cells in the urine. One case (Case 2) gave a history of headache; the same patient had subcutaneous nodules which turned out to be cysticercus cysts in the skeletal muscles. Cysticercosis cerebri was proven histologically in both; in one of them cysticercosis was generalized (Case 2), and in the other there was an associated paragonimiasis in the lungs (Case 1).

It is suggested that cysticercosis cerebri be seriously considered in the differential diagnosis of neurological entities that present themselves as apoplectiform attacks especially where there is a history of previous similar episodes or of epileptic seizures, or where subcutaneous nodules which are free from the overlying skin but are attached to the underlying tissues, e.g. they move with muscular contraction, are present.

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